A MULTI-INFORMANT, MULTI-METHOD EXPLORATION OF RESILIENCE AND PAIN ADAPTATION IN YOUTH WITH JUVENILE IDIOPATHIC ARTHRITIS

by

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Dalhousie University is located in Mi'kma'ki,
the ancestral and unceded territory of the Mi'kmaq.
We are all Treaty people.

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ABSTRACT

The application of resilience theory to the context of secondary chronic pain conditions such as juvenile idiopathic arthritis (JIA) is a burgeoning research area. To better understand how to promote pain adaptation in this population, this dissertation aimed to identify the psychosocial correlates and prognostic factors associated with JIA pain (Study 1), examine the role of youth and parent perfectionism in contributing to the youth's psychological health (Study 2), and explore the relative importance (RI) of resilience resources and mechanisms contributing to the pain adaptation of youth with JIA (Study 3). Upon systematically reviewing the literature (Study 1), 516 unique associations between psychosocial factors and JIA pain intensity, frequency, and sensitivity were identified. Greater pain was correlated with unhelpful pain beliefs, lower parent/child self-efficacy, reduced social functioning, parent/child internalizing symptoms, and lower child well-being and health-related quality of life (HRQoL). Prognostically, greater pain was predicted by pain beliefs, internalizing symptoms, and lower well-being. Studies 2 and 3 used data collected from 156 youth with JIA (13-18 years) and a parent through an online survey. Structural equation models in Study 2 demonstrated support for select preregistered hypotheses, wherein perfectionism largely served as a risk factor. Positive relationships between dimensions of youth perfectionism and internalizing symptoms were partially explained through pain catastrophizing (self-oriented perfectionism) and self-concealment of symptoms (socially prescribed perfectionism). Parent self-oriented perfectionism was associated with greater catastrophizing and pain-related fears, and fewer youth depression symptoms; although, no indirect effects were observed. In Study 3, the RI of evidence-based youth and parent resilience resources and mechanisms were simultaneously explored in predicting distinct recovery, sustainability, and growth outcomes (i.e., pain intensity, functional disability, HRQoL, benefit finding). Within-person resources and mechanisms were positively correlated. The RI of predictors varied across outcomes; however, child pain acceptance, followed by youth/parent self-efficacy and youth optimism were the most robust predictors of pain adaptation across models. Taken together, these studies identify the resources and mechanisms that are key to promoting pain adaptation in the context of JIA which has important theoretical and clinical implications for helping youth with JIA to optimize living in the face of adversity.

LIST OF ABBREVIATIONS AND SYMBOLS USED

ACT	Acceptance and Commitment Therapy
AFQ-Y8	Avoidance and Fusion Questionnaire for Youth
ANCOVA	Analysis of Covariance
ANOVA	Analysis of Variance
BDI	Beck Depression Inventory
BFSC	Benefit Finding Scale for Children
BRS	Brief Resilience Scale
BTPS	Big Three Perfectionism Scale
C	Child
CAD	Canadian Dollar
CAPS	Child and Adolescent Perfectionism Scale
CAPTCHA	Completely Automated Public Turing test to tell Computers and Humans Apart
CASE	Children's Arthritis Self-Efficacy Scale
CBCL	Child Behavior Checklist
CBS	Caregiver Burden Scale
CBT	Cognitive Behavioral Therapy
CDI	Children's Depression Inventory
CES-DC	Centre for Epidemiological Studies Depression Scale for Children
CFI	Robust Comparative Fit Index
CGAS	Clinically Derived Global Score for Psychosocial Functioning
CHAQ	Childhood Health Assessment Questionnaire
CHQ	Child Health Questionnaire
CI	Confidence Interval
CINAHL	Cumulative Index to Nursing and Allied Health Literature
Corr	Correlation
CPAQ-A8	Chronic Pain Acceptance Questionnaire for Adolescents
CPT	Cold Pressor Task
CSQ-C	Coping Strategies Questionnaire for Children
CVS	Child Vulnerability Scale
DES-IV	Differential Emotions Scale - IV
E	Enthesitis-Related Arthritis
f^2	Cohen's effect size
FAS	Facial Affective Scale
FDI	Functional Disability Inventory
FES	Family Environment Scale
FOPQC-SF	The Fear of Pain Questionnaire Child – Short Form
FPS	Faces Pain Scale
FPS-R	Faces Pain Scale - Revised

FSC-Q	Health-Related Felt Stigma and Concealment Questionnaire
GCPS	Graded Chronic Pain Scale
GEE	Generalized Estimating Equations
HADS	Hospital Anxiety and Depression Scale
HAQ	Child Health Assessment Questionnaire
HCP	Healthcare provider
HR	Hierarchical Regression
HRQoL	Health Related Quality of Life
IBD	Inflammatory Bowel Disease
IP	Internet Protocol
IQR	Interquartile Range
JAMAR	Juvenile Arthritis Multidimensional Assessment Report
JAQQ	Juvenile Arthritis Quality of Life Questionnaire
JBI	Joanna Briggs Institute
JIA	Juvenile Idiopathic Arthritis
κ	Cohen's Kappa
LiR	Linear Regression
LMM	Linear Mixed Models
LoR	Logistic Regression
LOT-R	Life Orientation Test - Revised
M	Mean
Mdn	Median
MFQ	Mood and Feelings Questionnaire
MLM	Multilevel Models
MR	Multiple Regression
N	No
N	Population Sample Size
n	Sub-sample Size
N/A	Not Applicable
NRS-11	Eleven-point Numeric Rating Scale
O	Oligoarticular Arthritis
OOP	Other-Oriented Perfectionism
OSF	Open Science Framework
P	Parents/Caregivers
p	P-value for Significance Testing
PANAS-C	Positive and Negative Affect Scale for Children
PASE	Parent Arthritis Self-Efficacy Scale
PCQ	Pain Coping Questionnaire
PCS-C	Pain Catastrophizing Scale for Children
PCS-P	Pain Catastrophizing Scale for Parents

PedsQL	Pediatric Quality of Life Inventory
PF	Pain Frequency
PFOPQ	Parent Fear of Pain Questionnaire
PHQ-4	Patient Health Questionnaire - 4
PI	Pain Intensity
PIS	Pain Intensity Scale
Po	Polyarticular Arthritis
PPAQ	Parent Pain Acceptance Questionnaire
PPFQ-10	Parental Psychological Flexibility Questionnaire - 10
PPQ	Pediatric Pain Questionnaire
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-analysis
PROMIS	Patient-Reported Outcomes Measurement Information System
PROSPERO	International Prospective Register of Systematic Reviews
Ps	Psoriatic Arthritis
PS	Pain Sensitivity
PSC	Pediatric Symptom Checklist
PSS-10	Perceived Stress Scale - 10
QoL	Quality of Life
QoML	Quality of My Life Scale
QST	Quantitative Sensory Testing
R	Statistical Software
r	Standardized correlation
R^2	Proportion of variance explained in regression
RCADS	Revised Child Anxiety and Depression Scale
RCADS-25	Revised Child Anxiety and Depression Scale Short Version (25 items)
RCMAS	Revised Children's Manifest Anxiety Scale
Reg	Regression
RI	Relative Importance
RMSEA	Root Mean Square Error of Approximation
RPI	Recalled Pain Inventory
S	Systemic Arthritis
SCARED	Screen for Child Anxiety Related Disorders
SCCAMPI	The Stress and Coping Cyclical Amplification Model of Perfectionism in Illness
SD	Standard Deviation
SEM	Structural Equation Models
SOP	Self-Oriented Perfectionism
SOPA	Survey of Pain Attitudes
SPP	Socially Prescribed Perfectionism
SPPA	Self-Perception Profile for Adolescents

SPPC	Self-Perception Profile for Children
SPQ	Structured Pain Questionnaire
SRMR	Standardized Root Mean Square Residual
SRQ-20	Self-Reporting Questionnaire
SSQR	Social Support Questionnaire – Revised
SSRS	Social Skills Rating System
STAI-C	State-Trait Anxiety Inventory for Children
SUPER-KIDZ	Standardized Universal Pain Evaluation for Rheumatology Providers for Children and Youth
TLI	Robust Tucker-Lewis Index
TSC-C	Trauma Symptom Checklist for Children
TTC	Teens Taking Charge Self-Management Intervention
U	Undifferentiated/Other Arthritis or Unclear
VAS	Visual Analogue Scale
Y	Yes
YLOT	Youth Life Orientation Test
WHO-5	World Health Organization Well-Being Index
WHYMPI	West Haven-Yale Multidimensional Pain Inventory
$\bar{x}$	Mean
α	Alpha
β	Standardized regression coefficient
Δ	Change

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CHAPTER 1: INTRODUCTION

1.1. Rheumatic Diseases in Childhood

Rheumatic diseases involve abnormalities predominantly affecting the musculoskeletal system, inclusive of conditions such as arthritis and fibromyalgia (Petty, Laxer, Lindsley, et al., 2021). Although there is truth to the belief that rheumatic conditions predominantly affect older populations (Statistics Canada, 2022), children can also be affected. The most frequently presenting rheumatic disease in childhood is juvenile idiopathic arthritis (JIA). JIA is a group of chronic inflammatory diseases with an unknown etiology wherein the key diagnostic indicator is synovitis, or inflammation of the synovial membrane of the joint (Rapoff et al., 2017), and the key presenting symptoms are pain and stiffness (Canadian Paediatric Society, 2009). According to the International League of Associations of Rheumatology, JIA is an umbrella term for seven heterogeneous subtypes of the disease: systemic arthritis, persistent and extended oligoarthritis, rheumatoid factor positive polyarthritis, rheumatoid factor negative polyarthritis, psoriatic arthritis, enthesitis-related arthritis, and undifferentiated arthritis (Petty et al., 2004). These diagnoses have different incidence rates, ages of onset, and sex distributions (Thierry et al., 2014) and differ in factors such as the presence of other clinical features (e.g., rashes) and the number of joints affected (Petty et al., 2004). There is nevertheless disagreement about this classification system (Lee et al., 2022) as the similarities across subtypes can make it difficult to distinguish one from the other (e.g., all chronic inflammatory diseases presenting before 16 years of age). Across subtypes, JIA largely affects females (Cattalini et al., 2019), and best available evidence suggests that it affects up to 8 million children worldwide, most of whom are undiagnosed (Petty,

Laxer, & Wedderburn, 2021). Nationally, that equates to approximately 1 in every 1000 Canadian children (Public Health Agency of Canada, 2020).

Merely a generation ago, children with JIA often experienced complications such as joint damage, deformity, uveitis (i.e., inflammation in the uvea, or the middle layer of the eye), and growth abnormalities (Stoll & Cron, 2014). Fortunately, the past few decades have brought revolutionary changes in the management of JIA pharmacologically (e.g., advances in biologic therapies; Stoll & Cron, 2014), physiologically (e.g., encouragement of physical activity; West et al., 2019), and psychologically (e.g., provision of psychosocial supports; Cohen et al., 2017).

These advances have successfully reduced the inflammation and disease activity in many children (Cavallo et al., 2017; Ehrmann-Feldman et al., 2007). For example, in a Canadian cohort of over 1100 children with JIA (Guzman et al., 2015), the probability of attaining inactive disease (defined as an active joint count of 0; a physician global assessment of the disease as <10mm on a 0-100mm scale; and the absense of systemic manifestations, enthesitis, and uveitis) within 2 years of diagnosis was over 70%. Within 5 years of diagnosis, the probability of attaining remission (defined as at least 12 months of inactive disease) was approximately 50%. Thus, although JIA is a chronic condition, it is also a manageable condition. With the proper supports and treatments, many children with JIA can lead a life without significant joint deformity or damage. Unfortunately, despite these extraordinary advances over the past few decades, a key symptom remains absent from the definitions of disease inactivity and remission – the experience of pain (Giancane et al., 2017).

1.2. Pain in Juvenile Idiopathic Arthritis

Pain is one of the most frequently reported symptoms amongst youth with JIA (Canadian Paediatric Society, 2009). It is a subjective experience defined as "an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage" (Raja et al., 2020). While JIA pain can be procedural, originating from a specific treatment regimen such as an infusion or self-injection (Brandelli et al., 2019a), the most frequent experience of pain is that which does not have a specific origin and is chronic in nature (Weiss et al., 2014). Chronic pain is defined as "pain that lasts or recurs for longer than three months" (Treede et al., 2019). It is considered to be a multidimensional experience that is associated with a combination of biological (e.g., genetics, disease activity, abnormal pain processing, tissue damage, medications), psychological (e.g., anxiety, depression, coping, sleep), social (e.g., limited social interactions), and environmental (e.g., family pain history, other's responses to one's pain) factors (Stinson & Prescott, 2021).

From a neurophysiological perspective, Stinson and Prescott (2021) review how the experience of pain (i.e., nociception) begins with the activation of sensory neurons (i.e., afferents) that exist throughout peripheral tissues such as the skin, muscles, and joints. Afferents become activated in response to noxious stimuli (e.g., temperature, pressure, mechanical force, chemicals) and relay sensory information to the central nervous system and amygdala (the ascending pain pathway). In the context of JIA, the afferents may initially become activated in response to synovitis. In the amygdala, the pain is processed alongside one's thoughts, memories, and emotions, which subsequently initiates the physiological response to pain, or the withdrawal from the noxious stimuli if

possible (the descending pain pathway). In the case of chronic pain, the pain pathway can become activated in the absence of a noxious stimuli due to sensitization at different points along the way. Peripheral sensitization can occur when the primary afferents become sensitized, thus leading to hyperalgesia (i.e., exaggeration of the pain response) or allodynia (i.e., pain in response to innocuous stimuli). Central sensitization can also occur when the central nervous system becomes sensitized. Thus JIA pain is truly "more than simple nociception," as Munro and Singh-Grewal (2013) describe in their aptly titled editorial. Abnormal pain processing has been observed in various samples of youth with JIA (Arnstad et al., 2020; Cornelissen et al., 2014; Hogeweg et al., 1995; Leegaard et al., 2013; Thastum et al., 2001; Thastum et al., 1997). As an example, Cornelissen et al. (2014) used an experimental pain paradigm, quantitative sensory testing, to assess the pain sensitivity of 60 children with JIA. They found a generalized hypersensitivity amongst children with JIA (with and without active disease) compared to healthy controls in response to pressure, light touch, cold, and heat pain. Thus, although pain may have its origins in part in the biology of JIA, it also has the potential to develop into its own issue.

The pain associated with JIA is a multidimensional experience that can be understood in terms of its sensory (e.g., intensity, duration, frequency, location, descriptor words), affective (e.g., unpleasantness, emotional impact), and evaluative (e.g., perception of interference with functioning) components (Melzack, 1975; Stinson & Prescott, 2021). In terms of its sensory components, the literature has demonstrated that JIA pain varies within and between days (Tupper et al., 2013). One daily diary study found that patients reported pain on average 73% of days across a 2-month period (Schanberg et al., 2003). While most children reported mild to moderate pain

experiences, approximately 1/3 reported severe pain. Shiff et al. (2018) studied pain longitudinally in 1,062 children enrolled within 90 days of their diagnosis of JIA for up to five years. Five trajectories were identified: 56% of children had a mild-decreasing pain trajectory, 29% of children had a moderate-decreasing pain trajectory, 7% had a chronically-moderate pain trajectory, 4% had a minimal pain trajectory (i.e., a consistently low pain intensity), and 4% had a mild-increasing pain trajectory. For many, JIA pain continues to persist despite treatment advances and improvements in disease activity (Bromberg et al., 2014; Butbul Aviel et al., 2011; Cornelissen et al., 2014), likely in part due to peripheral and central sensitization. Moreover, the experience of JIA pain (Tollisen et al., 2018) and abnormal pain processing (Arnstad et al., 2020) can persist into adulthood.

Despite these experiences, and despite pain management having been declared as a basic human right (Brennan et al., 2019), healthcare providers (HCP; e.g., rheumatologists, nurses, physical therapists) report low confidence in assessing for pain and a general reluctance to engage in conversations regarding JIA pain (R. R. Lee et al., 2020). In contrast, families continue to report a need for increased support around their child's pain management (Brandelli et al., 2019b) to the point that JIA pain has emerged as a top research priority amongst families of children with arthritis (Correll et al., 2020). Given this discrepancy between patient and family needs and the challenges HCP continue to face in assessing and managing JIA pain, there is an important need for research in this area.

1.3. Implications of JIA Pain

By way of the affective and evaluative components of pain, the consequences of

JIA pain are far-reaching (Stinson & Prescott, 2021). There is a plethora of literature demonstrating the detrimental effects JIA pain can have on one's quality of life, mental health, functioning, participation, and sleep quality (Butbul Aviel et al., 2011; Fair et al., 2019; Feinstein et al., 2011; Guite et al., 2011; Stinson et al., 2011), many of which can persist into adulthood (Barth et al., 2016; Packham & Hall, 2002; Tollisen et al., 2018). Furthermore, many of these detrimental effects can impact those in the child's support system as well (Brandelli et al., 2019a; Brandelli, Tutelman, et al., 2022; Vuorimaa et al., 2011; Yuwen et al., 2017). As an example, Yuwen et al. (2017) explored nine parents' experiences caring for a young child with JIA. Through inductive content analyses numerous experiences were identified, two of which included "feeling my child's pain" and "feeling drained by the whole process".

Research has largely focused on understanding JIA pain through a risk perspective, focusing on individual and environmental risk factors for worse and persistent pain and the subsequent consequences. Although it is intuitive to focus on the negative associations of an undesirable experience as intervention targets, exclusively focusing on the negative components of an experience may preclude the advancement of knowledge and the promotion of human strengths as a possible route for intervention. This is in line with the growing evidence for Acceptance and Commitment Therapy (ACT) as an intervention for pediatric chronic pain (Pielech et al., 2017) and health conditions (Chihwen et al., 2013; Masuda et al., 2011), which focuses on strengthening one's engagement in value-directed tasks rather than avoiding aversive experiences (Pielech et al., 2017).

The experience of JIA pain and the presence of risk factors does not necessitate

undesirable outcomes. Individual variation in pain experiences and how one adapts to JIA pain has been observed (Hynes et al., 2019; Stinson et al., 2011), suggesting the presence of pain in JIA is not deterministic. As Tong et al. (2012) found in their qualitative synthesis, some youth with JIA experience a mastery over their body and their pain experiences. One youth described it as being the "controller controlling the body". Similarly, a published abstract by our team used descriptive content analysis to explore parent's perspectives of their child's arthritis pain (Brandelli et al., 2020). One category that was identified was their perspective of their child's strength and resilience. As an example, a mother of a 17-year-old girl with JIA described:

> "My daughter knows now that she has ups and downs. She knows her meds keep her in a good place. She knows when she stops physio she's in trouble. … She knows getting out and doing things makes her feel better and isolation at home is way more depressing… She is very involved in after school activities and spends lots of time with her friends who understand her pain and help her."

These findings suggest there are underlying factors that allow some youth to better manage the adversity of JIA pain, which is important knowledge when it comes to assessing for and managing JIA pain.

1.4. Resilience

The shift in emphasis to a strengths-based approach is not only more aligned with patient interests (Birnie, Dib, et al., 2019), it also offers important insights on how some youth successfully navigate the experience of JIA pain, which in turn, has the potential to inform prevention and intervention efforts (Masten, 2019). Conceptually, this is the study of resilience.

Resilience is a complex and evolving construct with no unifying definition. Masten (2001) refers to resilience as the study of "ordinary magic", as it is not unusual to see success in the face of adversity, yet there is something elusive about the concept. The tenets of what comprises resilience are largely agreed upon (Southwick et al., 2014; Ungar, 2018). Specifically, resilience: 1) is a process rather than a static trait (i.e., one is not "resilient"), and therefore it is something that can be changed; 2) involves exposure to adversity (in this case, JIA pain); 3) engages a capacity to adapt successfully in response to said adversity (i.e., demonstrate stability or recovery in their social, mental, or physical functioning); and 4) emphasizes the broader social-ecological system over the individual (which is particularly relevant for children, whose development is based on interactions between genetic, neurobiological, social, and cultural experiences). The American Psychological Association (2006) defines resilience as, "the process and outcome of successfully adapting to difficult or challenging life experiences, especially through mental, emotional, and behavioral flexibility and adjustment to external and internal demands". What is missing from this definition is the emphasis on the system, thus a more encompassing definition was proposed by Masten (2014), wherein resilience is defined as, "the capacity of a dynamic system to adapt successfully to disturbances that threaten system function, viability, or development".

Resilience has its roots in the study of risk, promotive, and protective factors (i.e., associated with worse outcomes, better outcomes, and a dampening effect on risk factors, respectively; Racine et al., 2022); however, it is more than any of these factors in isolation. No single demographic, personality, biological, or social factor has been found to enhance adaptation by more than a small degree (Southwick et al., 2014). The study of

resilience is thus more complex, and focused on how the variables relate within and between systems to create the heterogeneous responses to adversity that exist (Rutter, 2012).

1.5. Ecological Resilience-Risk Model in Pediatric Chronic Pain

The complexity of this construct is well described by the Ecological Resilience-Risk Model in Pediatric Chronic Pain (Cousins, Kalapurakkel, et al., 2015), the theoretical underpinning for this dissertation. Modelled off of the risk-resilience model in adult chronic pain (Sturgeon & Zautra, 2013) and Bronfenbrenner's (1979) ecological systems theory, this model situates the child within the family and social environment, which is further situated within the culture and time. Within each of these interacting systems, there are resources (i.e., stable traits such as one's personality) and mechanisms (i.e., dynamic, modifiable processes such as one's coping style) that come in to play when one is faced with pain. These resources and mechanisms can be adaptive or maladaptive (i.e., resilience resources and risk factors; resilience mechanisms and risk mechanisms). Together these adaptive and maladaptive resources and mechanisms interact within and between systems to produce the conditions necessary for pain adaptation. Three domains comprise the outcome of pain adaptation in this model (Reich et al., 2010). The first is recovery or resumed functioning, assessed through measures of physical health, pain, psychological health, and quality of life. The second outcome is sustainability, defined as a perseverance of valued activities and measured through constructs such as academic success and valued living. The third outcome is growth, defined as a realization of one's capabilities, which can be assessed through measures of benefit finding or posttraumatic growth (e.g., finding a sense of purpose, deepening

relationships) and self-regulation in response to pain (e.g., modulating thoughts, emotions, and behaviors towards a goal). Together, the complex and multifaceted interaction of resilience and risk resources and mechanisms as situated within the culture and time in contributing to outcomes of pain adaptation represents the process of resilience.

From the broader literature and earlier work exploring risk, promotive, and protective factors, we know of some resources and mechanisms that reflect adaptive systems. These include supportive relationships, problem-solving and self-regulation skills, self-efficacy, optimism, and belief in the meaning of life (Masten, 2019). With that said, very few of these have been explored in relation to JIA pain, let alone in complex analyses with multiple variables involved (e.g., mediations, moderations, path analyses).

A systematic review conducted by Hynes et al. (2019) identified a total of seven studies exploring resilience within the context of JIA. Beeckman, Hughes, et al. (2019) found that child and parent psychological flexibility (i.e., being aware of and open to unwanted and uncontrollable experiences while still having the ability to act in line with broader life values; Hayes et al., 2006) and child pain acceptance were associated with adaptive functioning in youth with JIA. Seid et al. (2014) found that child perceived social support, child self-efficacy, and medication adherence were significantly associated with functioning. Connelly (2005) found a negative association between family dysfunction and child hope. Sawyer (2005; 2004) demonstrated that pain coping strategies were related to functional disability and health related quality of life. Frank et al. (1998) found that parental distress was significantly associated with more swollen joints, thereby hindering child adaptation to JIA. Finally, Timko et al. (1993) identified

that parental distress was associated with greater functional disability in children. Across these studies, the only resilience resource explored was the child's perceived social support, and the only resilience mechanisms assessed were self-efficacy, youth and family psychological flexibility, medication adherence, and pain acceptance. They were all explored in relation to the child's recovery or resumed function, with no studies assessing other components of adaptation such as sustainability or growth. Furthermore, few addressed parent or environmental factors, and a minority explored resilience in the context of JIA pain specifically.

Clearly there is a dearth of literature on resilience in JIA pain overall. More specifically, there is a lack of literature exploring other resources and systems (Hynes et al., 2019). As an example, although there is broader evidence to suggest optimism may serve as a resilience resource (e.g., Cousins, Cohen, et al., 2015; Cousins et al., 2016; Tomlinson et al., 2021), that has not been explored in this population. Moreover, there are gaps pertaining to the role of the broader system (e.g., parent resilience resources and mechanisms), the assessment of outcomes other than quality of life (e.g., benefit finding), and the use of analyses that extend beyond single variable main effects (e.g., path analyses, multiple regressions) (Cousins, Kalapurakkel, et al., 2015; Hynes et al., 2019; McKillop & Banez, 2016).

More specifically, one personality trait (i.e., resource) in particular is beginning to receive increased attention in the pediatric pain literature – perfectionism. This is defined as a multidimensional personality trait that involves striving for flawlessness, setting exceedingly high standards, making overly critical evaluations of oneself and others, feeling pressure to meet high standards imposed by others, and expecting perfection of

others (Frost et al., 1990; Hewitt & Flett, 1991). Perfectionism is a frequently presenting clinical phenomenon that is theorized to amplify challenges in youth with pain and their parents such that it undermines coping and pain adaptation. It is posited to work through biological (i.e., via increased stress and subsequent alterations in pain processing and inflammatory processes), psychological (i.e., via cognitive and behavioral correlates that can precipitate, maintain, or exacerbate pain), and social (i.e., via greater interpersonal challenges) processes (Randall, Gray, et al., 2018). With that said, while perfectionism may serve as a risk factor in some situations, certain dimensions may also have the potential to serve as a resilience resource (e.g., increasing self-management; Piercy et al., 2020), and thus requires further exploration and application to this population (Randall, Gray, et al., 2018).

Taken together, the studies that comprise this dissertation are among the few spearheading the application of resilience theory to the context of JIA pain to address the aforementioned gaps. Building on the multi-informant and systems approach of the Ecological Resilience-Risk Model in Pediatric Chronic Pain, this dissertation seeks to further our understanding of how to foster and support resilience amongst youth with JIA pain and their families.

1.6. Methodological Considerations

In addition to incorporating a series of studies that follows best available evidence as it pertains to patient engagement and open science, this dissertation utilizes a rigorous multi-informant and multi-method approach to inform the understanding of resilience in the context of JIA pain. This dissertation incorporates a series of independent, interrelated research studies using complementary methodologies encompassing data from

youth/parent dyads to ascertain broader implications for an overall research goal (Anguera et al., 2018). An advantage of this approach is that it allows researchers to examine phenomena from different perspectives with different levels and types of data to offer a more comprehensive picture, which is highly suited to further the study of resilience in the context of JIA pain given that pain is a personal, subjective, and multidimensional experience (Raja et al., 2020). These methodologies include the use of a systematic review and online data collection that incorporates the dyad (i.e., youth with JIA and parents) and the application of psychometrically appropriate measurements of pain. Below, an overview of these design considerations and methodologies is provided, including their benefits and the ways in which they have been incorporated.

1.6.1. Patient Engagement

Patient engagement is the meaningful and active collaboration of patients with lived experience and/or their caregivers in conducting research and knowledge translation (Canadian Institutes of Health Research, 2019). It stems from the principles of inclusiveness, support, mutual respect, and co-building in research (Canadian Institutes of Health Research, 2019). As with other team members who bring a unique domain of expertise to the research team, patient and family partners bring expertise in the form of lived experiences with a health condition, or in supporting someone with a health condition.

Patient engagement exists on a spectrum, wherein partners can be involved at any stage of the research process, from planning (e.g., determining the relevance of the research question), to recruitment (e.g., piloting and reviewing study materials, advertising within their networks), to analysis (e.g., sharing their interpretation of study

results, situating the results within the real-world context), to dissemination (e.g., co-presenting results, being involved in the publication of results; Ontario Brain Institute, 2019). Meaningful patient engagement, however, goes beyond this. Black et al. (2018) recommend fostering the partnership early on, and Hamilton et al. (2018) emphasize the importance of explicitly addressing potential challenges related to procedures (e.g., compensation), timelines, team interactions, the research environment, and feeling sufficiently equipped to engage in the study. Training opportunities (e.g., Kids Brain Health Network et al., 2022) and best practice guidelines (e.g., Richards et al., 2020; Richards et al., 2018) exist to support researchers and partners in this collaboration.

The benefits of patient engagement are expansive and impact both members of the collaboration. Involving a patient or family partner encourages researchers to become more accountable and transparent, and to address more relevant research questions (Domecq et al., 2014). Partner involvement is also associated with increased study enrollment and success with grant applications (Domecq et al., 2014). Partners involved in research have reported generally positive experiences, and appreciate the opportunity to co-build research and feel heard in sharing their expertise (Leese et al., 2018). Although there are drawbacks pertaining to logistics (e.g., time, funding; Domecq et al., 2014), the need for institutional support (Domecq et al., 2014), and the potential for burden on partners (Leese et al., 2018), the benefits tend to outweigh the risks (Vat et al., 2019).

The studies of this thesis are based on the research priorities identified by youth and families in the pain and arthritis communities (Birnie, Dib, et al., 2019; Correll et al., 2020). Cassie + Friends, a Canadian non-profit organization advocating for greater

awareness around childhood rheumatic diseases (Cassie + Friends, 2024), has been involved with this dissertation since its inception as a stakeholder. Furthermore, prior to the onset of this dissertation an open call was initiated to recruit a diverse team of youth and family partners who have provided instrumental support throughout all phases of the research process (Brandelli, Jordan, et al., 2022).

1.6.2. Open Science

Open science is defined as the "transparent and accessible knowledge that is shared and developed through collaborative networks" (Vicente-Saez & Martinez-Fuentes, 2018). This includes practices such as making scientific data and materials available (e.g., preregistering hypotheses, making data available to be reproduced) and publishing in open access journals (Foster, n.d.). This scientific movement has been gaining momentum in recent years given the reproducibility crisis in the psychological sciences (Open Science Collaboration, 2015) and the ongoing risk of scientific misconduct (e.g., fabrication, falsification, and questionable research practices like p-hacking, selective reporting, and hypothesizing after results are known; Gross, 2016).

Although there are some drawbacks to practicing open science, including greater time, open access publishing fees, and extra considerations around privacy and confidentiality, the benefits are far reaching. The practice of open science has important implications not only for individual researchers and the broader discipline, it also greatly impacts the community (Foster, n.d.). Benefits to individual researchers include greater collaboration, accountability, and visibility. Benefits to the discipline include greater scientific rigor, more cost-effective research, and a culture of collaboration and open engagement. Most importantly, benefits to the community include increased trust in

science, accelerated knowledge transfer (helping to reduce the 17 year gap that exists in translating health research into practice; Morris et al., 2011), and reduced burden on vulnerable populations to participate in multiple research studies.

The studies that comprise this dissertation make use of various practices that align with open science, including the preregistration of hypotheses and protocols, the availability of data through open science platforms, and the publication of manuscripts in open access journals.

1.6.3. Systematic Review

Data synthesis methods form an important pillar of evidence-based health care (Jordan et al., 2019). One method in particular, the systematic review, aims to provide a comprehensive, unbiased synthesis of many relevant studies in a single document using rigorous and transparent methods (Aromataris & Pearson, 2014) to understand the current state of the evidence, identify conflicting results, and produce statements to guide decision making and areas for future research. Five key tenets of a systematic review include: articulating the objective, inclusion, and exclusion criteria a-priori; comprehensively searching the literature; critically appraising the included studies; analysing and synthesizing the extracted data; and transparently reporting the methods (Aromataris & Pearson, 2014).

A systematic review methodology was utilized in Chapter 2 to inform the known correlations and prognostic relationships between psychosocial risk and resilience factors and pain in youth with JIA. This was necessary because no previous systematic synthesis of this literature has been conducted and because it remains unclear in the literature whether certain psychosocial factors are harmful or helpful (e.g., various coping styles).

Moreover, this information was critical to informing the design and conduct of the subsequent studies of this dissertation.

There are numerous organizations worldwide that guide the conduct of systematic reviews, such as the Cochrane Collaboration (Higgins et al., 2019) and the Joanna Briggs Institute (JBI; Aromataris & Munn, 2020). While both are evidence-based alternatives, these methodologies differ in their emphasis on the types of studies included in their reviews. JBI was selected as the guiding methodology for the conduct of the review included in Chapter 2, given its inclusivity of diverse study designs (e.g., observational designs) which is in line with the current literature in the field of JIA pain.

1.6.4. Online Research

Interest in online research (i.e., online recruitment and data collection) has increased exponentially due to the COVID-19 pandemic and the resulting public health restrictions. These restrictions have trickled down to affect the feasibility of in-person data collection at many health centres and academic institutions (Villarosa et al., 2021). As an example, within the Canadian Alliance of Pediatric Rheumatology Investigators JIA registry, there was a reported decrease of 50% in research registry enrollment, which was directly linked to recommendations to halt research recruitment temporarily (Dushnicky et al., 2022).

The shift to online research is a welcome strategy, conferring advantages to participants and researchers alike. These include, but are not limited to, fewer barriers to participation, reduced research expenses, greater heterogeneity in participants, and automatization of data collection (Hoerger, 2010). In addition to the above-mentioned benefits, several other factors make online data collection a promising avenue for health

researchers specifically. Findings from the Pew Research Center demonstrated that over 85% of adults (Perrin, 2021) and 95% of youth (Vogels et al., 2022) have access to digital devices (e.g., smartphones and computers), many of whom are accessing social media platforms such as Facebook, Instagram, and YouTube multiple times per day (Auxier & Anderson, 2021; Vogels et al., 2022). This makes youth and adults natural foci for advertising research studies over social media (Amon et al., 2014). Moreover, social media use is especially high in individuals seeking health-related information (Hamm et al., 2014; Silver et al., 2019; Tonsaker et al., 2014), including over 70% of youth with JIA (van Pelt et al., 2015). Many youth and families who are faced with rare health conditions such as JIA are keen to advance their self-management skills (Stinson et al., 2020) and broaden their network/receive peer support from others with similar health conditions (Stinson, Feldman, et al., 2012; van Pelt et al., 2015) as evidenced by recent peer support initiatives (e.g., Cassie + Friends, 2022; Stinson et al., 2016). Evidently this intersection of societal changes has led to the online world being an opportune area for the recruitment of youth with JIA and their caregivers, and past research can attest to the success of this method (Brandelli et al., 2019a).

There are nevertheless potential drawbacks to online research in two broad domains necessitating specific considerations in this research methodology. The first is the legitimacy of the population from whom the data is being collected. The pandemic led to a drastic increase in cybercrime (Zhang et al., 2022), and the impact on research is no exception. Sham respondents (i.e., disingenuous and software automated responses, Teitcher et al., 2015) are increasingly capitalizing on online research for financial gain. Recommended strategies to prevent their participation include not publicly sharing the

survey link, minimizing the promotion of incentives, manually screening participants, using security features such as CAPTCHAs (Completely Automated Public Turing test to tell Computers and Humans Apart) and ballot box stuffing prevention, and including attention and logic checks (Griffin et al., 2022; Simone, 2019; Storozuk et al., 2020; Teitcher et al., 2015). Despite best attempts to prevent their participation, it is likely that some may still permeate the abovementioned barriers, making it critical to verify data integrity prior to analyses. Recommended strategies to do so include screening open-ended questions, verifying Internet Protocol (IP) addresses, cross-checking duplicate demographic information, and removing outlier response times (Griffin et al., 2022; Simone, 2019; Storozuk et al., 2020; Teitcher et al., 2015). The second drawback is participant retention. Particularly in the context of the pandemic when screen fatigue can result from the increased online interactions (McClain et al., 2021), there is a greater risk of participant drop-out. Research had shown that approximately 10% of participants can be expected to drop out automatically, with an additional 2% for every 100 questions asked (Hoerger, 2010). As such, it is particularly important to be mindful of the survey length. Other recommended strategies for participant retention include offering incentives, inserting breaks, minimizing open ended questions, and including progress trackers (Afkinich & Blachman-Demner, 2020; Hoerger, 2010).

Taken together, while online research presents as a valuable methodology amidst the pandemic, there are important considerations to ensure data integrity. The latter two studies (Chapters 3 and 4) capitalize on this strategy while attending to the best available evidence around conducting web-based research.

1.6.5. Measurement of Pain in Juvenile Idiopathic Arthritis

Given the subjectivity of the pain experience (Raja et al., 2020), it is challenging to ascertain information from proxy-reporters regarding pain (Stinson & Prescott, 2021) and pain-related functioning (Cohen et al., 2010). There is, in fact, consistent evidence that proxy-reports of pain are highly discrepant and less reliable than child pain reports, particularly when the child's disease activity is high and/or youth are experiencing symptoms of depression (Boerner et al., 2013; Chambers et al., 1998; Doherty et al., 1993; Lal et al., 2011; Palermo, Zebracki, et al., 2004). With that said, pain self-reports should be considered in the context of other factors (e.g., developmental stage, psychological factors, environment, challenges with recall bias), and thus multi-informant perspectives can add great value (Cohen et al., 2010; Palermo & Chambers, 2005). There is also consistent evidence that asking youth to complete pain reports at multiple time points rather than recalling pain over a specified period may yield a more accurate representation of the pain experience (Palermo, Valenzuela, et al., 2004; Stinson, Jibb, et al., 2014), thus multi-timepoint measures are also recommended when possible (Stinson et al., 2011). Finally, as pain is comprised of multiple dimensions, measures should also reflect this (Melzack, 1975; Stinson & Prescott, 2021). Taken together, while self-report assessments of JIA pain are the gold standard, they are elevated in the context of multiple perspectives, dimensions, and timepoints.

Although various pain measures exist (Cohen et al., 2008; Lootens & Rapoff, 2011), few incorporate all the above-mentioned criteria. Moreover, at a component level, there is limited evidence on the quality of existing tools assessing pain intensity in the context of pediatric chronic pain (Birnie, Hundert, et al., 2019). The two biggest contenders are the visual analogue scales (VAS) and the eleven-point numeric rating

scale (NRS-11); however, even these scales demonstrate weak evidence at best. There is one multi-dimensional pain measure that incorporates the NRS-11 among other measures of pain frequency, duration, descriptions, unpleasantness, and limitations - the Standardized Universal Pain Evaluation for Rheumatology Providers for Children and Youth (SUPER-KIDZ; Luca, 2013; Luca et al., 2017). This measure has demonstrated appropriate psychometrics, has been validated in youth with JIA between 4 to 18 years of age, and incorporates both youth and parent reports. Given these features, components of this measure were used in Chapters 3 and 4 of this dissertation. Although it is only used once rather than over repeated timepoints in an effort to acquire a sufficient sample size and bolster retention, multiple perspectives are collected and care was taken to reduce the recall time (i.e., asking about current pain and pain over the past month).

1.7. Overview of Dissertation Studies

To further support adopting a strengths-based approach to the study and management of JIA pain, this dissertation explores various facets of the Ecological-Resilience-Risk Model in Pediatric Chronic Pain and illuminates ways to promote resilience in the context of JIA pain through three studies that incorporate the above-mentioned innovative methodologies. The dissertation aims to: 1) synthesize the current state of our knowledge regarding the known dyadic risk and resilience factors (resources and mechanisms) associated with and predictive of JIA pain intensity, frequency, and sensitivity (recovery outcomes); 2) examine whether perfectionism in parents and/or youth serves as a risk factor or a resilience resource in predicting youth internalizing symptoms (recovery outcomes); and 3) explore the synergy between, and relative importance of, various evidence-based youth and parent resilience resources and

mechanisms in predicting usual pain, functioning, generic- and rheumatology specific health-related quality of life (HRQoL; recovery and sustainability outcomes), and benefit finding (growth outcome). These studies are reviewed in Chapters 2-4 of this thesis. A general discussion of the results, strengths and limitations, and clinical and research implications is outlined in Chapter 5.

The first study, outlined in Chapter 2, was a systematic review of the psychosocial factors that are known to be associated or predictive of JIA pain. The literature exploring psychosocial factors related to JIA pain is scattered, methodologically diverse (e.g., using different instruments and reporters), and produces inconsistent findings (e.g., certain coping styles are related to worse JIA pain experiences in some but not all studies). A thorough synthesis of this literature can allow the community to ascertain the landscape of existing information, and to accurately interpret findings in the context of methodological differences. The first study of this dissertation addresses this gap. The objective was to synthesize the literature on factors associated with JIA-related pain to determine what psychosocial factors were 1) associated with, and 2) predictive of (i.e., prognostic factors) JIA pain (intensity, frequency, and sensitivity). The JBI methodology for etiology and risk (Aromataris & Munn, 2020) guided the conduct of this review, which included a three-step search strategy and transparent process for the selection of studies. Key databases were searched for terms related to JIA and pain in youth 0-17 years old, and original quantitative studies were included. This age range was selected as it aligns with the time at which youth present to pediatric health care systems (Clemente et al., 2016). Qualitative studies were excluded given the expansiveness of this review and the focus on quantitative outcomes (i.e., pain intensity, frequency, and sensitivity).

The protocol was pre-registered through the International Prospective Register of Systematic Reviews (PROSPERO; CRD42021266716). Eligible studies were critically appraised using JBI tools that assess details such as the inclusion criteria, validity of measures, and the appropriateness of statistics (see Tables 2.2 and 2.3 for specific items and ratings). While studies were not excluded based on their appraisal, it was considered in the interpretation of findings (e.g., null results may have been related to inadequate sample sizes). Finally, results were synthesized in a narrative and tabular format and were retrospectively organized by the lead author according to the Transactional Model of Stress and Coping (Lazarus & Folkman, 1984). This was recommended during the peer review process to enhance organization and interpretation. While it is expected that nuances may exist in what psychosocial factors are relevant given details such as the child's age, diagnosis, and disease activity, the aim of this review was to provide a comprehensive synthesis of the literature. These differences were nevertheless narratively described and noted within the tabular synthesis.

Given the sparsity of resilience factors identified in the literature, particularly at the resource and dyadic levels, the second study (Chapter 3) examined perfectionism as a novel resource within this population. This study, guided by the Ecological Resilience-Risk Model in Pediatric Chronic Pain (Cousins, Kalapurakkel, et al., 2015) and the Stress and Coping Cyclical Amplification Model of Perfectionism in Illness (SCCAMPI; Molnar et al., 2016), aimed to explore the role of youth and parent perfectionism as either a risk factor and/or a resilience resource in the context of JIA pain. It was hypothesized that youth and parent perfectionism would contribute to greater youth anxiety and depression by way of 1) greater negative self-evaluations in response to JIA pain, and 2)

greater self-concealment of JIA pain. Hypotheses were pre-registered through Open Science Framework (OSF). Demographic data, clinical variables, and psychosocial factors were collected from 156 youth with JIA (13-18 years of age) and their caregivers through a large-scale online survey. Data were analyzed through structural equation modelling, as informed by the actor-partner interdependence model (Cook & Kenny, 2016).

The third and final study, outlined in Chapter 4, took a novel approach to the study of factors contributing to resilience in youth with JIA. A limitation of the existing literature exploring resilience in the context of JIA pain is the emphasis on resources and mechanisms and their independent contributions (Hynes et al., 2019). This is a prerequisite to the study of resilience; however, the process of resilience is best understood by including a multitude of resources and mechanisms within the individual and their environment (Rutter, 2012; Ungar, 2011, 2018). The objectives of this study were to 1) explore the relevance of youth and parent resilience resources and mechanisms that have been identified in the broader literature (hypothesizing significant and positive bivariate correlations), and 2) to explore their relative importance (RI) in contributing to recovery/sustainability (i.e., pain intensity, functioning, HRQoL) and growth (i.e., benefit finding) outcomes. This study used the same data as collected in Chapter 3, inclusive of additional measures of resilience as were identified in Chapter 2 and the existing literature (Hynes et al., 2019). Correlations, multiple regression run through five structural equation models, and RI (calculated based on the Pratt Index; Pratt, 1987), were applied to these dyadic data to address the aims and hypotheses.

CHAPTER 2: A SYSTEMATIC REVIEW OF THE PSYCHOSOCIAL FACTORS ASSOCIATED WITH PAIN IN CHILDREN WITH JUVENILE IDIOPATHIC ARTHRITIS

The manuscript based on this study is detailed below. Yvonne Brandelli, under the supervision of Drs. Christine Chambers and Sean Mackinnon, was responsible for developing the research question, search strategy (in consultation with an evidence synthesis specialist), and data extraction procedures, as well as for leading the preregistration of this review (PROSPERO CRD42021266716). She contributed substantially and oversaw a team of staff and students who were also involved in the systematic search, screening, and data extraction processes. She led data analyses and wrote the current manuscript. Co-authors feedback was integrated prior to submission. The manuscript was peer-reviewed and Yvonne Brandelli led the relevant revisions. This manuscript was published in the open access journal, *Pediatric Rheumatology*, on June 16, 2023. The full reference for this manuscript is:

Brandelli, Y. N., Chambers, C. T., Mackinnon, S. P., Parker, J. A., Huber, A. M., Stinson, J. N., Wildeboer, E. M., Wilson, J. P., & Piccolo, O. (2023). A systematic review of the psychosocial factors associated with pain in children with juvenile idiopathic arthritis. *Pediatric Rheumatology Online Journal, 21*(57), 1-35. https://doi.org/10.1186/s12969-023-00828-5

2.1. Abstract

Background: Pain is one of the most frequently reported experiences amongst children with Juvenile Idiopathic Arthritis (JIA); however, the management of JIA pain remains challenging. As pain is a multidimensional experience that is influenced by biological, psychological, and social factors, the key to effective pain management lies in understanding these complex relationships. The objective of this study is to systematically review the literature on psychosocial factors of children with JIA and their caregivers 1) associated with and 2) predictive of later JIA pain intensity, frequency, and sensitivity in children 0-17 years of age.

Methods: The Joanna Briggs Institute methodology for etiology and risk and Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) statement guided the conduct and reporting of this review. Terms related to pain and JIA were searched in English without date restrictions across various databases (PubMed, CINAHL, PsycINFO, Embase, Scopus, and the Cochrane Central Register of Controlled Trials) in September 2021. Two independent reviewers identified, extracted data from, and critically appraised the included studies. Conflicts were resolved via consensus.

Results: Of the 9,929 unique studies identified, 61 were included in this review and reported on 516 associations. Results were heterogeneous, likely due to methodological differences and moderate study quality. Results identified predominantly significant associations between pain and primary and secondary appraisals (e.g., more child pain beliefs, lower parent/child self-efficacy, lower child social functioning), parent/child internalizing symptoms, and lower child well-being and health-related quality of life. Prognostically, studies had 1-to-60-month follow-up periods. Fewer beliefs of harm,

disability, and no control were associated with lower pain at follow-up, whereas internalizing symptoms and lower well-being were predictive of higher pain at follow-up (bidirectional relationships were also identified).

Conclusions: Despite the heterogeneous results, this review highlights important associations between psychosocial factors and JIA pain. Clinically, this information supports an interdisciplinary approach to pain management, informs the role of psychosocial supports, and provides information to better optimize JIA pain assessments and interventions. It also identifies a need for high quality studies with larger samples and more complex and longitudinal analyses to understand factors that impact the pain experience in children with JIA.

2.2. Introduction

Pain is a common experience reported by children with Juvenile Idiopathic Arthritis (JIA; Canadian Paediatric Society, 2009). The pain is variable in intensity (Schanberg et al., 2003; Tupper et al., 2013), enduring (Rashid et al., 2018; Shiff et al., 2018), only mildly associated with disease activity (Bromberg et al., 2014; Kimura et al., 2006), and associated with a host of negative outcomes (e.g., reduced participation, quality of life, and mental health challenges; Fair et al., 2019; Sawyer et al., 2005; Stinson et al., 2011). In a recent qualitative study, healthcare providers (HCP) identified a lack of training and confidence in managing JIA pain, which led some to actively avoid talking about pain (R. R. Lee et al., 2020). Evidently, there are important unmet needs pertaining to the understanding, assessment, and management of pain in JIA (Giancane et al., 2017).

Pain is defined as "an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage […] that is a personal experience that is influenced to varying degrees by biological, psychological, and social factors" (Raja et al., 2020). In other words, pain is developed and maintained by biological (e.g., genetics, disease activity, medications), psychological (e.g., emotions, cognitions), and social/environmental (e.g., parents, peers) factors. Thus, while biological factors such as a diagnosis of JIA can increase one's susceptibility and sensitivity to noxious stimuli, psychological and social (i.e., psychosocial) factors can also influence how pain is perceived. This is particularly important in the context of pediatric pain, wherein parent and family factors can interact with a child's development to affect their pain experience (Palermo et al., 2014). In considering the transactional model of stress

and coping (Lazarus & Folkman, 1984), while the presence of JIA pain may present as a potential stressor, primary appraisals (e.g., whether it is perceived as dangerous), secondary appraisals (e.g., whether an individual has sufficient internal and external resources to manage it), how one copes, and its subsequent outcomes (e.g., well-being, mental health) can all influence the pain experience. Understanding the components that develop and maintain one's pain are crucial to advancing the knowledge and management of JIA pain.

The relationships between biological, psychological, and social factors and JIA pain have been explored to varying degrees over the past four decades. Biological and disease-related factors have been explored extensively. Worse pain has been associated with enthesitis-subtype (Weiss et al., 2012), greater active joint count (Weiss et al., 2012), greater functional impairment (Rashid et al., 2018), and greater sleep disturbance (Stinson, Hayden, et al., 2014), whereas engagement in physical activity has been shown to be associated with decreased pain (Cavallo et al., 2017; Kuntze et al., 2018; Takken et al., 2008; Tarakcı et al., 2021). Age and sex have more inconsistent results (Stinson, Luca, et al., 2012), although recent research has suggested that females are at slightly greater risk of worse pain (Zweers et al., 2021). Psychosocial factors have been explored to a lesser degree. While the child's mood/mental health (Fair et al., 2019), quality of life/well-being (e.g., Taxter et al., 2015), cognitions and coping strategies (e.g., Thastum et al., 2005), family functioning (e.g., Thompson et al., 1987), and psychological therapies (Butler et al., 2022; Cohen et al., 2017) have also been explored in relation to JIA pain, results across these variables are not always consistent and have been measured in different ways.

The sensation of pain, for example, can be measured in terms of its intensity, frequency, or sensitivity in response to a noxious stimuli (i.e., hyperalgesia). Even these measures can be assessed in different ways (e.g., paper or electronic diaries, current or retrospective reports, self- or proxy-reports; Palermo, Zebracki, et al., 2004), all of which can affect the interpretation and comparability of results. As such, a formalized review is needed to make sense of discrepancies across studies and accurately interpret findings in the context of methodological differences. Moreover, the synthesis of details such as study sample size, age, diagnosis, measures, and research design (e.g., whether factors are correlated or predictive) allows readers to fully ascertain the landscape of information.

Given the greater emphasis and consistency in the literature about what biological and disease-related factors are most relevant to consider, the emphasis of this review is on psychosocial factors. The objective of this study is to synthesize the literature on factors associated with JIA-related pain to determine what psychosocial factors in both individuals with JIA and others in their social environment (e.g., caregivers) are 1) associated with and 2) predictive of (i.e., prognostic factors) JIA pain (intensity, frequency, sensitivity).

2.3. Methods

This systematic review followed the Joanna Briggs Institute (JBI) methodology for etiology and risk (Aromataris & Munn, 2020) and The Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA; Page et al., 2021). This study was pre-registered with the international prospective register of systematic reviews (PROSPERO CRD42021266716).

2.3.1. Eligibility Criteria

Population

This review included studies about children (0-17 years of age) with a diagnosis of JIA. The cut-off age was 17 years as many youth transition from pediatric to adult health systems around that age (Clemente et al., 2016). Studies reporting on children with comorbidities or rheumatic diseases other than JIA (Petty et al., 2004) were excluded to avoid potential confounds. Studies including broader age ranges (e.g., 0-18 years of age) or diagnoses (e.g., juvenile rheumatic diseases) were retained only if data were reported separately for children ages 0-17 years with JIA. Self- and proxy-reported data were included.

Exposure and Outcome

Studies were included if they explored psychosocial factors associated with pain. This review used the most frequently assessed sensory components of JIA pain as the outcome: pain intensity, frequency, and sensitivity. Psychosocial factors were defined as factors within oneself (e.g., beliefs, coping, mood/affect) and the environment (e.g., parent/family factors, school and social functioning) that were associated with pain (Goubert et al., 2021). Psychosocial factors were included with Aim 1 if they were associated with pain at any point in time (i.e., correlated with or predicted by pain) and in Aim 2 if they predicted later pain (i.e., temporal precedence was established).

Types of Studies

All quantitative studies published in the English language were included. No date restrictions were applied; however, dates were considered in the synthesis of results given an important shift in the treatment of JIA in the 2000s with the advent of biological

agents. Observational designs were considered associations, whereas cohort designs were considered prognostic depending on the analyses. Qualitative studies, studies not reporting original data (e.g., reviews), and the grey literature were excluded.

2.3.2. Search Strategy

The search strategy aimed to identify all published studies pertaining to this review. Following the JBI methodology, a three-step search strategy was applied with the support of an evidence synthesis librarian (LB). First, a limited search was conducted of PubMed, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), and Medline at OVID with keywords related to JIA, pain (Schinkel et al., 2017), and pediatrics (Leclercq et al., 2013), to ensure the search strategy encompassed pertinent terms. Second, the comprehensive search, inclusive of any keywords and index terms identified in the limited search, was completed on September 21st, 2021 (Additional File 1). The databases searched included Medline at OVID, CINAHL, PsycINFO, the Cochrane Central Register of Controlled Trials, Embase, and Scopus. Third, the reference list (backwards) and citing articles (forwards) of the included articles were searched for any additional studies. The search was updated on June 7th, 2022 to identify any recently published articles.

2.3.3. Study Selection

References were uploaded to Covidence systematic review software (Veritas Health Innovation, Melbourne, Australia). Duplicates were removed automatically and manually. Titles and abstracts were double screened for eligibility by two independent reviewers (always YNB, either EMW or OP). Relevant full texts were located, uploaded, and double screened for eligibility by the same reviewers. Inter-rater agreement was

established using a weighted Cohen's Kappa (poor: $\kappa < 0.00$; slight: $\kappa = 0.00 – 0.20$, fair: $\kappa = 0.21 – 0.40$, moderate: $\kappa = 0.41 – 0.60$, substantial: $\kappa = 0.61 – 0.80$, and almost perfect: $\kappa = 0.81 – 1.00$; Landis & Koch, 1977). Discrepancies were resolved via consensus (YNB, EMW, and OP).

2.3.4. Methodological Quality Assessment

The methodological quality of the included studies was critically appraised by two independent reviewers (always YNB, either EMW or OP) using the JBI critical appraisal instruments (Aromataris & Munn, 2020). These standardized instruments assess the presence of various methodological limitations (e.g., participant selection, measurement bias, confounds) in a "yes", "no", or "unclear" format. Different instruments were used based on the study design and way in which the data relevant to this review were collected (i.e., separate instruments were used for analytical cross-sectional studies, cohort studies). No attempts were made to contact authors for additional information. Discrepancies were resolved via consensus (YNB, EMW, and OP).

2.3.5. Data Extraction

A data extraction template was developed and pilot tested for this review. The template included information regarding the study, population, measures, and results (Additional File 2). Two independent reviewers (always YNB, either EMW or OP) extracted data from the included articles and discrepancies were resolved through consensus (YNB, EMW, and OP).

2.3.6. Data Synthesis

Given the heterogeneity of associations explored, data were synthesized narratively and in tabular form. Studies were grouped together based on the psychosocial

factors. Similarities (e.g., significance of associations) and differences (e.g., reporter) across studies were explored.

2.4. Results

2.4.1. Study Inclusion

The systematic search returned 9,929 unique studies, 61 of which were included in this review (Amine et al., 2009; Anthony et al., 2011; Armbrust et al., 2016; Baildam et al., 1995; Baloueff, 1996; Barlow et al., 2000, 2001; Barlow et al., 2002; Bromberg, 2009; Bromberg et al., 2012; Bruns et al., 2008; Connelly et al., 2012; Cornelissen et al., 2014; Dimitrijevic Carlsson et al., 2019; Doherty et al., 1993; El-Najjar et al., 2014; Hagglund et al., 1995; Hanns, 2018; Hoff et al., 2006; Jaworski, 1992; Klotsche et al., 2014; Kovalchuk et al., 2017; Kovalchuk et al., 2018; Lavigne et al., 1992; Listing et al., 2018; Lomholt et al., 2015; Lomholt et al., 2013; Luca et al., 2017; Mahler et al., 2017; Margetić et al., 2005; Oen et al., 2021; Oen et al., 2009; Rashid et al., 2018; Ross et al., 1993; Sällfors et al., 2004; Schanberg et al., 2003; Schanberg et al., 2005; Selvaag et al., 2003; Selvaag et al., 2005; Shelepina et al., 2011; Stinson, 2006; Stinson et al., 2016; Stinson et al., 2020; Tarakcı et al., 2011; Tarkiainen et al., 2019; Thastum & Herlin, 2011; Thastum et al., 2005; Thastum et al., 1997; Thastum et al., 1998; Thompson et al., 1987; Tupper, 2012; Tupper et al., 2013; Upadhyay et al., 2021; Vandvik & Eckblad, 1990; Vuorimaa et al., 2009, 2011; Vuorimaa et al., 2008; Walco et al., 1992; Yan et al., 2020). The PRISMA chart (Figure 2.1) relays the search results and inclusion process (Page et al., 2021). Between rater reliability was moderate to substantial at the Title/Abstract screening stage ($\kappa = 0.58$ & 0.61) and substantial at the Full Text screening stage ($\kappa = 0.61$ & 0.73).

2.4.2. Description of Studies

The 61 included studies came from 59 articles and 49 unique datasets. Studies reporting on the same datasets were included only if new associations were identified (i.e., identical associations in multiple publications on the same dataset were removed). Publication dates ranged from 1987 to 2021. Most of the articles included were peer-reviewed publications, however two conference abstracts (Mahler et al., 2017; Shelepina et al., 2011) and six theses were also included (Baloueff, 1996; Bromberg, 2009; Hanns, 2018; Jaworski, 1992; Stinson, 2006; Tupper, 2012). The six theses were selected over published manuscripts as additional associations were identified. Articles came from 17 countries, with the United States, Canada, the United Kingdom, and Denmark being the most represented. Most recruitment took place in clinics apart from two studies wherein it was unclear (Kovalchuk et al., 2017; Shelepina et al., 2011). Participants were predominantly children with JIA; however, 34 studies included parent/caregiver reports and two studies included HCP reports. Sample sizes ranged from 11 to 1906 participants (Mdn = 85; IQR = 99). Participants were largely female children (Mdn = 67%, IQR = 11%) and caregivers (Mdn = 83%, IQR = 17%), although some studies were missing these data. Other demographic information could not be aggregated given the variability of information reported on (e.g., medians or means, varying categories, missing information); however, most studies reported on children in the adolescent period (with only 7 studies including children younger than 5), with polyarticular and oligoarticular JIA as the most represented diagnoses.

Of the 516 unique associations, 234 were significant as per the α level used in each study. Fifty-one were classified as prognostic factors. Validated measures were

generally used to measure pain intensity (Bieri et al., 1990; Billings et al., 1987; Cella et al., 2007; Cleeland, 2009; Filocamo et al., 2011; Hicks et al., 2001; Luca et al., 2017; Singh et al., 1994; Stinson et al., 2008; Varni et al., 1987; Von Korff et al., 1992); although, 109 associations provided no or unclear references. Pain frequency (Filocamo et al., 2011; Hicks et al., 2001; Mikkelsson et al., 1996; Varni et al., 1987) and sensitivity (Meier et al., 2001; Zeltzer et al., 1989) were largely assessed using standardized measures and protocols. Pain was measured via self-report in 46 studies, proxy-report in 15 studies, and an unclear reporter in seven studies. Psychosocial factors were organized based on the transactional model of stress and coping (Lazarus & Folkman, 1984) and included both child and parent factors. Validated measures were used to assess children's primary appraisals (i.e., interpretations of whether JIA pain is positive, irrelevant, or threatening/harmful) (Cleeland, 2009; Jensen et al., 1994; Stinson et al., 2008); children's internal (Barlow et al., 2001; Cella et al., 2007; Cohen et al., 1983; Harter, 1985, 1988; Landgraf et al., 1996) and external (Achenbach & Edelbrock, 1983; Billings et al., 1987; Gresham & Elliott, 1990; Harter, 1985, 1988; Kerns et al., 1985; Landgraf et al., 1996; Lavigne et al., 1992; Lomholt et al., 2015; Moos & Moos, 1987; Reynolds & Richmond, 1985; Sarason et al., 1987; Singh et al., 1994; Stinson et al., 2016; Stinson et al., 2020; Varni, 1998a, 1998b; Walco et al., 1992) and parent's internal (Barlow et al., 2000; Landgraf et al., 1996) secondary appraisals (i.e., assessment of resources available to manage JIA pain); children's coping (Crombez et al., 2003; Gil et al., 1991; Rosenstiel & Keefe, 1983; Thastum et al., 1998); and outcomes including children's (Achenbach & Edelbrock, 1983; Angold et al., 1995; Birmaher et al., 1995; Briere, 1996; Cella et al., 2007; Chorpita et al., 2000; Cleeland, 2009; Faulstich et al., 1986; Forsyth et al., 1996;

Harter, 1985, 1988; Kazdin et al., 1983; Kotsch et al., 1982; Kovacs, 1985; Kroenke et al., 2009; Landgraf et al., 1996; Laurent et al., 1999; McGrath et al., 1996; Pagano et al., 2000; Reynolds & Richmond, 1985; Shields & Cicchetti, 1997; Spielberger et al., 1983; Stinson et al., 2008; Varni, 1998a, 1998b; Zeman et al., 2001) and parent's (Beck et al., 1996; DeLongis et al., 1988; Harding et al., 1980; Landgraf et al., 1996; Lanyon, 1978; Medeiros et al., 1998; Zigmond & Snaith, 1983) mental health, and children's health-related quality of life (HRQoL; i.e., the impact of one's health on their life; Feldman et al., 2000) and well-being (i.e., one's sense of how well their needs are being met; Costanza et al., 2007) (Duffy et al., 1997; Feldman et al., 2000; Landgraf et al., 1996; Shaffer et al., 1983; Singh et al., 1994; Varni, 1998a, 1998b; World Health Organization, 1998). Twenty associations exploring well-being provided no citation. Table 2.1 outlines the exact measures and their frequency of use. Six quasi-experimental studies explored pain in relation to psychosocial interventions (Lavigne et al., 1992; Lomholt et al., 2015; Stinson et al., 2016; Stinson et al., 2020; Walco et al., 1992). The manipulation set them aside from other studies included in this review, thus the results have been included in Additional Files 3.1-3.3 and the figures.

2.4.3. Methodological Quality

The included studies were critically appraised using JBI tools (Moola et al., 2020; Tufanaru et al., 2020) based on the associations used in the review rather than the stated study design (e.g., daily diary studies were categorized as cross-sectional or cohort depending on how the data were analyzed, studies with pain predicting psychosocial factors were considered cross-sectional designs). For the two theses that contained two studies each, separate appraisals were conducted. Fifty-one studies were cross-sectional

and five were cohort. No studies were excluded based on the critical appraisal.

The median critical appraisal score was 75% (IQR = 20%). For the 51 cross-sectional studies, scores ranged from 38% to 100%, with the identification and management of confounds as the greatest weakness (Table 2.2). For the five cohort studies, scores ranged from 40% to 89%, with the validity of the outcome measurement (i.e., pain) as the lowest rated item (Table 2.3).

2.4.4. Findings of the Review

Findings of the review have been grouped based on the study aims, categories as they map to the transactional model of stress and coping (Lazarus & Folkman, 1984), and child/parent factors. See Table 2.4 for study details and Figure 2.2/Additional File 4 for a summary.

Aim 1: Psychosocial Correlates

Primary Appraisals

Child correlates. There were 5 studies reporting on 28 associations between primary appraisals and pain in children with JIA. Pain unpleasantness was positively associated with pain intensity (in 5/5 associations; herein referred to as 5/5) (Stinson, 2006). Pain beliefs were significantly associated with pain intensity in 14/20 associations (Thastum & Herlin, 2011; Thastum et al., 2005) and pain frequency in 2/3 associations (Lomholt et al., 2013). Specifically, beliefs that pain causes harm and disability were positively associated with pain (5/5 each). Beliefs that one lacks control over their pain were positively associated with pain intensity (3/3) but not frequency (0/1). Beliefs there is no cure and that others should help with their pain (i.e., solicitude) were partially associated with pain intensity (1/3 and 1/2, respectively); whereas beliefs that emotions

affect pain were not (0/2).

Taken together, although primary appraisals have been studied infrequently, perceptions of pain unpleasantness and beliefs that pain causes harm, disability, and loss of control appear to be consistently related to worse pain experiences in youth with JIA.

Secondary Appraisals – Internal Factors

Child correlates. There were 7 studies reporting on 22 associations between internal factors a child may consider in their secondary appraisal and JIA pain, 8 of which were significant. Self-efficacy was negatively associated with pain intensity in 3/3 associations. Barlow, Shaw, and Wright (2001) developed a measure to assess self-efficacy in children with arthritis. Each of the subscales (activity, emotion, and symptom) demonstrated a significant negative correlation to pain intensity. Vuorimaa and colleagues (2011) used the same measure with a different factor structure (Vuorimaa et al., 2007) in relation to pain frequency, wherein 2/6 associations were significant (i.e., social self-efficacy but not psychological or somatic self-efficacy). Four additional internal factors were explored in relation to JIA pain. Neither children's perceptions of their physical appearance (0/3) (Baloueff, 1996) nor child- or parent-reported self-esteem (0/4) (Baloueff, 1996; Kovalchuk et al., 2018) were associated with pain intensity. Stress was positively related to pain intensity in 2/4 associations (Dimitrijevic Carlsson et al., 2019; Schanberg et al., 2005; Upadhyay et al., 2021); however, it is worth noting that nonsignificant results were only observed in one study with a small sample size (n = 16). Interestingly, difficulties with cognitive functioning were negatively correlated with pain intensity in select analyses (1/2) (Upadhyay et al., 2021).

Parent correlates. Four studies reported on 17 associations between parent

cognitive factors and pain in children with JIA. Of those, 8/17 were significant. Parent self-efficacy was negatively associated with pain intensity in 4/10 associations (Barlow et al., 2000; Barlow et al., 2002) and pain frequency in 4/6 associations (Vuorimaa et al., 2011). Specifically, psychosocial and symptom self-efficacy were negatively related to child pain intensity in 3/5 and 1/5 associations, respectively (Barlow et al., 2000; Barlow et al., 2002). Somatic and social self-efficacy, but not psychological self-efficacy, were negatively related to child pain frequency in 2/2 associations each (Vuorimaa et al., 2011). Parent self-esteem was not related to children's JIA pain (0/1) (Kovalchuk et al., 2018).

Taken together, despite the small sample sizes used in many of these studies, various domains of parent and child self-efficacy and children's perceptions of stress have shown important associations to children's JIA pain experiences.

Secondary Appraisals – External Factors

Child correlates. Sixteen studies reported on the relationship between social factors (i.e., school and social functioning, parent responses to pain, family functioning) and pain in children with JIA, with 30/105 significant associations. School functioning was significantly associated with pain intensity in 13/19 associations (Armbrust et al., 2016; Baloueff, 1996; Doherty et al., 1993; Klotsche et al., 2014; Sällfors et al., 2004; Schanberg et al., 2005; Shelepina et al., 2011) and pain frequency in 1/1 association (Sällfors et al., 2004). Greater pain was associated with more school absences or reduced school activity (6/8) (Armbrust et al., 2016; Doherty et al., 1993; Sällfors et al., 2004) and home-schooling compared to traditional schooling (1/1) (Shelepina et al., 2011). Pain did not appear to be associated with children's perceptions of their scholastic competence

(0/3) (Baloueff, 1996). Similarly, social functioning and pain were significant related in 9/35 associations. More specifically, social functioning was significantly associated with pain intensity in 8/34 associations (Baloueff, 1996; Hagglund et al., 1995; Klotsche et al., 2014; Schanberg et al., 2005; Stinson, 2006; Thompson et al., 1987) and frequency in 1/1 association (Schanberg et al., 2003). Klotsche and colleagues (2014) found decreases in pain over time predicted better school and social functioning across 7/8 timepoints within one year. Schanberg and colleagues (2003) also found a positive correlation between social concerns and pain frequency, and that pain scores were associated with increased odds of foregoing social activity (2/2) (Schanberg et al., 2003; Schanberg et al., 2005). No other associations were significant between pain and components of social functioning including social support, competence, skills, self-control, acceptance, communication, assertion, cooperation, or empathy (0/25) (Baloueff, 1996; Hagglund et al., 1995; Stinson, 2006; Thompson et al., 1987).

Five studies reported on relationships between parent specific resources and children's pain intensity, all of which had a sample size of less than 60 parents. Parent influences on the child's mood (Vuorimaa et al., 2011) and responses to the child's pain (Jaworski, 1992) were not associated with pain frequency or intensity (0/11); however, the measures used were not validated in this population. Family factors were variably related to pain intensity (Kovalchuk et al., 2018; Ross et al., 1993; Thompson et al., 1987). In some analyses, independence (1/3), achievement orientation (1/3), intellectual-cultural orientation (1/3), activities (1/2), cohesion (1/5), and expressiveness (1/3) were negatively associated with pain intensity, whereas harmony (1/2) was a positive relationship. Other factors including conflict, control, relationships, moral-religious

emphasis, active-recreational orientation, and organization demonstrated no relationships (0/18).

Taken together, JIA pain is consistently associated with lower school and social functioning, but less related to actual skills. Although parent and family factors demonstrated less of a relationship, the studies included used small sample sizes and adapted measures.

Coping

Child correlates. Pain coping strategies were frequently assessed, and significantly associated with pain intensity in 15/61 associations (Bromberg, 2009; Dimitrijevic Carlsson et al., 2019; Stinson, 2006; Thastum et al., 2005; Thastum et al., 1997; Thastum et al., 1998), pain frequency in 3/6 associations (Lomholt et al., 2013; Vuorimaa et al., 2011), and pain sensitivity in 2/21 associations (Cornelissen et al., 2014; Thastum et al., 1997; Thastum et al., 1998). Greater coping ability and efficacy were negatively associated with pain (3/3) (Bromberg et al., 2012; Vuorimaa et al., 2011). Distraction is often cited as an adaptive coping strategy; however, only behavioral distraction was negatively associated with pain (4/9) (Lomholt et al., 2013; Thastum et al., 2005; Thastum et al., 1998). Neither broad measures of distraction (0/6) (Stinson, 2006; Thastum et al., 1997) nor measures of cognitive distraction (0/9) (Lomholt et al., 2013; Thastum et al., 2005; Thastum et al., 1998) were associated with pain. The use of positive self-statements is also presumed to be an adaptive coping style and was negatively associated with pain intensity (but not frequency or sensitivity) in 4/9 associations (Lomholt et al., 2013; Thastum et al., 2005; Thastum et al., 1998). Catastrophizing is often cited as a maladaptive coping strategy, which was positively

associated with pain intensity, frequency, and sensitivity in 7/22 associations (Cornelissen et al., 2014; Dimitrijevic Carlsson et al., 2019; Lomholt et al., 2013; Thastum et al., 2005; Thastum et al., 1997; Thastum et al., 1998). The remaining coping strategies were minimally or not associated with pain: externalizing (1/9) (Thastum et al., 2005; Thastum et al., 1998); emotion focused avoidance (1/2) (Stinson, 2006); and seeking social support, information seeking, approach, and reinterpretation (0/19) (Stinson, 2006; Thastum et al., 2005; Thastum et al., 1997; Thastum et al., 1998). Many studies exploring pain coping had relatively small sample sizes, likely contributing to the heterogeneity in results.

Taken together, despite some variability, children's coping strategies of catastrophizing, behavioral distraction, and positive self-statements tended to show an important relationship to JIA pain.

Outcomes

Child correlates. Forty-two studies reported on 183 associations between pain and outcomes such as children's pain interference, mental health, and well-being, 104 of which were significant. Although a comprehensive review of the physical/functional limitations imposed by JIA pain were beyond the scope of this review, three studies found that the interference that pain imposed on daily activities was positively associated with pain intensity in 13/13 associations (Kovalchuk et al., 2018; Stinson, 2006).

Broad measures of child mental health were not significantly associated with pain intensity (0/8 associations) (Baildam et al., 1995; Kovalchuk et al., 2018; Thompson et al., 1987; Vandvik & Eckblad, 1990) or sensitivity (0/8) (Cornelissen et al., 2014). Externalizing symptoms (e.g., behavior) were also not associated with pain intensity

(0/12) (Baloueff, 1996; Kovalchuk et al., 2018; Ross et al., 1993; Thompson et al., 1987; Vandvik & Eckblad, 1990), a finding that was stable across measures, reporters (parent, child), sample sizes (i.e., 23-60), and analyses (e.g., correlations, regressions). Internalizing symptoms (e.g., distress, emotional functioning) were positively associated with pain intensity in 10/16 associations (Anthony et al., 2011; Dimitrijevic Carlsson et al., 2019; Klotsche et al., 2014; Ross et al., 1993; Thompson et al., 1987; Vandvik & Eckblad, 1990; Vuorimaa et al., 2009) and with pain frequency in 1/1 association (Vuorimaa et al., 2008). Most of the nonsignificant relationships used a proxy report to measure internalizing symptoms. Anxiety symptoms were positively associated with pain in 11/23 associations. More specifically, anxiety symptoms were positively associated with pain intensity in 4/10 associations (Margetić et al., 2005; Ross et al., 1993; Schanberg et al., 2003; Stinson, 2006; Tarakcı et al., 2011; Upadhyay et al., 2021), pain frequency in 5/5 associations (Schanberg et al., 2003; Vuorimaa et al., 2011), and pain sensitivity in 2/8 associations (Cornelissen et al., 2014). Across these studies, nonsignificant relationships tended to be more prevalent in studies with smaller sample sizes (i.e., 6/10 associations where $n \leq 52$). Depression symptoms were positively associated with pain in 21/44 associations. Specifically, depression symptoms were positively associated with pain intensity in 19/42 associations (Bromberg et al., 2012; Connelly et al., 2012; El-Najjar et al., 2014; Hagglund et al., 1995; Hanns, 2018; Jaworski, 1992; Margetić et al., 2005; Rashid et al., 2018; Ross et al., 1993; Schanberg et al., 2003; Tarakcı et al., 2011; Tupper, 2012; Upadhyay et al., 2021; Yan et al., 2020) and pain frequency in 2/2 associations (Vuorimaa et al., 2011). While most scales assessed various depression symptoms (e.g., Children's Depression Inventory, Mood and Feelings

Questionnaire), some studies explored individual symptoms. Negative affect (Bromberg et al., 2012; Tupper, 2012), but not hopelessness or sadness (Hagglund et al., 1995), was found to be positively associated with greater pain intensity. Using a daily diary methodology, Connelly and colleagues (2012) explored the relationship between emotion regulation and pain intensity. Although lower pain intensity was not correlated with child- or parent-reported emotion regulation or the adaptive upregulation of positive emotions, findings suggested that children with lower pain intensity were better able to manage their negative emotions and had fewer mood fluctuations day-to-day (i.e., less variability in positive and negative affect). Two studies explored the impact of pain on depression symptoms longitudinally. Hanns (2018) found that higher baseline pain intensity was associated with worse depression symptoms over 12 months; results that were in keeping with other studies (Yan et al., 2020). Across these associations, nonsignificant results were common in studies published before the year 2000; however, these studies also tended to report on younger samples (e.g., childhood) and using parent reports of depression symptoms (i.e., 7/7).

Greater HRQoL was significantly associated with lower pain intensity (28/37) (Amine et al., 2009; Klotsche et al., 2014; Kovalchuk et al., 2017; Kovalchuk et al., 2018; Listing et al., 2018; Luca et al., 2017; Oen et al., 2021; Oen et al., 2009; Selvaag et al., 2003; Stinson, 2006; Tarakcı et al., 2011; Tarkiainen et al., 2019; Vandvik & Eckblad, 1990) and lower pain intensity variability (1/1) (Tupper et al., 2013), and greater well-being was significantly associated with lower pain intensity (15/16) (Kovalchuk et al., 2018; Mahler et al., 2017; Oen et al., 2009; Rashid et al., 2018; Sällfors et al., 2004; Selvaag et al., 2005; Tarakcı et al., 2011) and pain frequency (4/4)

(Sällfors et al., 2004; Vuorimaa et al., 2011). These findings were consistent across measures (e.g., Childhood Health Assessment Questionnaire, Pediatric Quality of Life Inventory), timeframes (e.g., usual, past week), reporters (child, parent, HCP), and analyses (e.g., correlations, regressions). In addition to cross-sectional studies, Listing and colleagues (2018) found that greater pain intensity at baseline was not only associated with lower HRQoL at baseline, but also 36 months later. Similar results were found by others (Klotsche et al., 2014; Oen et al., 2009; Tarkiainen et al., 2019). Nonsignificant results were more prevalent in studies with small sample sizes (i.e., 3/5 studies where $n \leq 36$) and those assessing psychosocial HRQoL especially with the Child Health Questionnaire (7/11 studies).

Parent correlates. Six studies reported on 22 associations between parent mental health outcomes and JIA pain. Mothers' mental health was over-represented (samples ranged from 83% to 100% female). Across these studies, 9/22 associations were significant. Parent internalizing symptoms (e.g., distress) were positively related to child pain intensity in 2/3 associations (Bruns et al., 2008; Ross et al., 1993). Parental symptoms of anxiety were not associated with child pain intensity or frequency (0/3) (Barlow et al., 2002; Vuorimaa et al., 2011). Parental symptoms of depression were positively associated with pain frequency (3/4) (Vuorimaa et al., 2011), but not intensity (0/2) (Anthony et al., 2011; Barlow et al., 2002); however, the latter two studies had smaller sample sizes ($n \leq 51$). Parent identified limitations that pain imposed on their daily activities were positively associated with their child's pain in 4/10 associations (Anthony et al., 2011; Bruns et al., 2008; Kovalchuk et al., 2018). More specifically, Bruns and colleagues (2008) were unable to demonstrate a relationship between caregiver

burden and child pain intensity; however, Kovalchuk and colleagues (2018) found that time and emotional impact were positively correlated with parent- (but not child-) reported pain intensity. Furthermore, Anthony and colleagues (2011) found that although parent-reported hassles (i.e., perceptions of daily events like the weather and their workload as negative) were not significantly associated with child pain intensity, the frequency of parent-reported uplifts (i.e., parents identifying daily events as positive) was interestingly associated with greater child-reported pain.

Taken together, internalizing symptoms in children (anxiety, depression, and interference) and parents (depression, impacts on time and emotions, and more frequent uplifts) tend to demonstrate reliable associations to greater pain in children in studies with sufficient sample sizes using validated self-report measures, whereas greater HRQoL/well-being appears to be robustly related to lower JIA pain in children with JIA.

Aim 2: Prognostic Factors

Primary Appraisals

Child factors. The relationship between pain beliefs and pain were assessed prognostically in one study (Thastum & Herlin, 2011), wherein 4/5 associations were significant. Following up on their earlier work, Thastum and Herlin (2011) explored the impact of pain beliefs on pain intensity two years later. They found that baseline beliefs of harm, disability, and lack of control (but not that there is no medical cure) were positively correlated with later pain intensity, and that cognitive beliefs (i.e., the sum of the above beliefs) predicted greater pain intensity two years later. Taken together, pain beliefs are an important prognostic factor for later JIA pain experiences.

Outcomes

Child factors. Prognostically, the relationship between depression symptoms and pain intensity were explored in four studies (Connelly et al., 2012; Hanns, 2018; Hoff et al., 2006; Rashid et al., 2018). Of those, depression symptoms significantly predicted pain intensity in 7/14 associations. Connelly and colleagues (2012) used a 28-day daily diary study to explore whether emotion regulation predicted pain intensity. Through linear mixed models, they found similar results longitudinally as were reported cross-sectionally. Namely, greater variability in positive and negative emotions predicted greater pain intensity over time, and the adaptive upregulation of positive emotions following a drop in emotions predicted lower pain intensity over time. Two studies using the same database (Hanns, 2018; Rashid et al., 2018) found that more depression symptoms at baseline predicted greater pain intensity and less improvement in pain over at least one year. Rashid and colleagues (2018) went on to conduct a group-based trajectory analysis, however no differences in depression symptoms across pain groups were observed. Finally, Hoff and colleagues (2006) assessed depression symptoms and pain intensity dyadically over 12 months. Although child-reported baseline depression symptoms did not predict later parent-reported pain intensity, it predicted later child-reported pain intensity when pain was low at baseline.

The relationship between well-being and pain was also explored by Rashid and colleagues (2018), wherein 4/8 associations were significant. Worse baseline well-being was significantly correlated with less change in pain intensity over 12 months; however, change in well-being was not correlated with change in pain intensity. Moreover, in their group-based analyses, the “consistently high” and “improved pain” groups had significantly worse baseline well-being than the “consistently low” pain group, and

improvements in well-being at six months were more likely in the "improved pain" group compared to the "consistently low" pain group.

In sum, the predictive value of depression symptoms on later pain experiences appeared to be contingent on the specific symptoms assessed and the reporter of these variables. Nevertheless, greater depression symptoms and lower well-being were predictive of worse pain intensity over time, but both relationships are likely more complex.

2.5. Discussion

Pain is a common experience that affects children with JIA in many ways. Across 61 studies, 516 unique associations between pain and psychosocial factors were identified. Most studies explored these associations cross-sectionally, with 51 associations explored longitudinally. The studies were of moderate quality, with the identification of confounds, and validity of outcome (i.e., pain) measures as the biggest areas for improvement. All studies were nevertheless included. Various factors were explored in relation to JIA pain, speaking to the complex relationships that exist; however, the emphasis was predominantly on child outcomes (e.g., mental health, well-being) and less on primary and secondary appraisals within the child and caregiver. Within and between studies, only a few variables were always related to JIA pain (unpleasantness and interference; beliefs of harm, disability, and control). The heterogeneity of most results is likely attributable to the moderate study quality, variability in measures and reporters, and small sample sizes; publication year did not appear to impact results substantially across these categories. Various factors are nevertheless important to consider as the associations were generally significant and

trending in the same direction.

With regards to children's primary appraisals, two constructs were looked at in relation to JIA pain – perceptions of pain unpleasantness and pain beliefs. These perceptions and beliefs are assumptions of reality through which events such as arthritis pain can be interpreted, and are thereby presumed to affect coping efforts and the pain experience (Turner et al., 2000). For example, a child who believes their JIA pain is purely physical in nature may feel a lack of control over their pain, thus increasing the attention given to their pain experience. While only a few studies explored these associations, results consistently demonstrated that perceptions of unpleasantness and beliefs that pain signifies harm, causes functional disability, and is unable to be controlled were significantly associated with worse JIA pain cross-sectionally and longitudinally. Less consistently, beliefs that there is no cure, that emotions impact pain, and that others should respond solicitously tended to be associated with greater pain. Pain beliefs appear to be a promising area for future research, especially in conjunction with pain neuroscience as an intervention to target unhelpful beliefs.

A few constructs were explored pertaining to the child's and parent's assessments of their internal and external resources available to manage JIA pain (i.e., secondary appraisals). While some internal resources (self-esteem, cognitive functioning, stress, perceptions of physical appearance) were minimally explored, one was explored in greater depth. Self-efficacy is one's expectations of success in performing the behaviors required to meet a specific outcome (Bandura, 1977), which has theoretical implications for the actions one takes, the amount of effort exerted, and the nature of one's thoughts and emotions (Barlow et al., 2001). It is thought to be an key mechanism of change in

fostering resilience (Tomlinson et al., 2017). Although a relatively nascent construct in pediatric pain, within broader pain populations it has also been associated with lower pain severity (Jackson et al., 2014). Two teams explored self-efficacy in this population using different subscales and pain outcomes. Across these studies, both child and parent self-efficacy (albeit in different domains) were generally related to better pain experiences. Thus, self-efficacy is a vital construct for further exploration.

Various factors pertaining to external resources were also explored. While JIA pain was not associated with impaired social skills, it was generally associated with worse school (e.g., attendance, paying attention in class, keeping up with schoolwork) and social (e.g., getting along with others, having friends) functioning. These findings parallel the pain literature (Forgeron et al., 2010; Groenewald et al., 2019) and can be understood through the interpersonal fear avoidance model of pain (Goubert & Simons, 2014). The child's internal pain experience is theorized to lead to negative cognitions, which can contribute to avoidant behaviors (e.g., avoiding school or friends). This can limit the child's social support which, upon future secondary appraisals, can further aggravate the child's pain. Longitudinal designs are required to fully understand these pathways. This model also highlights how parents contribute to children's pain experiences. Parent pain responses (e.g., responding protectively, reinforcing activity restriction, distracting) were not significantly related to JIA pain in this review, which is in line with a recent meta-analysis demonstrating that they are more closely related to functional disability (Harrison et al., 2020). Family variables (e.g., harmony, cohesion) have also been postulated to affect pain intensity in JIA; however, in this review, as in the broader literature (Lewandowski et al., 2010), these relationships were unreliable. Pain

was inconsistently associated with greater harmony and less achievement, achievement orientation, expressiveness, activities, cohesion, and intellectual-cultural orientation. It is possible that JIA pain may cause a unique dynamic within the family, wherein the family engages in fewer activities, is less cohesive, and is more co-dependent. Greater family harmony was an interesting finding, which was theorized to be because an overly harmonious and responsive environment may reinforce pain behaviors (Ross et al., 1993). These results must be interpreted with caution given the small sample sizes of the studies exploring them. More research with larger samples, new pain-specific family measures, and longitudinal studies showing how family functioning varies with pain flares is warranted.

Coping, or the intentional use of thoughts and behaviors to manage stressful experiences (Compas et al., 2014), was also explored in relation to JIA pain. Certain coping strategies are posited to be adaptive and have the potential to improve the child's well-being and pain experience (e.g., seeking information and social support, problem solving, positive self-statements, distraction). Other strategies are viewed as maladaptive and are thought to be associated with worse well-being and pain (e.g., emotion-focused avoidance, catastrophizing, externalizing; Reid et al., 1998). With that said, there is significant variability in the pediatric pain literature regarding coping theories, measures, and responses (Nabbijohn et al., 2021), which was also observed in this review. While the associations identified in this review trended in the expected directions, results were neither straightforward nor unanimous. Specifically, only positive self-statements and behavioral distraction were generally associated with reduced pain, and only catastrophizing tended to be associated with greater pain. Strategies such as seeking

social support and information, externalizing, emotion-focused avoidance, and approach were not significant in either direction. These results are likely a function of the broader variability in the literature (Nabbijohn et al., 2021) as well as the small sample sizes of the included studies. Moreover, no studies investigated these findings longitudinally or explored parent coping. As such, there is a clear need for more theoretically-driven research understanding the role of child and parent coping in JIA pain cross-sectionally and longitudinally.

Outcomes in relation to JIA pain (e.g., mental health, well-being) were explored most frequently and are presumed to be a result of the primary and secondary appraisals and coping efforts and can subsequently influence future appraisals. One of the most consistent findings of this review was the negative relationship between pain and measures of HRQoL and well-being. Results were demonstrated cross-sectionally and longitudinally in both directions (i.e., pain predicting lower well-being and the reverse). In considering the multidimensional nature of pain, HRQoL comprises the evaluative component, or the way in which pain affects one's broader well-being such as their functioning (Melzack, 1975; Stinson & Prescott, 2021). Thus, the consistent and bidirectional relationships identified in this review are well grounded in the literature. Although nonsignificant results were observed, they were more prevalent in studies with smaller sample sizes and those using the Child Health Questionnaire (a measure reported to be confusing due to the varying response options and recall periods across items; Hullmann et al., 2011). Although broad measures of child mental health and externalizing symptoms were not related to JIA pain, significant associations were often observed with measures assessing internalizing symptoms, and more specifically symptoms of

interference, anxiety, and depression. Nonsignificant results tended to occur in younger samples, when proxy reports of internalizing symptoms or pain were used, and in studies with smaller sample sizes. As pain and internalizing symptoms are internal experiences, proxy reporters may not fully understand the child's experiences with either, leading to null results. Nevertheless, these findings parallel what has been seen in the broader pediatric pain literature (McKillop & Banez, 2016). With regards to the relationships between pain and depression symptoms, interestingly results were retained in longitudinal designs, with some studies finding that pain predicted later depression symptoms, and other studies demonstrating the reverse. Current frameworks suggest that rather than one causing the other, there is a shared vulnerability wherein pain and internalizing symptoms may develop and maintain one another (for reviews see: Jastrowski Mano et al., 2019; Soltani et al., 2019; Vinall et al., 2016).

The role of parent mental health is also salient in these frameworks. In this review, a small number of studies cross-sectionally explored the relationship between parent (largely maternal) mental health and JIA pain. Although anxiety symptoms were not related to pain, few studies examined this. Broader measures of internalizing and depression symptoms demonstrated a relationship to greater JIA pain in some but not all associations, as did scales assessing the impacts pain has on parents' time and emotions. This is consistent with the small to null effects found in a recent meta-analysis on the role of parent factors in pediatric pain (Donnelly et al., 2020). As suggested by the abovementioned frameworks, it is likely that the relationship does exist, however is more complex than correlations may suggest. According to social learning theory, a parent observing their child in pain may experience internalizing symptoms which through

modelling and specific responses may contribute to the child's own internalizing symptoms and draw greater attention to their pain experience. More research is needed to further test these frameworks, particularly as it relates to paternal mental health. Another interesting finding emerged, wherein more parent-reported uplifts, or positive events in the day, was associated with greater pain (Anthony et al., 2011). It was posited that increased pain led to parents being more attentive to positive daily experiences or that parents were more attentive to their child's pain when there were more positive events in the day; however, future research is warranted to test these theories.

In sum, numerous psychosocial correlates have been identified in relation to JIA pain, all of which have important implications in the child's future appraisals of JIA pain and are key targets for pain assessment and intervention. This study had strengths in its inclusion of multiple dimensions of the pain experience, a broad array of psychosocial factors, multiple reporters, and unlimited inclusion dates and quantitative designs. There are also limitations. The search was restricted to children 0-17 years of age; some studies were excluded because they included youth 18 years and older, thus limiting the scope of this review. Similarly, only studies that included "pain" or some variation of the term in their abstracts were included. It is possible that some studies were missed as they did not mention pain or used a different dimension of pain all together (e.g., impact, number of painful joints). Finally, given the heterogeneity of the associations and samples included, the focus was on significance and directionality. Future research may benefit from using effect sizes and meta-analytic techniques to further explore these relationships (McKillop & Banez, 2016), though at present methodology and measurement is so diverse across studies that this may be premature.

The results of this review identify important research directions. Most studies assessed correlational relationships between psychosocial factors and JIA pain. To advance our understanding of factors predictive of JIA pain, there is a need for high quality longitudinal designs. With regards to methodological considerations, participants were largely females with polyarticular or oligoarticular JIA. Future research should seek to explore the pain experience in other populations such as males, other JIA subtypes, and diverse ethnic backgrounds. Furthermore, over 20 studies did not clearly cite or describe their pain measure, 15 relied on a proxy report of pain, and seven did not clarify who the reporter was. While some of these studies may have predated best practice in pediatric pain research, it is recommended that future studies obtain self-reports of pain in children ages 5-6 years old and older (von Baeyer, 2006) and behavioral observations for younger or nonverbal children (von Baeyer & Spagrud, 2007). Assessment of pain in younger or nonverbal children nevertheless remains an important area where further research is required, especially in the context of JIA. These results similarly highlight the inconsistency in measures used to assess psychosocial factors, suggesting the need for greater consensus and psychometric support across measures in this population. Moreover, it is well known that these relationships are more complex than can be expressed through correlations or main effects. An important next step will be to use larger samples and/or open databases that allow for complex analyses that will offer insight into how biopsychosocial factors interact to affect JIA pain (e.g., functioning, rheumatoid factor, cyclic citrullinated peptide antibodies, the child's growth and development, bone and mineral metabolism) (McKillop & Banez, 2016), and how the relationship between psychosocial factors and pain may differ based on subgroups of

individuals (e.g., the 10-15% of children with JIA who experience more chronic JIA) (Shiff et al., 2018; Tesher et al., 2022). Finally, this review has highlighted a restricted set of psychosocial correlates, despite a call nearly 2 decades ago to explore the role of parent/family factors in relation to pain (Palermo & Chambers, 2005), and more recent calls to take a strengths-based approach (Cousins, Kalapurakkel, et al., 2015). As such, in addition to more rigorously assessing the identified associations, there are many factors that were not identified in this review and as such have yet to be explored in relation to JIA pain (e.g., parent factors, temperament/personality dimensions, resilience).

These findings have important clinical implications. Of primary importance is that pain should be assessed comprehensively and regularly in clinics. Stinson and Prescott have outlined several brief and validated pain assessment measures to use with youth diagnosed with JIA (Stinson & Prescott, 2021). The psychosocial factors identified play an important role in the child's pain experience, regardless of whether they cause, are caused by, or are only tangentially related to JIA pain. In line with the interdisciplinary approach to pain management, while pharmacological and physical strategies may be appropriate, psychosocial supports may also be warranted given these results. With regards to psychological interventions, there is preliminary support for their efficacy in reducing pain (and improving other outcomes) in children with JIA (Butler et al., 2022; Cohen et al., 2017). The findings of this review can help refine and design new interventions tailored to address factors associated with worse pain and promote factors associated with reduced pain.

2.6. Conclusions

JIA pain is a complex and pervasive issue. This study has identified psychosocial

factors that tend to be associated with or predictive of JIA pain, including child pain beliefs, internal and external resources (e.g., self-efficacy, social factors, intervention participation), and outcomes such as internalizing symptoms and well-being. Results however should be interpreted with caution given the heterogeneity of findings. These results can help guide the clinical care of children with JIA and can better inform interventions. Moreover, this study has identified several directions for future research, including the use of validated pain measures and larger samples to explore the interactions amongst variables.

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2.8. Conflicts of Interest

The authors declare that they have no competing interests.

2.9. Data Availability Statement

The search string used to identify relevant studies in the current review is available in the supplementary materials. The search string has also been saved at searchrxiv.org.

2.10. References

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2.11. Figures

Figure 2.1. PRISMA Chart Detailing the Search Results and Inclusion Process

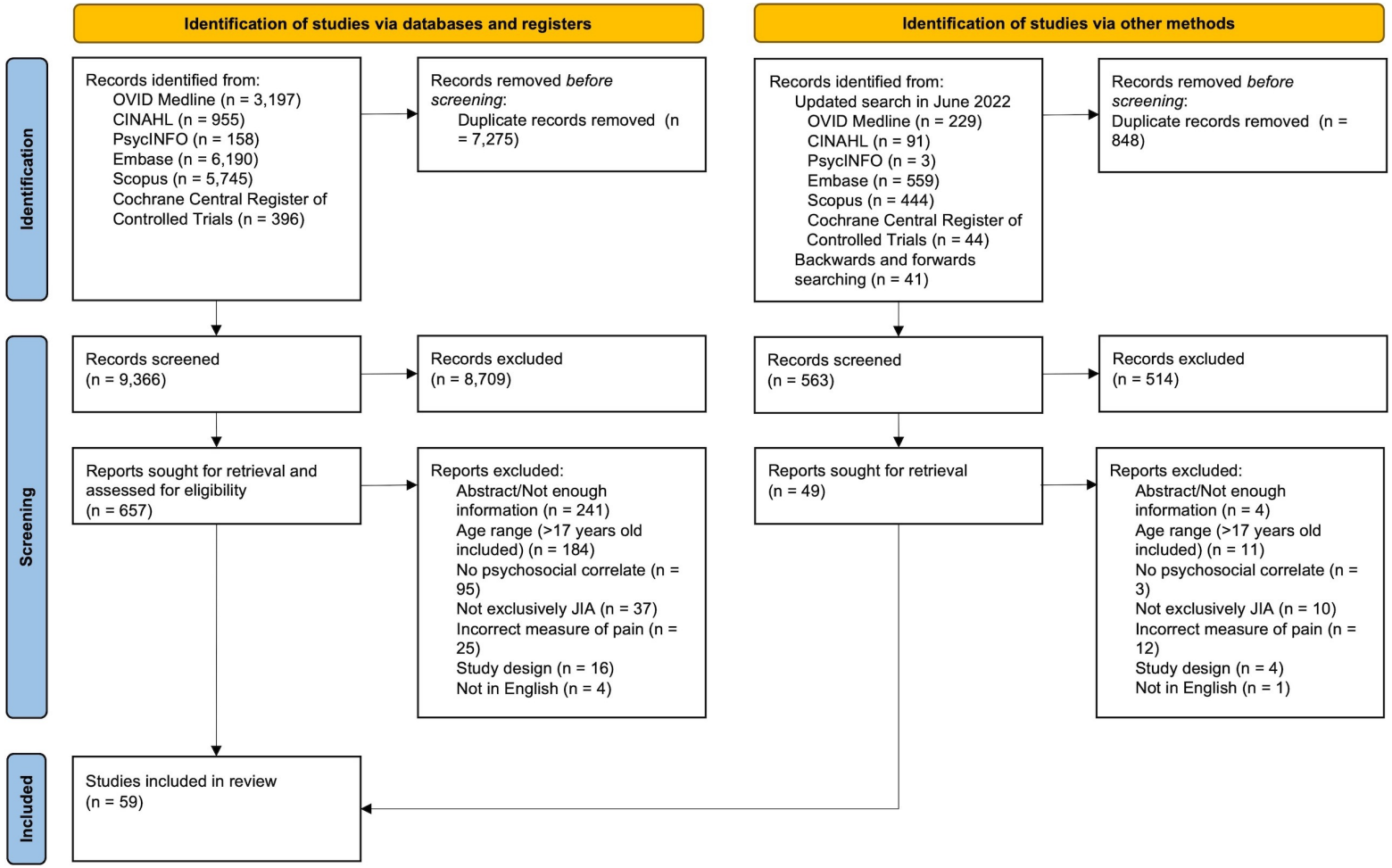
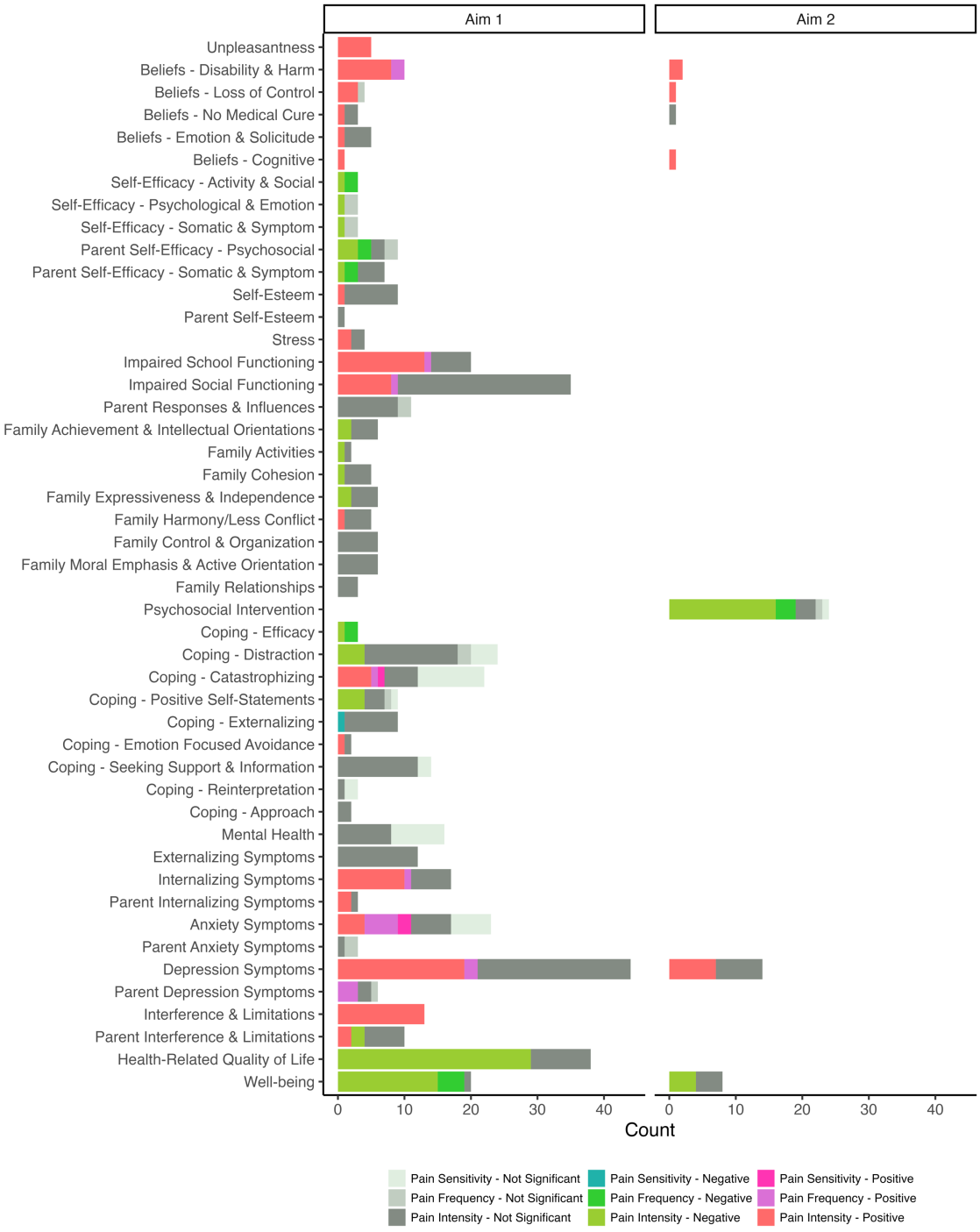


Figure 2.2. Psychosocial Factors Identified and their Associations with Pain Intensity, Frequency, and Sensitivity in Youth with JIA



2.12. Tables

Table 2.1. Measures Used in the 61 Included Studies (N = 516 associations)

Domain	Construct	Measure	Acronym	Frequency
Pain	Intensity	Pediatric Pain Questionnaire (Varni et al., 1987)	PPQ	168
		E-Ouch (Stinson et al., 2008)	--	44
		Faces Pain Scale & Faces Pain Scale Revised (Bieri et al., 1990; Hicks et al., 2001)	FPS(-R)	43
		Childhood Health Assessment Questionnaire (Singh et al., 1994)	CHAQ	40
		Patient-Reported Outcomes Measurement Information System (Cella et al., 2007)	PROMIS	9
		Recalled Pain Inventory (Cleeland, 2009; Stinson et al., 2008)	RPI	4
		Graded Chronic Pain Scale	GCPS	3
		Pain Intensity Scale (Filocamo et al., 2011)	PIS	3
		Child Health Assessment Questionnaire (Billings et al., 1987)	HAQ	2
		SUPERKIDZ (Luca et al., 2017)	--	2
		Juvenile Arthritis Multidimensional Assessment Report (Von Korff et al., 1992)	JAMAR	1
		No reference	--	109
	Frequency	Structured Pain Questionnaire (Mikkelsson et al., 1996)	SPQ	29
		Faces Pain Scale & Faces Pain Scale Revised (Bieri et al., 1990; Hicks et al., 2001)	FPS(-R)	7
		Pain Intensity Scale (Filocamo et al., 2011)	PIS	3
		Pediatric Pain Questionnaire (Varni et al., 1987)	PPQ	4
		No reference	--	4
	Sensitivity	Quantitative Sensory Testing (Meier et al., 2001)	QST	24
		The Cold Pressor Task (Zeltzer et al., 1989)	CPT	16
		No reference	--	1
Primary Appraisals	Pain Unpleasantness	E-Ouch (Stinson et al., 2008)	--	3
		Recalled Pain Inventory (Cleeland, 2009; Stinson et al., 2008)	RPI	2
	Pain Beliefs	Survey of Pain Attitudes (Jensen et al., 1994)	SOPA	28
Secondary Appraisals - Internal	Self-Efficacy	Children's Arthritis Self-Efficacy Scale (Barlow et al., 2001)	CASE	9
	Self-Esteem	Self-Perception Profile for Children and Adolescents (Harter, 1985, 1988)	SPPC/A	3
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	2
	Stress	Patient-Reported Outcomes Measurement Information System (Cella et al., 2007)	PROMIS	2
		Perceived Stress Scale (Cohen et al., 1983)	PSS-10	1
		No reference	--	1
	Physical Appearance	Self-Perception Profile for Children and Adolescents (Harter, 1985, 1988)	SPPC/A	3
	Cognitive Function	Patient-Reported Outcomes Measurement Information System (Cella et al., 2007)	PROMIS	2
Parent Secondary Appraisals - Internal	Self-Efficacy	Parent Arthritis Self-Efficacy Scale (Barlow et al., 2000)	PASE	16
	Self-Esteem	Child Health Questionnaire (Landgraf et al., 1996)	CHQ	1
Secondary Appraisals - External	School Functioning	Pediatric Quality of Life Inventory – Core & Arthritis Modules (Varni, 1998a, 1998b)	PedsQL	8
		Childhood Health Assessment Questionnaire (Singh et al., 1994)	CHAQ	3
		Self-Perception Profile for Children and Adolescents (Harter, 1985, 1988)	SPPC/A	3

Domain	Construct	Measure	Acronym	Frequency
		Child Health Assessment Questionnaire (Billings et al., 1987)	HAQ	2
		Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1985)	RCMAS	1
		No reference	--	3
	Social Functioning	Social Skills Rating System (Gresham & Elliott, 1990)	SSRS	15
		Pediatric Quality of Life Inventory – Core & Arthritis Modules (Varni, 1998a, 1998b)	PedsQL	10
		Child Behavior Checklist (Achenbach & Edelbrock, 1983)	CBCL	3
		Self-Perception Profile for Children and Adolescents (Harter, 1985, 1988)	SPPC/A	3
		Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1985)	RCMAS	2
		Social Support Questionnaire – Revised (Sarason et al., 1987)	SSQR	2
	Parent Pain Responses	West Haven-Yale Multidimensional Pain Inventory (Kerns et al., 1985)	WHYMPI	9
	Family Relationships	Family Environment Scale (Moos & Moos, 1987)	FES	35
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	4
		No citation	--	2
	Interventions[a]	Pain Management Intervention (Lavigne et al., 1992)	--	9
		Cognitive Behavioral Therapy Intervention (Walco et al., 1992)	CBT	1
		Cognitive Behavioral Therapy Group Intervention (Lomholt et al., 2015)	CBT	10
		Peer-Led Intervention (Stinson et al., 2016)	iPeer2Peer	1
		Self-Management Intervention (Stinson et al., 2020)	TTC	3
Coping	Coping	Pain Coping Questionnaire[b] (Thastum et al., 1998)	PCQ	76
		Pain Catastrophizing Scale for Children (Crombez et al., 2003)	PCS-C	9
		Coping Strategies Questionnaire for Children (Gil et al., 1991; Rosenstiel & Keefe, 1983)	CSQ-C	1
		No reference	--	2
Outcomes	General Mental Health	Pediatric Symptom Checklist (Pagano et al., 2000)	PSC	8
		Child Behavior Checklist (Achenbach & Edelbrock, 1983)	CBCL	5
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	2
		Rutter Parental Screening Questionnaire (Graham & Rutter, 1968)	--	1
	Externalizing Symptoms	Child Behavior Checklist (Achenbach & Edelbrock, 1983)	CBCL	5
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	4
		Self-Perception Profile for Children and Adolescents (Harter, 1985, 1988)	SPPC/A	3
	Internalizing Symptoms	Pediatric Quality of Life Inventory – Core & Arthritis Modules (Varni, 1998a, 1998b)	PedsQL	8
		Child Behavior Checklist (Achenbach & Edelbrock, 1983)	CBCL	3
		Child Vulnerability Scale (Forsyth et al., 1996)	CVS	1
		Patient Health Questionnaire (Kroenke et al., 2009)	PHQ-4	1
		No reference	--	4
	Anxiety Symptoms	State-Trait Anxiety Inventory for Children (Spielberger et al., 1983)	STAI-C	12
		Revised Children's Manifest Anxiety Scale (Reynolds & Richmond,1985)	RCMAS	4
		Pediatric Quality of Life Inventory – Core & Arthritis Modules (Varni, 1998a, 1998b)	PedsQL	2
		Patient-Reported Outcomes Measurement Information System (Cella et al., 2007)	PROMIS	2

Domain	Construct	Measure	Acronym	Frequency
		Trauma Symptom Checklist for Children (Briere, 1996)	TSC-C	2
		Screen for Child Anxiety Related Disorders (Birmaher et al., 1995)	SCARED	1
	Mood/Depression Symptoms	Children's Depression Inventory (Kovacs, 1985)	CDI	12
		Mood and Feelings Questionnaire (Angold et al., 1995)	MFQ	12
		Positive and Negative Affect Scale for Children (Laurent et al., 1999)	PANAS-C	8
		Child Behavior Checklist (Achenbach & Edelbrock, 1983)	CBCL	6
		Facial Affective Scale (McGrath et al., 1996)	FAS	4
		Patient-Reported Outcomes Measurement Information System (Cella et al., 2007)	PROMIS	3
		Children's Emotion Management Scale (Zeman et al., 2001)	--	2
		Differential Emotions Scale – IV (Kotsch et al., 1982)	DES-IV	2
		Emotion Regulation Scale (Shields & Cicchetti, 1997)	--	2
		Hopelessness Scale for Children (Kazdin et al., 1983)	--	2
		Revised Child Anxiety and Depression Scale (Chorpita et al., 2000)	RCADS	2
		Trauma Symptom Checklist for Children (Briere, 1996)	TSC-C	2
		Centre for Epidemiological Studies Depression Scale for Children (Faulstich et al., 1986)	CES-DC	1
	Pain Interference/ Limitations	Recalled Pain Inventory (Cleeland, 2009; Stinson et al., 2008)	RPI	6
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	4
		E-Ouch (Stinson et al., 2008)	--	3
	Health-Related Quality of Life (HRQOL)	Pediatric Quality of Life Inventory – Core & Arthritis Modules (Varni, 1998a, 1998b)	PedsQL	22
		Juvenile Arthritis Quality of Life Questionnaire (Duffy et al., 1997)	JAQQ	7
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	5
		Quality of My Life Scale (Feldman et al., 2000)	QoML	3
		Clinically Derived Global Score for Psychosocial Functioning (Shaffer et al., 1983)	CGAS	1
	Well-being	Global Assessment of Well-being Visual Analogue Scale	--	20
		Childhood Health Assessment Questionnaire (Singh et al., 1994)	CHAQ	7
		World Health Organization Well-Being Index (World Health Organization, 1998)	WHO-5	1
Parent Outcomes	General Mental Health	Lanyon Psychological Screening Inventory (Lanyon, 1978)	--	2
		Self-Reporting Questionnaire (Harding et al., 1980)	SRQ-20	1
	Anxiety Symptoms	Hospital Anxiety and Depression Scale (Zigmond & Snaith,1983)	HADS	3
	Mood/Depression Symptoms	Hospital Anxiety and Depression Scale (Zigmond & Snaith,1983)	HADS	3
		Beck Depression Inventory (Beck et al., 1996)	BDI	3
	Pain Interference/ Limitations	Revised Hassles and Uplifts Scale (DeLongis et al., 1988)	--	5
		Child Health Questionnaire (Landgraf et al., 1996)	CHQ	4
		Caregiver Burden Scale (Medeiros et al., 1998)	CBS	1

Note. [a]See supplementary material for results. [b]Some studies used a preliminary version of this scale.

Table 2.2. Critical Appraisal Results for Analytical Cross Sectional Studies

Author & Year	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	%
Amine 2009	Y	Y	U	Y	N	N	U	Y	50%
Anthony 2011‡	Y	Y	Y	Y	Y	Y	Y	Y	100%
Armbrust 2016	Y	Y	N	Y	Y	Y	U	Y	75%
Baildam 1995	Y	Y	Y	Y	N	N	Y	N	63%
Baloueff 1996	Y	Y	Y	Y	Y	Y	Y	Y	100%
Barlow 2000	Y	N	Y	Y	N	N	U	Y	50%
Barlow 2001	Y	N	Y	Y	N	N	U	Y	50%
Barlow 2002	N	N	Y	Y	N	N	Y	Y	50%
Bromberg 2009‡	Y	Y	Y	Y	N	N	Y	Y	75%
Bromberg 2012‡	Y	Y	Y	Y	Y	Y	Y	Y	100%
Bruns 2008	Y	Y	Y	Y	N	N	U	Y	63%
Cornelissen 2014	Y	Y	Y	Y	N	N	Y	Y	75%
Dimitrijevic Carlsson 2019	Y	Y	N	Y	N	N	Y	Y	63%
Doherty 1993	Y	Y	Y	Y	N	N	Y	Y	75%
El-Najjar 2014	Y	Y	Y	Y	N	N	U	Y	63%
Hagglund 1995	Y	Y	Y	Y	Y	Y	Y	Y	100%
Hanns 2018-2‡‡	Y	Y	Y	Y	Y	Y	U	Y	88%
Jaworski 1992	Y	N	Y	Y	N	N	Y	Y	63%
Klotsche 2014	Y	Y	U	Y	Y	Y	U	Y	75%
Kovalchuk 2017	N	N	U	Y	N	N	Y	Y	38%
Kovalchuk 2018	N	N	Y	Y	N	N	U	Y	38%
Listing 2018	Y	Y	Y	Y	U	U	U	Y	63%
Lomholt 2013††	Y	Y	Y	Y	N	N	Y	Y	75%
Luca 2017	Y	N	Y	Y	N	N	Y	Y	63%
Mahler 2017	Y	Y	U	Y	N	N	U	Y	50%
Margetić 2005	U	N	Y	Y	N	N	Y	Y	50%
Oen 2009§	Y	Y	U	Y	Y	Y	U	Y	75%
Oen 2021§	Y	N	U	Y	Y	Y	U	Y	63%
Ross 1993	Y	Y	Y	Y	Y	Y	Y	Y	100%
Sällfors 2004	Y	Y	Y	Y	N	N	N	Y	63%
Schanberg 2003‡	Y	Y	Y	Y	N	N	Y	Y	75%
Schanberg 2005‡	Y	Y	Y	Y	Y	Y	Y	Y	100%
Selvaag 2003	N	Y	N	Y	Y	Y	N	Y	63%
Selvaag 2005	N	N	Y	Y	N	N	U	Y	38%
Shelepina 2011	N	N	Y	U	N	N	U	Y	25%
Stinson 2006-1†	Y	Y	Y	Y	N	N	Y	Y	75%
Stinson 2006-2	Y	Y	Y	Y	N	N	Y	Y	75%
Tarakci 2011	Y	Y	Y	Y	N	N	Y	Y	75%
Tarkiainen 2019	Y	U	Y	Y	Y	Y	U	Y	75%
Thastum 1997	Y	Y	Y	Y	N	N	Y	Y	75%
Thastum 1998	N	N	Y	Y	N	N	Y	Y	50%
Thastum 2005††	Y	Y	Y	Y	Y	Y	Y	Y	100%
Thompson 1987	Y	U	Y	Y	N	N	Y	Y	63%
Tupper 2012	Y	Y	Y	Y	N	N	Y	Y	75%
Tupper 2013†	U	Y	Y	Y	Y	Y	Y	Y	88%

Author & Year	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	%
Upadhyay 2021	N	N	Y	Y	N	N	Y	Y	50%
Vandvik 1990	Y	N	Y	Y	N	N	Y	Y	63%
Vuorimaa 2008§§	Y	Y	Y	Y	N	N	U	U	50%
Vuorimaa 2009§§	Y	Y	Y	Y	N	N	U	U	50%
Vuorimaa 2011§§	Y	Y	Y	Y	N	N	Y	Y	75%
Yan 2020	Y	Y	Y	Y	N	N	Y	Y	75%
%	80%	69%	82%	98%	29%	29%	61%	94%	

Note. JBI critical appraisal for quasi-experimental studies: Q1 = Were the criteria for inclusion in the sample clearly defined? Q2 = Were the study subjects and the setting described in detail? Q3 = Was the exposure measured in a valid and reliable way? Q4 = Were objective, standard criteria used for measurement of the condition? Q5 = Were confounding factors identified? Q6 = Were strategies to deal with confounding factors stated? Q7 = Were the outcomes measured in a valid and reliable way? Q8 = Was appropriate statistical analysis used?
‡, ‡‡, †, ††, §, §§ Studies with overlapping datasets. Y = Yes; N = No; U = Unclear.

Table 2.3. Critical Appraisal Results for Analytical Cohort Studies

Author & Year	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	%
Connelly 2012	N/A	N/A	Y	N	N	N	Y	Y	Y	Y	Y	67%
Hanns 2018-1‡‡	N/A	N/A	Y	Y	Y	N	U	Y	N	N	Y	56%
Hoff 2006	N/A	N/A	Y	Y	Y	N	Y	Y	Y	Y	Y	89%
Rashid 2018‡‡	U	N/A	U	Y	Y	N	U	Y	N	N	Y	40%
Thastum 2011††	N/A	N/A	Y	Y	Y	N	Y	Y	Y	Y	Y	89%
%	0%	N/A	80%	80%	80%	0%	60%	100%	60%	60%	100%	

Note. JBI critical appraisal for cohort studies: Q1 = Were the two groups similar and recruited from the same population? Q2 = Were the exposures measured similarly to assign people to both exposed and unexposed groups? Q3 = Was the exposure measured in a valid and reliable way? Q4 = Were confounding factors identified? Q5 = Were strategies to deal with confounding factors stated? Q6 = Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)? Q7 = Were the outcomes measured in a valid and reliable way? Q8 = Was the follow up time reported and sufficient to be long enough for outcomes to occur? Q9 = Was follow up complete, and if not, were the reasons to loss to follow up described and explored? Q10 = Were strategies to address incomplete follow up utilized? Q11 = Was appropriate statistical analysis used?

‡, ‡‡, ‡‡‡, †, ††, §, §§ Studies with overlapping datasets. Y = Yes; N = No; U = Unclear; N/A = Not applicable

Table 2.4. Study Characteristics and Results

Author, Year, Publication Type	Sample Size(s)	Age(s) x or Mdn (Range)	% Girls	% JIA Type	Pain: Construct (Reporter) – Measure	Psychosocial Factor(s): Construct (Reporter) – Measure	Main Findings: Analysis - Result
Amine 2009 Article	80 C 80 P	x=11 (6-17) --	59 --	Po:32; O:43; S:26	PI (--) – VAS CHAQ	HRQoL (--) – JAQQ	Corr – Lower well-being was significantly associated with greater PI
Anthony 2011‡ Article	51 C 51 P	x=12 (8-16) --	61 96	Po:63; E:8; S:24; Ps:5	PI (C) – VAS PPQ (current)	P Depression symptoms (P) – BDI Vulnerability (P) – CVS P Hassles & Uplifts Intensity & Frequency (P) – Hassles and Uplifts Scale	Corr – Parent depression symptoms, child vulnerability, parent identified daily hassles (intensity and frequency), and parent identified daily uplifts (intensity) were not significantly associated with PI Corr & HR controlling for age, gender, active joint count, and disease severity – More parent reported daily uplifts were significantly associated with greater PI
Armbrust 2016 Article	80 C	Mdn=10 (8-13)	65	Po:35; O:45; E:4; S:11; Ps:5	PI (C) – VAS	School Attendance (C) – -- (yes/no)	Corr & LoR controlling for age, disease activity, medications, disability, and fatigue – Lower school attendance was significantly associated with greater PI
Baildam 1995 Article	29 C 29 P	x=11 (7-16) --	48 --	Po:48 O:52	PI (C) – VAS (worst past week)	Mental Health (P) – Rutter Parental Screening Questionnaire high (≥13) / low (<13)	Mann-Whitney U Test – Children with higher and lower Rutter scores did not significantly differ in PI
Baloueff 1996 Thesis	60 C	x=12 (8-17)	73	Po:33; O:57; S:10	PI (C) – VAS PPQ (average of current and past week) mean and high (>2.5cm)/ low (<2.5cm)	Behavioral Conduct, Self-Esteem, Scholastic Competence, Appearance, & Social Acceptance (C) – SPPC/A Assertion, Cooperation, Empathy, Self-Control, & Social Skills (C) – SSRS	Corr, MR & one-way ANOVA – Behavioral conduct, physical appearance, scholastic competence, social acceptance, self-esteem, assertion, cooperation, empathy, self-control, and social skills were not significantly associated with PI, nor did they significantly differ between high and low pain groups
Barlow 2000 Article	116 C 178 P	* (7-17) *	64 65	--	PI (Mother, Father, & C) – VAS (current)	P Psychosocial & Symptom Self-Efficacy (Mother & Father) – PASE	Corr – Greater mother's psychosocial self-efficacy was significantly associated with lower mother and child reports of PI Greater father's psychosocial self-efficacy was significantly associated with lower PI as reported by the child but not themselves Greater mother's symptom self-efficacy was significantly associated with lower PI as reported by themselves but not their child Father's symptom self-efficacy was not significantly

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							associated with their own and child reports of PI
Barlow 2001 Article	89 C 151 P	x=12 (7-17) *	62 58	--	PI (C) – VAS (current)	Activity, Emotion, & Symptom Self-Efficacy (C) – CASE	Corr – Greater child activity, emotion, and symptom self-efficacy were significantly associated with lower PI
Barlow 2002 Article	30 C 30 P	x=11 (--) x=38 (--)	67 100	Po:26; O:61; S:13	PI (C) – VAS PPQ (current)	P Depression & Anxiety symptoms (Mother) – HADS P Psychosocial & Symptom Self-Efficacy (Mother) – PASE	Corr – Maternal depression and anxiety symptoms, and psychosocial and symptom self-efficacy were not significantly associated with PI
Bromberg 2009‡ Thesis	51 C	x=12 (8-16)	65	Po:100	PI (C) – VAS PPQ (1x/day for 2 mos)	Coping Efficacy (C) – CSQ-C assessed 1x/day for 2 mos	HR controlling for age, disease severity, and sleep quality – Greater coping efficacy was significantly associated with lower PI
Bromberg 2012‡ Article	51 C 51 P	x=12(8-16) --	65 --	Po:100	PI (C) – VAS PPQ (1x/day for 2 mos)	Mood (C) –VAS FAS assessed 1x/day for 2 mos	Hierarchical MLM controlling for age, disease severity, and between and within child sleep quality – Higher daily reported mood (within subjects), but not mean mood (between subjects), was significantly associated with lower PI that day
Bruns 2008 Article	70 C 70 P	x=10 (0-16) x=37 (--)	67 91	Po:63; O:16; S:21	PI (--) – VAS (past week)	P Caregiver Burden (P) – CBS P Mental Health (P) – SRQ-20	Corr – Caregiver burden and parent mental health were not significantly associated with PI
Connelly 2012 Article	43 C 43 P	x=13 (8-17) --	86 90	--	PI (C) – electronic VAS (3x/day for 28 days)	Variability in positive & negative mood, ability to adaptively attenuate negative emotions, & ability to upregulate positive emotions (C) – PANAS-C assessed 3x/day for 28 days Emotion Regulation (P) – The Emotion Regulation Scale (baseline) Emotion Regulation (C) – Children's Emotion Management Scale (baseline)	Corr and LMM – Greater variability in positive and negative emotions were significantly associated with and predictive of greater PI A child's ability to adaptively attenuate negative emotions was associated with, but not predictive of, lower PI A child's ability to adaptively upregulate positive emotions to average levels following a drop was not significantly associated with but was predictive of lower PI Parent-reported and self-reported emotion regulation at baseline was not significantly associated with or predictive of PI
Cornelissen 2014 Article	60 C	Mdn=13 (7-17)	73	Po:48; Ps:22	PS (C) – Cold Detection, Cold Pain, Warm	Catastrophizing (C) – PCS-C Mental Health (C) – PSC Trait Anxiety symptoms (C) – STAI-C	LR – Catastrophizing and mental health were not significantly associated with PS as measured by the child's cold detection,

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					Detection, Warm Pain, Mechanical Detection, Mechanical Pain, Vibration Detection, & Pressure Pain Thresholds		cold pain, warm detection, heat pain, mechanical detection, mechanical pain, vibration detection, or pressure pain thresholds Greater trait anxiety symptoms were significantly associated with greater PS as measured by the child's lower mechanical detection and mechanical pain thresholds, but not by their cold detection, cold pain, warm detection, heat pain, vibration detection, or pressure pain thresholds
Dimitrijevic Carlsson 2019 Article	45 C	Mdn=12 (6-16)	73	Po:33; O:44	PI for temporo-mandibular joints (C) – GCPS (average of current, past week, and worst in the past week)	Catastrophizing (C) – PCS-C Distress (C) – PHQ-4 Stress (C) - PSS	Corr – Greater catastrophizing, distress, and perceived stress were significantly associated with greater temporomandibular joint PI
Doherty 1993 Article	20 C 20 P	x=11 (8-15) --	55 100	Po:15; O:55; S:30	PI (C & P) – VAS Child HAQ	School absences (P) – Child HAQ	Corr – More school absences were significantly associated with greater parent, but not child, reported PI
El-Najjar 2014 Article	54 C 54 P	x=11 (6-15) --	67 --	Po:28; O:39; E:11; S:22	PI (--) – VAS	Depression symptoms (C) – CES-DC	Corr – More depression symptoms were significantly associated with greater PI
Hagglund 1995 Article	60 C	x=11 (7-17)	62	Po:35; O:55; S:10	PI (C) – VAS (past month)	Social Support (C) – SSQR Hopelessness (C) – Hopelessness Scale for Children Sadness (C) – DES-IV	Corr and HR controlling for age, gender, socioeconomic status, disease duration, and articular severity – Social support, hopelessness, and sadness were not significantly associated with PI
Hanns 2018-1‡‡ Thesis	219 C	x=13 (11-16)	57	Po:22; O:35; E:13; S:6; Ps:13; U:11	PI (C) – VAS (baseline, 6, and 12 mos) mean and high (7.4)/low (0.4)	Depression symptoms (C) – MFQ at baseline, 6, and 12 mos average and low (2 points)/high (31 points)	LMM controlling for active/limited joint count and disability – More depression symptoms at baseline significantly predicted greater PI over time, and greater PI at baseline predicted more depression symptoms over time Mann Whitney U-Test – More depression symptoms at baseline significantly predicted greater PI over 12 mos, and higher PI at baseline significantly predicted greater depression symptoms over 12 mos

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Hanns 2018-2‡‡ Thesis	102 C	Mdn=13 (11-16)	57	Po:30; O:52; E:18	PI (C) – VAS	Depression symptoms (C) – MFQ mean and high (≥27)/low (<27)	Corr and MR controlling for age, medications, diagnosis, gender – Greater depression symptoms were significantly associated with greater PI Mann Whitney U-Test – Children with high and low depression symptoms did not significantly differ in PI
Hoff 2006 Article	63 C 63 P	x=12 (8-17) x=40 (--)	81 --	Po:29; O:41; E:8; S:5; U:18	PI (C & P) – FPS (last few days at baseline, 6, and 12 mos)	Depression symptoms (C) – RCADS at baseline	LMM controlling for age, gender, income, and disease severity – Greater depressive symptoms at baseline significantly predicted child reported, but not parent reported, PI over time when PI was low at baseline
Jaworski 1992 Thesis	30 C 30 P	x=11 (6-17) --	73 --	Po:73; O:27	PI (C & P) – VAS PPQ	Depression symptoms (C) – CDI Depression symptoms (P) – CBCL P Punishing, Distracting, & Solicitous Pain Responses (P) – WHYMPI	Corr – Child reported depression symptoms were significantly associated with greater parent reported PI for the whole sample, and 12–17-year-olds, but not 6–11-year-olds Parent reported depression symptoms, punishing, distracting, and solicitous pain responses were not significantly associated with child or parent reported PI in the whole sample, 6–11-year-olds, or 12–17-year-olds
Klotsche 2014 Article	61 C 61 P	x=11 (3-17) --	66 --	Po:67; O:21; E:5; S:2; Ps:3; U:2	PI (P) – VAS CHAQ (9 timepoints: baseline, 1 mos, 2 mos, 3 mos, 4 mos, 5 mos, 6 mos, 9 mos, and 12 mos)	HRQoL Total, Emotional Functioning, School Functioning, & Social Functioning (--) – PedsQL (9 timepoints)	Univariate and Multivariate Reg controlling for disease activity, joints, stiffness, disability, & comorbidities – Lower well-being at baseline was significantly associated with greater PI at baseline Latent Growth Curve Mixture Modelling – A rapid increase in well-being across the first 4 timepoints was significantly associated with lower PI at baseline Linear Reg – Lower PI across timepoints significantly predicted better total well-being across time Lower PI across timepoints 1-7, but not 8 and 9 significantly predicted better emotional functioning across time Lower PI across timepoints 1-8, but not 9, significantly predicted better school and social functioning across time
Kovalchuk 2017 Article	55 C 55 P	* (6-17) --	53 --	Po:53; O:47	PI (P) – VAS CHAQ	HRQoL Psychosocial (P) – CHQ	Corr – Psychosocial well-being was not significantly associated with PI

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Kovalchuk 2018 Article	60 C 60 P 60 HCP	x=13 (5-17) -- --	48 100 --	Po:48; O:52	PI (C & P) – VAS (current)	HRQoL Behavior, Global Behavior, Self-Esteem, Family Cohesion, Family Activities, Mental Health, Time Impact, Emotional Impact, Emotional Role Limitations, Physical Role Limitations, & Psychosocial (P) – CHQ Well-being (P, HCP, & C) – Global Assessment VAS	Corr – Behavior, global behavior, self-esteem, family cohesion, mental health, and psychosocial summary scores were not significantly associated with parent or child reported PI Reduced engagement in family activities and greater impact on parents' time and emotions were significantly associated with parent (but not child) reported PI More emotional and physical role limitations in parents, and lower parent, child, and healthcare provider global assessments of well-being were significantly associated with greater parent and child reported PI
Listing 2018 Article	953 C 953 P	x=8 (--) --	67 --	Po:28; O:46; E:11; S:4; Ps:4; U:8	PI (P) – NRS	HRQoL (--) – PedsQL	LR – Greater well-being at baseline was significantly associated with lower PI at baseline Stepwise Reg – Greater PI at baseline significantly predicted lower well-being at 36 mos
Lomholt 2013†† Article	41 C	x=14 (8-17)	71	Po:44; O:24; E:5; S:22; Ps:5	PF (C) – FPS-R (2x/day for 2 weeks) pain/pain-free groups	Coping Behavioral Distraction, Cognitive Distraction, Catastrophizing, & Positive Self-Statement – PCQ Pain Beliefs (Control, Disability, & Harm (C) PA	Mann Whitney U-Test – Behavioral distraction, cognitive distraction, the use of positive self-statements, and beliefs of control did not significantly differ between the pain and pain-free groups Greater catastrophizing, beliefs of harm, and beliefs of disability were significantly higher amongst the pain group compared to the pain-free group
Luca 2017 Article	17 C 17 P	* (4-7) --	* --	*	PI (C) – SUPERKIDZ (current and past week)	HRQoL (C – PedsQL Arthritis	Corr – Child reported and parent reported well-being were not significantly associated with current and past week PI, respectively
Mahler 2017 Abstract	51 C 51 P	Mdn=13 (6-16) --	76 --	Po:27; O:37; E:4; S:10; Ps:11; U:11	PI (--) – VAS JAMAR (past week)	Well-being (--) – WHO-5	Corr – Child well-being was not significantly associated with PI
Margetić 2005 Article	36 C	x=13 (8-16)	61	--	PI (C) – VAS (current)	Anxiety and Depression symptoms (C) – TSC-C	Corr and Reg – Greater depression, but not anxiety symptoms, were significantly associated with greater PI

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Oen 2009[§] Article	356 C 356 P	Mdn=9 (0-17) --	66 --	Po:24; O:41; E:10; S:7; Ps:7; U:12	PI (--) – VAS (baseline and 6 mos)	Well-being (--) – Global Assessment VAS assessed at baseline and 6 mos later HRQoL (--) – JAQQ assessed at baseline and 6 mos later	Corr – Lower well-being (VAS & JAQQ) at baseline was significantly associated with greater PI at baseline Univariate & Multivariate Reg controlling for number of joints affected, baseline JAQQ, and time since diagnosis – Greater PI at baseline predicted lower well-being (JAQQ) at 6 mos
Oen 2021[§] Article	561 C	Mdn=10 (--) --	65 --	Po:23; O:41; E:15; S:5; Ps:6; U:10	PI (C) – VAS (past week at diagnosis, 3-9 mos post, and during flares)	HRQoL (C) – JAQQ psychosocial assessed at diagnosis, 3-9 mos post, and during flares HRQoL (C) – QoML assessed at diagnosis, 3-9 mos post, and during flares	Corr in SEM – Greater PI at diagnosis and 3-9 mos post diagnosis were significantly associated with lower well-being (JAQQ & QoML) at diagnosis and 3-9 mos post diagnosis, respectively Greater PI during flares was significantly associated with lower well-being (QoML but not JAQQ) during flares
Rashid 2018[‡‡] Article	851 C 851 P	Mdn=8 (1-16) --	66 --	Po:29; O:48; E:5; S:6; Ps:8; U:3	PI (--) – VAS PPQ (baseline, 6 mos, and annually up to 60 months) average and 3 pain trajectories: consistently low/ improved/consistently high	Well-being (P) – Global Assessment VAS assessed at baseline, 6 mos, and annually Depression symptoms (--) MFQ assessed at baseline, 6 mos, and annually	Corr – Lower well-being and greater depression symptoms at baseline were significantly associated with greater PI at baseline and less change in PI over time Greater PI at baseline was significantly associated with less change in well-being within 6 mos Change in PI within 12 mos was not significantly associated with change in well-being over 12 mos Multinomial LoR – Well-being was significantly lower in the consistently high and improved pain groups compared to the consistently low pain group, and well-being significantly increased over 6 mos in the improved pain group compared to the consistently low pain group. No other differences emerged. Depression symptoms did not significantly differ across groups.
Ross 1993 Article	56 C 56 P	x=12 (7-17) --	73 --	Po:59; O:27; E:5; S:9	PI (C) – VAS (3x/day for 28 days) mean	Behavior (P) – CBCL Depression symptoms (C) – CDI Anxiety symptoms (C) – STAI-C Distress (C) – CDI and STAI-C P Maternal Distress (P) – Lanyon Psychological Screening Inventory P Family Harmony (P) – FES	Corr and HR controlling for range of motion, disease activity, joint activity, stiffness, number of joints affected, and other measured variables – Behavior was not significantly associated with PI Greater anxiety symptoms, child distress, and maternal distress were significantly associated with greater PI Greater depression symptoms were significantly associated with but not predictive of greater PI

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							Greater family harmony was not associated with but predicted greater PI
Sällfors 2004 Article	125 C	x=14 (10-17)	66	Po:46; O:53; S:1	PI (C) – VAS (usual) PI (C) – NRS PIS (4x/day for 1 week) PF (C) – PIS (pain free days)	Well-being (C) – VAS CHAQ Absences from school (C) – CHAQ	Corr and Stepwise Reg – Lower well-being was significantly associated with greater PI (VAS & PIS) and PF Corr – More school absences were significantly associated with greater PI (VAS & PIS) and PF
Schanberg 2003‡ Article	41 C	x=12 (8-17)	59	Po:59; E:7; S:27; Ps:7	PI (C) – VAS PPQ 1x/day at baseline, follow up, and for 2 mos) PF (C) – VAS PPQ (percentage of pain days)	Depression symptoms (C) – CDI assessed at baseline Anxiety symptoms, Social Concerns, Physiologic Anxiety, & Worry (C) – RCMAS assessed at baseline	Corr – Depression symptoms were not significantly associated with PI Greater physiologic anxiety was significantly associated with greater PI and PF Greater total anxiety symptoms, social concerns, and worry were significantly associated with greater PF
Schanberg 2005‡ Article	51 C	x=12 (8-17)	65	Po:63; E:8; S:24; Ps:6	PI (C) – VAS PPQ (1x/day at baseline, follow up, and for 2 mos)	Stress (C) – Daily Events Inventory assessed daily for 2 months Mood (C) – FAS assessed daily for 2 mos Social & School Activity Reduction (C) – RCMAS assessed daily for 2 mos	Longitudinal Mixed Effects Models – Greater same day stress and lower same day mood were significantly associated with greater same day PI LMM controlling for disability index, global assessment, sex, age, disease onset, stiffness, fatigue, mood, and stress – Social, but not school, activity reduction was significantly associated with greater PI
Selvaag 2003 Article	116 C 116 P	x=9 (4-17) x=38 (--)	60 *	Po:35; O:51; E:3; S:4; Ps:6; U:1	PI (P) – VAS	HRQoL psychosocial (--) – CHQ	Corr – Psychosocial well-being was not significantly associated with PI
Selvaag 2005 Article	-- 197 P	x=7 (1-16) --	61 --	Po:30; O:56; E:4; S:7; Ps:3	PI (P) – VAS	Well-being (P) – Global Assessment VAS	Corr – Lower well-being was significantly associated with greater PI
Shelepina 2011 Abstract	99 C	-- (14-17)	73	Po:49; O:16; E:15;	PI (P) – VAS	Schooling location (C) - -- school/home	-- – Children who were taught at home without medical indication reported significantly higher PI compared to those taught at school

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				S:19			
Stinson 2006-1† Thesis	76 C	x=13 (9-17)	78	Po:49; O:15; E:11; S:13; Ps:11; U:3	PI (C) – E-ouch VAS (3x/day for 14 days) PI (C) – NRS RPI (past week)	Pain Unpleasantness & Pain Interference (C) – E-ouch Pain Unpleasantness, Pain Interference Total, Pain Interference Mood, Pain Interference Relationships, Pain Interference Schoolwork, & Pain Interference Sleep (C) – RPI Coping via Approach, Distraction, & Emotion-Focused Avoidance (C) – PCQ HRQoL Total & Psychosocial, (C) – PedsQL HRQoL Arthritis Total, Worry, & Communication (C) – PedsQL Rheumatology	Corr – Greater pain unpleasantness (E-Ouch and RPI) was significantly associated with greater PI (E-Ouch and RPI) across both weeks Greater pain interference (E-Ouch and RPI total, mood, relationships, schoolwork, sleep) was significantly associated with greater PI (E-Ouch and RPI) Approach coping and distraction coping were not significantly associated with PI (E-Ouch) on either week Greater emotion focused avoidance coping was significantly associated with greater PI (E-Ouch) on week 2 but not week 1 Lower total well-being, lower psychosocial well-being, lower total arthritis well-being, and more worry were significantly associated with greater PI (E-Ouch) Communication was not significantly associated with PI (E-Ouch)
Stinson 2006-2 Thesis	36 C	x=13 (8-17)	67	Po:28; O:39; E:11; S:11; Ps:6; U:6	PI (C) – E-ouch VAS (3x/day for 31 days; at day 7 had joint injections) PI (C) – NRS RPI (past week)	Pain Unpleasantness & Pain Interference (C) – E-ouch Pain Unpleasantness & Pain Interference (C) – RPI Coping via Approach, Distraction, & Emotion-Focused Avoidance (C) – PCQ HRQoL Total & Psychosocial (C) – PedsQL HRQoL Arthritis Total, Worry, & Communication (C) – PedsQL Rheumatology	Corr – Greater pain unpleasantness (E-Ouch and RPI) and pain interference (E-Ouch and RPI) were significantly associated with greater PI (E-Ouch and RPI) Approach coping, avoidance coping, and emotion-focused avoidance coping were not significantly associated with PI (E-Ouch) Lower total well-being and total arthritis well-being were significantly associated with greater PI (E-Ouch) Psychosocial well-being, worry, and communication were not significantly associated with PI (E-Ouch)
Tarakci 2011 Article	52 C	x=12 (8-17)	63	Po:52; O:29; E:8; S:4; Ps:6; U:2	PI (C) – VAS CHAQ (past week)	Depression symptoms (C) – CDI Anxiety symptoms (C) - SCARED Well-being (C) – CHAQ	Corr – Depression and anxiety symptoms were not significantly associated with PI Lower well-being was significantly associated with greater PI

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Tarkiainen 2019 Article	-- 60 P	* (4-14) --	65 --	Po:85; E:13; Ps:2	PI (--) – VAS (8x over 1 year)	HRQoL psychosocial (C) – CHQ assessed 8 times throughout 1 year	Univariate LMM – Greater PI was significantly associated with less improvement psychosocial well-being over time
Thastum 1997 Article	15 C 15 P	x=12 (9-15) --	73 --	Po:20; O:80	PI (C) – VAS (current) PS (C) – Tolerance/time hand submerged PS (C) – Threshold/time moved to button	Coping via Catastrophizing, Distraction, & Reinterpretation (C) – preliminary PCQ	Reg – Greater catastrophizing was significantly associated with greater PI and lower pain threshold (PS), but not pain tolerance (PS) Distraction and reinterpretation were not significantly associated with PI or PS (tolerance or threshold)
Thastum 1998 Article	40 C	* (8-17)	58	--	PI (C) – VAS PPQ (current, average, worst) high (modest disease activity and pain)/low (disease activity but few pain complaints) PS (C) – Tolerance/ time hand submerged	Coping via Behavioral Distraction, Cognitive Distraction, Information Seeking, Seeking Social Support, Externalizing, Catastrophizing, & Positive Self-Statements (C) – PCQ	Corr and T-test – Greater behavioral distraction was significantly associated with lower PI (average, current, worst) but not experimental PI or PS. Behavioral distraction was significantly higher in the high pain group Cognitive distraction, information seeking, and seeking social support were not significantly associated with PI nor did it differ between high and low pain groups Greater externalizing was significantly associated with lower PS (i.e., higher tolerance); however, was not significantly associated with average, current, or worst PI (current and experimental) and did not differ between high and low pain groups Greater catastrophizing was significantly associated with greater experimental PI; however, was not significantly associated with average, current, or worst PI and did not differ between high and low pain groups Fewer positive self-statements were significantly associated with greater PI (average, current, worst); however, were not significantly associated with experimental PI and PS, and did not differ between high and low pain groups
Thastum 2005†† Article	56 C	x=11 (7-15)	80	Po:41; O:43; E:2; S:13; Ps:2	PI (C) – FPS (2x/day for 3 weeks) Mean and	Coping via Behavioral Distraction, Positive Self-Statements, Seeking Social Support, Cognitive	Corr and T-Test – Behavioral distraction and seeking social support were not significantly associated with PI, and they did not differ

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					high (pain ≥ 1.61 & disease activity <3)/low (remaining sample)	Distraction, Catastrophizing, Externalizing, & Total (C) – PCQ Pain Beliefs of Control, Harm, Disability, Solicitude, Medical Cure, Emotion, Total, Cognitive (disability + control + medical cure + harm), & Emotional (medical cure + emotion + solicitude) (C) – SOPA	between high and low pain groups Positive self-statements were not significantly correlated with PI; however, were significantly higher in the low pain group Corr, HR controlling for age, sex, disease duration, disease severity, disability, and pain beliefs, and T-Test – Cognitive distraction and externalizing were not significantly associated with PI and they did not differ between high and low pain groups Greater catastrophizing was significantly associated with greater PI (Corr, not Hierarchical Reg), and was significantly higher in the high pain groups Corr and T-Test – Lower control beliefs were significantly associated with greater PI and were significantly lower in the high pain group Greater harm and disability beliefs were significantly associated with greater PI and were significantly higher in the high pain group Emotion beliefs were not significantly associated with PI nor did they differ between high and low pain groups Lower medical cure beliefs and higher solicitude beliefs were significantly associated with greater PI; however, did not differ between the high and low pain groups HR controlling for age, sex, disease duration, disease severity, disability and pain coping – Worse pain beliefs (including cognitive beliefs but not emotional beliefs) were significantly associated with greater PI
Thastum 2011†† Article	47 C	* (7-15)	83	Po:40; O:45; S:13; Ps:2	PI (C) – FPS (2x/day for 3 weeks at baseline and 24 mos) Average and high (pain ≥ 1.61 & disease activity <3)/low (remainder)	Pain Beliefs of Control, Medical Cure, Harm, Disability, & Cognitive (disability + control + medical cure + harm) (C) – SOPA	Corr – Lower control beliefs at baseline and 24 mos were significantly associated with greater PI 24 mos later Medical cure beliefs at baseline and 24 months were not significantly associated with PI at 24 mos Corr and T-test – Greater harm and disability beliefs at baseline and 24 mos were significantly associated with greater PI 24 mos later, and significantly higher in the high pain group at 24 mos

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							HR controlling for disability, disease activity (and with/without baseline PI) – Greater cognitive beliefs at baseline significantly predicted PI at 24 mos
Thompson 1987 Article	23 C 23 P	x=10 (5-15) --	78 100	Po:48; O:22; S:26; U:4	PI (C) – VAS PPQ (current, worst, and high/low)	Number of elevated behavior and social competence subscales, Overall Adjustment, Externalizing, Internalizing & Social Competence (P) – CBCL P Family Relationships, Achievement, Active-Recreational Orientation, Cohesion, Conflict, Control, Expressiveness, Independence, Intellectua tural Orientation, Moral-Re s Emphasis, & Organizatıon (P) – FES	Welch's V – Children with 0, 1, 2, or 3 elevated behavior or social competence subscales did not significantly differ in current and worst PI Corr and Welch's V – Overall Adjustment, externalizing, internalizing, social competence, family relationships, conflict, active-recreational orientation, control, moral-religious emphasis, and organization were not significantly associated with PI, nor did they significantly differ between high and low pain groups Lower family achievement orientation was significantly associated with greater current, but not worst, PI, and it did not significantly differ between high and low pain groups Lower family cohesion and expressiveness were significantly associated with greater worst, but not current, PI, and they did not significantly differ between high and low pain groups. Lower family independence and intellectual-cultural orientation were significantly associated with greater current, but not worst, PI, and they did not significantly differ between high and low pain groups
Tupper 2012 Thesis	11 C	* (8-17)	*	Po:45	PI (C) – VAS PinGo (7x/day for 4 days) 4 categories: 0=None, 1-30=Mild, 31-69=Moderate, 70-100=Severe	Emotional valence (C) – FAS	GEE – There was a significantly greater probability of having no pain during times of high emotional valence (regardless of activation level)
Tupper 2013† Article	85 C	x=13 (8-17)	73	Po:42; O:22; E:9; S:14;	PI (C) – E-ouch VAS (3x/day for 7 days)	HRQoL (C) - PedsQL	LR controlling for disease activity, illness duration, age, and sex – Greater PI variability was significantly associated with lower well-being

Author, Year, Publication Type	Sample Size(s)	Age(s) x or Mdn (Range)	% Girls	% JIA Type	Pain: Construct (Reporter) – Measure	Psychosocial Factor(s): Construct (Reporter) – Measure	Main Findings: Analysis - Result
				Ps:7; U:5			
Upadhyay 2021 Article	16 C	x=13 (8-16)	69	Po:81; O:13; Ps:6	PI (C) – NRS PROMIS average and low (0-3)/high (>3)	Anxiety symptoms, Cognitive symptoms, Depression symptoms, and stress symptoms (C) – PROMIS	Corr and T-test – Anxiety symptoms, depression symptoms, and stress were not significantly associated with PI, nor did they significantly differ between high and low pain groups Lower cognitive function was significantly associated with greater PI, although it did not significantly differ between high and low pain groups
Vandvik 1990 Article	57 C 57 P	-- (7-16) --	67 --	Po:32; O:32; U:37	PI (C) – VAS	Psychosocial functioning (P) – CGAS Overall adjustment, Externalizing, & Internalizing (P) – CBCL	Corr – Psychosocial functioning, overall adjustment, externalizing, and internalizing were not significantly associated with PI
Vuorimaa 2008§§ Article	145 C	x=12 (8-15)	73	Po:50; O:40	PF (C) – SPQ (past 3 months)	Trait anxiety symptoms (C) – STAI-C Depression symptoms (C) – CDI Children were categorized into: 1) teenagers high in trait anxiety and depression; 2) children high in trait anxiety and low in depression; 3) children low in trait anxiety and depression; and 4) teenagers low in trait anxiety and depression	Discriminant Analyses – Cluster 1 (teenagers high in anxiety and depression symptoms) experienced significantly greater PF compared to the other clusters
Vuorimaa 2009§§ Article	142 C 142 P	x=12 (8-15) *	73 83	Po:50; O:50	PI (P) – VAS (current)	Trait anxiety symptoms (C) – STAI-C Depression symptoms (C) – CDI Children were categorized into: 1) teenagers high in trait anxiety and depression; 2) children high in trait anxiety and low in depression; 3) children low in trait anxiety and depression; and 4) teenagers low in trait anxiety and depression	Discriminant Analyses – Cluster 1 (teenagers high in anxiety and depression) experienced significantly greater PI compared to the other clusters
Vuorimaa 2011§§ Article	142 C 142 P	x=12 (8-15) *	73 83	Po:50; O:50	PF (C) – SPQ (past 3 months)	Depression symptoms (C) – CDI Anxiety symptoms (C) STAI-C Psychological, Somatic, & Social Self-Efficacy (C) – CASE	Corr and MR – Greater child depression and anxiety symptoms, lower child social self-efficacy, lower parent social self-efficacy, lower parent somatic self-efficacy, lower parent perception of the

Author, Year, Publication Type	Sample Size(s)	Age(s) x or Mdn (Range)	% Girls	% JIA Type	Pain: Construct (Reporter) – Measure	Psychosocial Factor(s): Construct (Reporter) – Measure	Main Findings: Analysis - Result
						P Depressive Symptoms (P) – BDI and HADS P Anxiety Symptoms (P) – HADS P Psychological, Social, & Somatic Self-Efficacy (C) – PASE P Parent Influence on Child Mood, Parent Perception of Child's Coping, & Parent Perception of Child's Well-being (P) – Author created	child's well-being, and lower parent perception of the child's coping were significantly associated with greater PF Greater parent depression symptoms (not MR with HADS) were significantly associated with greater PF Child psychological self-efficacy, child somatic self-efficacy, parent anxiety symptoms, parent psychological self-efficacy, and parent influence on child's mood were not significantly associated with PF
Yan 2020 Article	148 C	x=14 (8-17)	77	Po:18; O:53; E:13; S:7; Ps:2; U:7	PI (C) – NRS PROMIS (past week; multiple visits)	Depression symptoms (C) – PROMIS assessed across multiple visits	LMM – Increasing PI was significantly associated with an increase in depression symptoms

Note. Underlined text represents significant results. See Table 2.1 for master list of questionnaires and abbreviations.
‡, ‡‡, †, ††, §, §§ Studies with overlapping datasets; * Data provided but not specific to sample used in this review; -- Not reported
ANOVA = Analysis of Variance; ANCOVA = Analysis of Covariance; C = Child; Corr = Correlation; E = Enthesitis-Related Arthritis; GEE = Generalized Estimating Equations; HCP = Healthcare providers; HR = Hierarchical Regression; LiR = Linear Regression; LMM = Linear Mixed Models; LoR = Logistic Regression; MLM = Multilevel Models; MR = Multiple Regression; O = Oligoarticular Arthritis; P = Parents/Caregivers; Po = Polyarticular Arthritis; Ps = Psoriatic Arthritis; PF = Pain frequency; PI = Pain intensity; PS = Pain sensitivity/lower tolerance; Reg = Regression; S = Systemic Arthritis; SEM = Structural Equation Models; U = Undifferentiated/Other Arthritis

2.13. Supplementary Materials

Additional File 1: Search Strategy

Juvenile Idiopathic Arthritis terms	Pain terms (Schinkel et al., 2017)	Child terms (Leclercq et al., 2013)
Juvenile idiopathic arthritis Juvenile arthritis Juvenile chronic arthritis Juvenile rheumatoid arthritis Juvenile rhematic disease Inflammatory arthropathy Oligoarticular arthritis Pauciarticular arthrit* Polyarticular arthrit* Systemic arthrit* Enthesitis arthrit* Psoriatic arthrit* Undifferentiated arthrit*	Pain* Hurt* Discomfort* Chronic pain Acute pain Procedural pain Needle Injection* Syringe* Pain perception Nociception Pain threshold Hyperalgesi* Hypoalgesi* Enthesalgi* Central sensitivity Somatosensory profile Experimental pain Cold pressor Quantitative sensory test Water load Heat pain Thermal pain Pressure pain Exercise task Pain management Pain measurement	Infan* Perinat* Antepartum Ante-partum Postnatal* Post-natal* Baby* Babies Neonat* Neo-nat* Newborn* New-born* Child* Kid Kids Toddler* Girl* Girls Girlhood Boy* Boys Boyhood Preschool* Pre-school* Kindergarten* School* School child Juvenil* Minors* P?ediatric? Pe?diatric? Pediatric* Prepubescen* Pre-pubescen* Pubescen* Primary school Teen* Youth* Adolescen* Young adult* Young person* Young individual* Young people* Young population* Student* Highschool* High-school* High school Secondary school
OVID Medline search format: Arthritis, juvenile/ OR Stills disease/ OR Spondyloarthropathies/ OR	**OVID Medline search format:** Pain/ OR Chronic Pain/ OR Pain, Intractable/ OR Acute Pain/ OR Pain,	**OVID Medline search format:** Exp Infant/ OR Exp Infant, Newborn/ OR Exp Behavior, Infant/ OR Exp

Juvenile Idiopathic Arthritis terms	Pain terms (Schinkel et al., 2017)	Child terms (Leclercq et al., 2013)
Spondylitis/ OR Spondylitis, Ankylosing/ OR Arthritis, Psoriatic/ OR ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD" OR (Oligo* ADJ5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* ADJ5 arthrit*) OR (Poly* ADJ5 arthrit*) OR (Systemic* ADJ5 arthrit*) OR (Systemic-onset ADJ5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still? disease" OR "Still? syndrome" OR (Enthesit* ADJ5 arthrit*) OR (Enthesitis-related ADJ5 arthrit*) OR "Spondyloarthr?path*" OR "JuSpA" OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* ADJ5 arthrit*) OR (Undifferentiated ADJ5 arthrit*)).ti,ab,kw,kf.	Procedural/ OR Injections/ OR Injections, Intramuscular/ OR Injections, Intra-articular/ OR Injections, Subcutaneous/ OR Injections, Intravenous/ OR Syringes/ OR Perception, Pain/ OR Nociception/ OR Pain Threshold/ OR Management, Pain/ OR Analgesia/ OR Analgesics/ OR Measurement, Pain/ OR ("Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR "Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*" OR "Hyperalgesi*" OR "Hypoalgesi*" OR "Enthesalgi*" OR "Central sensitivity" OR "Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*").ti,ab,kw,kf.	Health, Infant/ OR Exp Child/ OR Exp Child, Preschool/ OR Exp Behavior, child/ OR Exp Health, Child/ OR Exp Pediatrics/ OR Exp Adolescent/ OR Exp Behavior, adolescent/ OR Exp Health, adolescent/ OR Exp Young Adult/ OR ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy" OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary ADJ2 school) OR (Primary ADJ2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young ADJ2 adult*) OR (Young ADJ2 person*) OR (Young ADJ2 individual*) OR (Young ADJ2 people*) OR (Young ADJ2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High ADJ2 school*) OR (Secondary ADJ2 school*)).ti,ab,kw,kf.
CINAHL search format: ((MH "Arthritis, juvenile rheumatoid") OR (MH "Spondyloarthropathies") OR (MH "Spondlyarthritis") OR (MH "Spondylitis, ankylosing") OR (MH "Arthritis, Psoriatic") OR (TI ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD" OR (Oligo* N5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* N5 arthrit*) OR (Poly* N5 arthrit*) OR (Systemic* N5 arthrit*) OR (Systemic-onset N5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still# disease" OR "Still# syndrome" OR (Enthesit* N5 arthrit*) OR (Enthesitis-related N5 arthrit*) OR "Spondyloarthr#path*" OR "JuSpA" OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* N5 arthrit*) OR (Undifferentiated N5 arthrit*))) OR (AB ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD"	**CINAHL search format:** ((MH "Pain") OR (MH "Chronic pain") OR (MH "Acute pain") OR (MH "Pain, procedural") OR (MH "Treatment related pain") OR (MH "Injections") OR MH ("Injections, intramuscular") OR (MH "Injections, intra-articular") OR (MH "Injections, subcutaneous") OR (MH "Injections, intravenous") OR (MH "Syringes") OR (MH "Nociceptive Pain") OR (MH "Pain threshold") OR (MH "Pain management") OR (MH "Analgesia") OR (MH "Analgesics") OR (MH "Pain measurement") OR (TI ("Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR "Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*" OR "Hyperalgesi"*" OR "Hypoalgesi"*" OR "Enthesalgi*" OR "Central sensitivity" OR "Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*")) OR (AB ("Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR	**CINAHL search format:** ((MH "Infant+") OR (MH "Infant, Newborn+") OR (MH "Infant Behavior") OR (MH "Child+") OR (MH "Child, Preschool") OR (MH "Child Behavior+") OR (MH "Child Health") OR (MH "Pediatrics+") OR (MH "Adolescence+") OR (MH "Adolescent behavior") OR (MH "Adolescent health") OR (MH "Young adult") OR (TI ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy*" OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary N2 school) OR (Primary N2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young N2 adult*) OR (Young N2 person*) OR (Young N2 individual*) OR (Young N2 people*) OR (Young N2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High N2 school*)

Juvenile Idiopathic Arthritis terms	Pain terms (Schinkel et al., 2017)	Child terms (Leclercq et al., 2013)
OR (Oligo* N5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* N5 arthrit*) OR (Poly* N5 arthrit*) OR (Systemic* N5 arthrit*) OR (Systemic-onset N5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still# disease" OR "Still# syndrome" OR (Enthesit* N5 arthrit*) OR (Enthesitis-related N5 arthrit*) OR "Spondyloarthr#path*" OR "JuSpA" OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* N5 arthrit*) OR (Undifferentiated N5 arthrit*))))	"Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*" OR "Hyperalgesi*" OR "Hypoalgesi*" OR "Enthesalgi*" OR "Central sensitivity" OR "Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*”)))	OR (Secondary N2 school*)) OR (AB ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy*" OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary N2 school) OR (Primary N2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young N2 adult*) OR (Young N2 person*) OR (Young N2 individual*) OR (Young N2 people*) OR (Young N2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High N2 school*) OR (Secondary N2 school*)))
PsycINFO search format: (TI ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD" OR (Oligo* N5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* N5 arthrit*) OR (Poly* N5 arthrit*) OR (Systemic* N5 arthrit*) OR (Systemic-onset N5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still# disease" OR "Still# syndrome" OR (Enthesit* N5 arthrit*) OR (Enthesitis-related N5 arthrit*) OR "Spondyloarthr#path*" OR "JuSpA" OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* N5 arthrit*) OR (Undifferentiated N5 arthrit*))) OR (AB ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD" OR (Oligo* N5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* N5 arthrit*) OR (Poly* N5 arthrit*) OR (Systemic* N5 arthrit*) OR (Systemic-onset N5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still# disease" OR "Still# syndrome" OR (Enthesit* N5 arthrit*) OR (Enthesitis-related N5 arthrit*) OR "Spondyloarthr#path*" OR "JuSpA"	**PsycINFO search format:** (DE "Pain") OR (DE "Chronic pain") OR (DE "Acute pain") OR (DE "Injections") OR (DE "Intramuscular injections") OR (DE "Subcutaneous injections") OR (DE "Intravenous Injections") OR (DE "Pain perception") OR (DE "Pain thresholds") OR (DE "Pain management") OR (DE "Analgesia") OR (DE "Analgesic drugs") OR (DE "Pain measurement") OR (TI ("Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR "Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*" OR "Hyperalgesi*" OR "Hypoalgesi*" OR "Enthesalgi*" OR "Central sensitivity" OR "Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*")) OR (AB ("Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR "Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*" OR "Hyperalgesi*" OR "Hypoalgesi*" OR "Enthesalgi*" OR "Central sensitivity" OR	**PsycINFO search format:** (DE "Child behavior") OR (DE "Child health") OR (DE "Pediatrics") OR (DE "Early Adolescence") OR (DE "Adolescent behavior") OR (DE "Adolescent health") OR (DE "Emerging Adulthood") OR (TI ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy*" OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary N2 school) OR (Primary N2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young N2 adult*) OR (Young N2 person*) OR (Young N2 individual*) OR (Young N2 people*) OR (Young N2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High N2 school*) OR (Secondary N2 school*))) OR (AB ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy*"

Juvenile Idiopathic Arthritis terms	Pain terms (Schinkel et al., 2017)	Child terms (Leclercq et al., 2013)
OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* N5 arthrit*) OR (Undifferentiated N5 arthrit*)))	"Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*"))	OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary N2 school) OR (Primary N2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young N2 adult*) OR (Young N2 person*) OR (Young N2 individual*) OR (Young N2 people*) OR (Young N2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High N2 school*) OR (Secondary N2 school*)))
Embase search format: ('Juvenile rheumatoid arthritis'/de) OR ('Polyarthritis'/de) OR ('Systemic juvenile idiopathic arthritis'/de) OR ('Enthesitis'/de) OR ('Spondyloarthropathy'/de) OR ('Spondylarthritis'/de) OR ('Ankylosing spondylitis'/de) OR ('Psoriatic arthritis'/de) OR ('Juvenile idiopathic arthrit*' OR 'JIA' OR 'Juvenile arthrit*' OR 'JA' OR 'Juvenile chronic arthrit*' OR 'JCA' OR 'Juvenile rheumatoid arthrit*' OR 'JRA' OR 'Juvenile rheumatic disease*' OR 'Inflammatory arthropathy' OR 'JRD' OR (Oligo* NEAR/5 arthrit*) OR 'Oligoarthrit*' OR 'oJIA' OR 'OligoJIA' OR (Pauci* NEAR/5 arthrit*) OR (Poly* NEAR/5 arthrit*) OR (Systemic* NEAR/5 arthrit*) OR (Systemic-onset NEAR/5 arthrit*) OR 'S-JIA' OR 'SJIA' OR 'SO-JIA' OR 'SOJIA' OR 'Still$ disease' OR 'Still$ syndrome' OR (Enthesit* NEAR/5 arthrit*) OR (Enthesitis-related NEAR/5 arthrit*) OR 'Spondyloarthr$path*' OR 'JuSpA' OR 'JSpA' OR 'Ankylosing spondylitis' OR 'JAS' OR (Psoria* NEAR/5 arthrit*) OR (Undifferentiated NEAR/5 arthrit*)):ti,ab,kw	**Embase search format:** ('Pain'/de) OR ('Chronic pain'/de) OR ('Intractable pain'/de) OR ('Procedural pain'/de) OR ('Injection'/de) OR ('Intramuscular drug administration'/de) OR ('Intraarticular drug administration'/de) OR ('Subcutaneous drug administration'/de) OR ('Intravenous drug administration'/de) OR ('Syringe'/de) OR ('Nociception'/de) OR ('Pain threshold'/de) OR ('Analgesia'/de) OR ('Analgesics'/de) OR ('Pain measurement'/de) OR ('Pain*' OR 'Hurt*' OR 'Discomfort*' OR 'Chronic pain' OR 'Acute pain' OR 'Procedural pain' OR 'Needle*' OR 'Injection*' OR 'Syringe*' OR 'Experimental pain' OR 'Cold pressor' OR 'Quantitative sensory test*' OR 'Water load' OR 'Heat pain' OR 'Thermal pain' OR 'Pressure pain' OR 'Exercise task' OR 'Nocicepti*' OR 'Pain* threshold*' OR 'Hyperalgesi*' OR 'Hypoalgesi*' OR 'Enthesalgi*' OR 'Central sensitivity' OR 'Somatosensory profile*' OR 'Pain* management*' OR 'Analgesi*' OR 'Pain* measurement'):ti,ab,kw	**Embase search format:** ('Infant'/exp) OR ('Newborn'/exp) OR ('Child'/exp) OR ('Preschool child'/exp) OR ('Child behavior'/exp) OR ('Child health'/exp) OR ('Pediatrics'/exp) OR ('Adolescent'/exp) OR ('Adolescence'/exp) OR ('Adolescent behavior'/exp) OR ('Adolescent health'/exp) OR ('Young adult'/exp) OR ('Infan*' OR 'Perinat*' OR 'Antepartum' OR 'Ante-partum' OR 'Postnatal*' OR 'Post-natal*' OR 'Baby*' OR 'Babies' OR 'Neonat*' OR 'Neo-nat*' OR 'Newborn*' OR 'New-born*' OR 'Child*' OR 'Kid' OR 'Kids' OR 'Toddler*' OR 'Girl*' OR 'Girls' OR 'Girlhood' OR 'Boy*' OR 'Boys' OR 'Boyhood' OR 'Preschool*' OR 'Pre-school*' OR 'Kindergarten*' OR 'School*' OR 'Juvenil*' OR 'Minors*' OR 'P$ediatric$' OR 'Pediatric*' OR 'Prepubescen*' OR 'Pre-pubescen*' OR 'Pubescen*' OR (Primary NEAR/2 school) OR (Primary NEAR/2 education) OR 'Teen*' OR 'Youth*' OR 'Adolescen*' OR (Young NEAR/2 adult*) OR (Young NEAR/2 person*) OR (Young NEAR/2 individual*) OR (Young NEAR/2 people*) OR (Young NEAR/2 population*) OR 'Student*' OR 'Highschool*' OR 'High-school*' OR (High NEAR/2 school*) OR (Secondary NEAR/2 school*)):ti,ab,kw
Scopus search format: TITLE-ABS-KEY ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD" OR (Oligo* W/5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* W/5 arthrit*)	**Scopus search format:** TITLE-ABS-KEY ("Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR "Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*"	**Scopus search format:** TITLE-ABS-KEY ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy*" OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-

Juvenile Idiopathic Arthritis terms	Pain terms (Schinkel et al., 2017)	Child terms (Leclercq et al., 2013)
OR (Poly* W/5 arthrit*) OR (Systemic* W/5 arthrit*) OR (Systemic-onset W/5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still? disease" OR "Still? syndrome" OR (Enthesit* W/5 arthrit*) OR (Enthesitis-related W/5 arthrit*) OR "Spondyloarthr?path*" OR "JuSpA" OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* W/5 arthrit*) OR (Undifferentiated W/5 arthrit*))	OR "Hyperalgesi*" OR "Hypoalgesi*" OR "Enthesalgi*" OR "Central sensitivity" OR "Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*")	school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary W/2 school) OR (Primary W/2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young W/2 adult*) OR (Young W/2 person*) OR (Young W/2 individual*) OR (Young W/2 people*) OR (Young W/2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High W/2 school*) OR (Secondary W/2 school*))
Cochrane search format: "Arthritis, juvenile" [MeSH] OR "Spondyloarthropathies" [MeSH] OR "Spondylitis, Ankylosing" [MeSH] OR "Spondylarthritis" [MeSH] OR "Arthritis, psoriatic" [MeSH] OR ("Juvenile idiopathic arthrit*" OR "JIA" OR "Juvenile arthrit*" OR "JA" OR "Juvenile chronic arthrit*" OR "JCA" OR "Juvenile rheumatoid arthrit*" OR "JRA" OR "Juvenile rheumatic disease*" OR "Inflammatory arthropathy" OR "JRD" OR (Oligo* near/5 arthrit*) OR "Oligoarthrit*" OR "oJIA" OR "OligoJIA" OR (Pauci* near/5 arthrit*) OR (Poly* near/5 arthrit*) OR (Systemic* near/5 arthrit*) OR (Systemic-onset near/5 arthrit*) OR "S-JIA" OR "SJIA" OR "SO-JIA" OR "SOJIA" OR "Still? disease" OR "Still? syndrome" OR (Enthesit* near/5 arthrit*) OR (Enthesitis-related near/5 arthrit*) OR "Spondyloarthr?path*" OR "JuSpA" OR "JSpA" OR "Ankylosing spondylitis" OR "JAS" OR (Psoria* near/5 arthrit*) OR (Undifferentiated near/5 arthrit*)):ti,ab,kw	**Cochrane search format:** "Pain" [MeSH] OR "Chronic pain" [MeSH] OR "Intractable pain" [MeSH] OR "Acute pain" [MeSH] OR "Pain, procedural" [MeSH] OR "Injections" [MeSH] OR "Injections, intramuscular" [MeSH] OR "Injections, intra-articular" [MeSH] OR "Injections, subcutaneous" [MeSH] OR "Injections, intravenous' [MeSH] OR "Syringes" [MeSH] OR "Pain perception" [MeSH] OR "Nociceptive pain" [MeSH] OR "Pain management" [MeSH] OR "Analgesia" [MeSH] OR "Analgesics" [MeSH] OR "Pain measurement" [MeSH] OR OR "Pain threshold" [MeSH] OR (“Pain*" OR "Hurt*" OR "Discomfort*" OR "Chronic pain" OR "Acute pain" OR "Procedural pain" OR "Needle*" OR "Injection*" OR "Syringe*" OR "Experimental pain" OR "Cold pressor" OR "Quantitative sensory test*" OR "Water load" OR "Heat pain" OR "Thermal pain" OR "Pressure pain" OR "Exercise task" OR "Nocicepti*" OR "Pain* threshold*" OR "Hyperalgesi*" OR "Hypoalgesi*" OR "Enthesalgi*" OR "Central sensitivity" OR "Somatosensory profile*" OR "Pain* management*" OR "Analgesi*" OR "Pain* measurement*"):ti,ab,kw	**Cochrane search format:** "Infant" [MeSH][exp] OR "Infant, Newborn" [MeSH][exp] OR "Infant behavior" [MeSH][exp] OR "Infant Health" [MeSH][exp] OR "Child" [MeSH][exp] OR "Child, preschool" [MeSH][exp] OR "Child behavior" [MeSH][exp] OR "Child health" [MeSH][exp] OR "Pediatrics" [MeSH][exp] OR "Adolescent" [MeSH] OR "Adolescent behavior" [MeSH][exp] OR "Adolescent health" [MeSH][exp] OR "Young adult" [MeSH][exp] OR ("Infan*" OR "Perinat*" OR "Antepartum" OR "Ante-partum" OR "Postnatal*" OR "Post-natal*" OR "Baby*" OR "Babies" OR "Neonat*" OR "Neo-nat*" OR "Newborn*" OR "New-born*" OR "Child*" OR "Kid" OR "Kids" OR "Toddler*" OR "Girl*" OR "Girls" OR "Girlhood" OR "Boy*" OR "Boys" OR "Boyhood" OR "Preschool*" OR "Pre-school*" OR "Kindergarten*" OR "School*" OR "Juvenil*" OR "Minors*" OR "P?ediatric?" OR "Pediatric*" OR "Prepubescen*" OR "Pre-pubescen*" OR "Pubescen*" OR (Primary near/2 school) OR (Primary near/2 education) OR "Teen*" OR "Youth*" OR "Adolescen*" OR (Young near/2 adult*) OR (Young near/2 person*) OR (Young near/2 individual*) OR (Young near/2 people*) OR (Young near/2 population*) OR "Student*" OR "Highschool*" OR "High-school*" OR (High near/2 school*) OR (Secondary near/2 school*)):ti,ab,kw

Additional File 2: Data Extraction Template

Name of Data Extractor	
Study Details	- **Covidence study ID:** - **Full article title:** - **Journal:** - **First Author:** Last Name, First Name - **Year of publication:** - **Country of first author:** - **Publication type:** published article, abstract, dissertation, other (specify) - **Possible conflicts of interest:** yes (specify), no

Study Population	- **Sample size:**
	- **Sample:** youth, caregivers, healthcare providers, other (specify)
	- **For each population, fill out the relevant details below:**
	  - Age
	  - **Measurement:** Mean or median
	  - **Age:**
	  - **Age range:**
	  - **Sex (percentage male, female, other):**
	  - **Diagnosis (percentage polyarticular, oligoarticular, enthesitis, systemic, psoriatic, undifferentiated, other):**
	  - **Disease status (percentage active, inactive, in remission):**
	  - Length of the disease
	  - **Measurement:** Disease duration or disease onset
	  - **Measurement:** Mean or median
	  - **Duration:**
	  - **Range:**
	  - **Other details:**

Study Design	- **Design:** cross-sectional, cohort, case control, case series, randomized control trial, quasi-experimental trial, other (specify) - **Start date:** - **End date:** - **Study Duration/Follow-Up:** - **Countries recruited from:** - **Setting:** clinics, community, other (specify) - **Was this sample from a cohort:** no, yes (specify) - **Other details:**

Measures	- Exposure (i.e., psychosocial factor)
	  - **Construct:**
	  - **Data:** numerical or categorial
	  - **Reporter:** youth, parent, healthcare provider, other (specify)
	  - **Measure:**
	  - **Citation:**
	- Outcome (i.e., pain)
	  - **Measurement**: intensity, frequency, or sensitivity
	  - **Data:** numerical or categorical
	  - **Reporter:** youth, parent, healthcare provider, other (specify)
	  - **Measure:**
	  - **Time (e.g., current, past week):**
	  - **Citation:**

Results	- Association (complete the following for each)
	  - **Exposure (i.e., psychosocial factor)**
	  - **Outcome (i.e., pain)**
	  - **Significance of association:** yes or no
	  - **Direction of association:** positive or negative
	  - **Other information (e.g., statistical technique, covariates):**

Additional File 3.1: Quasi-Experimental Studies

Methodological Quality

For the five quasi-experimental studies, critical appraisal scores ranged from 78% to 100%, with the lack of a comparison group and participant retention as the biggest limitations (Additional File 3.2).

Results

Child factors. Across five psychosocial interventions, which may be considered as an external source of support in completing a secondary appraisal, 19/24 associations were significant (Lavigne et al., 1992; Lomholt et al., 2015; Stinson et al., 2016; Stinson et al., 2020; Walco et al., 1992) . Walco et al. (1992) tested the efficacy of an 8-week cognitive-behavioral therapy (CBT) intervention in 13 children with JIA and their parents and followed up six and 12 months later. All 10 of the associations demonstrated reduced pain intensity post-intervention. Comparatively, Lomholt et al. (2015) ran a 6-week group-based CBT intervention for nine children with JIA. After the intervention; however, no differences in pain intensity were observed between the treatment and control groups. Lavigne et al. (1992) provided a 6-week treatment package for pain management to eight children with JIA and their parents. Despite the small sample size, 6/9 associations demonstrated the benefits of the intervention in reducing both pain intensity and frequency, and the remaining were trending in the expected direction. Stinson et al. (2016) developed the iPeer2Peer intervention and assessed the efficacy of an 8-week trial in 16 children with JIA and a control group. No differences emerged in pain intensity scores between the treatment and control group post intervention. Stinson et al. (2020) also developed the 12-week Teens Taking Charge intervention and compared the outcomes at 3, 6, and 12 months for 88 children who participated in the intervention to 131 controls. All three associations showed significantly reduced pain intensity in the treatment condition at all timepoints. Taken together, participation in CBT or pain specific interventions tends to be predictive of lower pain reports in children with JIA.

Discussion

The efficacy of five psychosocial interventions varying in their orientation and delivery in reducing JIA pain were reviewed. Most demonstrated significant reductions in JIA pain intensity and frequency post intervention or in comparison to the control group. While a complete review and comparison of these interventions is beyond the scope of this study, Cohen et al. (2017) and Butler et al. (2022) have recently published comprehensive reviews in this area. Nevertheless, psychosocial interventions are promising way to foster improvements in JIA pain along with other important outcomes.

Additional File 3.2: Critical Appraisal Results for Quasi Experimental Studies

Author & Year	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	%
Lavigne 1992	Y	U	Y	Y	Y	Y	Y	Y	Y	89%
Lomholt 2015	Y	Y	Y	Y	Y	Y	Y	Y	Y	100%
Stinson 2016	Y	U	Y	Y	Y	Y	Y	Y	Y	89%
Stinson 2020	Y	U	Y	Y	Y	N	Y	Y	Y	78%
Walco 1992	Y	Y	Y	N	Y	N	Y	Y	Y	78%
%	100%	40%	100%	80%	100%	60%	100%	100%	100%	

Note. JBI critical appraisal for quasi-experimental studies: Q1 = Is it clear in the study what is the 'cause' and what is the 'effect' (i.e., there is no confusion about which variable comes first)? Q2 = Were the participants included in any comparisons similar? Q3 = Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest? Q4 = Was there a control group? Q5 = Were there multiple measurements of the outcome both pre and post the intervention/ exposure? Q6 = Was follow up complete/were differences between groups in terms of their follow up adequately described and analyzed? Q7 = Were the outcomes of participants included in any comparisons measured in the same way? Q8 = Were outcomes measured in a reliable way? Q9 = Was appropriate statistical analysis used?

‡, ‡‡, ‡‡‡, †, ††, §, §§ Studies with overlapping datasets

Y = Yes; N = No; U = Unclear

Additional File 3.3: Quasi-Experimental Study Characteristics and Results

Author, Year, Publication Type	Sample Size(s)	Age(s) x̄ or Mdn (Range)	% Female	% JIA Type	Pain: Construct (Reporter) – Measure	Psychosocial Factor(s): Construct (Reporter) – Measure	Main Findings: Analysis - Result
Lavigne 1992 Article	8 C 7 P 5 HCP	Mdn=14 (9-17) -- --	88 100 --	Po:75; O:13; E:13	PI (C & P) – VAS 3x/day for 1 mos pre, post, and 6 mos later PF (C & P) – VAS the percentage of ratings above 5 PS (HCP) – --	Treatment (C) – 6 sessions of biweekly therapy for pain management / Waitlist Control	Mann Whitney U-Test – Treatment and Waitlist Control groups did not significantly differ in child reported PI and PF The treatment group had significantly lower parent reported PI and PF compared to Waitlist Control Repeated Measured ANOVA – Children and parents reported PI and PF, and HCP reported PS tended to decrease over time in response to the treatment
Lomholt 2015 Article	19 C	* (9-14)	79	Po:32; O:42; E:5; S:11; Ps:11	PI (C) – FPS-R assessed 2x/day for 1 week (averaged)	Treatment (C) – 6 sessions of Cognitive Behavioral Therapy group/Waitlist Control	ANCOVA controlling for pre-intervention data and disease status – Treatment and Waitlist Control groups did not significantly differ in PI post treatment
Stinson 2016 Article	32 C	x̄=14 (12-17)	97	Po:41; O:31; E:3; Ps:25	PI (C) – NRS RPI assessed at baseline and post intervention	Treatment (C) – iPeer2Peer intervention for 8 weeks / WLC	Marginal Linear Models – Treatment and WLC groups did not significantly differ in their PI at study completion
Stinson 2020 Article	219 C 197 P	x̄=14 (12-17) --	70 80	Po:32; O:32; E:16; S:2; Ps:11; U:7	PI (C) – NRS RPI assessed at baseline, 3 mos (post intervention), 6 mos, and 12 mos	Treatment (C) – Teens Taking Charge intervention for 12 weeks / WLC	LMM – The treatment group demonstrated significantly lower PI at 3, 6, and 12 mos compared to the WLC
Walco 1992 Article	13 C 13 P	x̄=10 (4-16) --	62 --	O:62; S:38	PI (C & P) – VAS PPQ assessed 2x/day at baseline, post intervention, 6 and 12 mos	Treatment (C) – 8 Week Cognitive Behavioral Therapy Intervention	T-test – Child and parent reported PI (AM and PM) significantly decreased from baseline to post intervention, 6 mos, and 12 mos

Note. Underlined text represents significant results.
ANOVA = Analysis of Variance; ANCOVA = Analysis of Covariance; C = Child; E = Enthesitis-Related Arthritis; HCP = Healthcare providers; LMM = Linear Mixed Models; O = Oligoarticular Arthritis; P = Parents/Caregivers; Po = Polyarticular Arthritis; Ps = Psoriatic Arthritis; PF = Pain frequency; PI = Pain intensity; S = Systemic Arthritis; U = Undifferentiated/Other Arthritis

CHAPTER 3: UNDERSTANDING PERFECTIONISM IN YOUTH WITH JUVENILE IDIOPATHIC ARTHRITIS AND THEIR CAREGIVERS

The manuscript based on this study is detailed below. Yvonne Brandelli, under the supervision of Drs. Christine Chambers and Sean Mackinnon, was responsible for developing the research question, methodology, and analytic approach; preregistering the hypotheses (https://osf.io/wnxb8); and obtaining ethical approval. She developed the study protocol and data collection procedures, recruited participants, and led the data analysis and interpretation with the support of her supervisors and co-authors. Ms. Brandelli wrote the initial draft of this manuscript and received and incorporated feedback from the study co-authors. The manuscript was submitted to the Journal of Pediatric Psychology on February 14, 2024. The current reference for this manuscript is:

Brandelli, Y. N., Mackinnon, S. P., Chambers, C. T., Parker, J. A., Huber, A. M., Stinson, J. N., Johnson, S. A., & Wilson, J. P. (Revise and Resubmit). Understanding perfectionism in youth with juvenile idiopathic arthritis and their caregivers. *Journal of Pediatric Psychology.*

3.1. Abstract

Objective: Youth with juvenile idiopathic arthritis (JIA) experience elevated rates of internalizing symptoms, although more research is required to understand this phenomenon. Perfectionism, a multidimensional personality trait that involves dimensions such as striving for flawlessness (self-oriented perfectionism; SOP) and feeling that others demand perfection (socially prescribed perfectionism; SPP), is a well-known risk factor that has received minimal attention in pediatric populations. A priori hypotheses explored the relationships between youth and parent perfectionism and internalizing symptoms (i.e., depression and anxiety) in youth with JIA, as mediated by 1) youth/parent negative self-evaluations and 2) youth self-concealment.

Methods: 156 dyads comprised of youth (13-18 years) with JIA and a caregiver completed an online survey. Participants independently completed questionnaires about trait perfectionism, negative self-evaluations (i.e., pain catastrophizing and fear of pain), self-concealment, and internalizing symptoms.

Results: Preregistered hypotheses were partially supported. Positive relationships were observed between parent and youth SOP and negative self-evaluations, youth SOP and internalizing symptoms, and youth negative self-evaluations and internalizing symptoms. A negative relationship was found between parent SOP and depression symptoms. Indirect effects were observed for youth SOP predicting internalizing symptoms through pain catastrophizing. Exploratory mediations suggested youth SPP might predict internalizing symptoms directly and indirectly through self-concealment.

Conclusion: Perfectionism in youth and parents appears to play a role in the internalizing symptoms of youth with JIA and may manifest through negative self-evaluations and

self-concealment. While future research is needed, screening of perfectionistic tendencies in youth with JIA and their parents may help guide assessment, prevention, and treatment efforts.

3.2. Introduction

Juvenile idiopathic arthritis (JIA) is a chronic inflammatory disease affecting up to 8 million children worldwide (Petty, Laxer, & Wedderburn, 2021). Pain is one of the most frequently reported symptoms (Canadian Paediatric Society, 2009), with one daily diary study reporting that youth experienced pain on average 73% of days across a 2-month period (Schanberg et al., 2003). Both the diagnosis of a chronic health condition and the increased rates of pain put youth with JIA at risk for worse mental health outcomes such as anxiety and depression (Brandelli et al., 2023; Fair et al., 2019; Li et al., 2023). In a systematic review of the literature, Fair et al. (2019) found that many youth with JIA experience clinically significant symptoms of depression (7-36%) and anxiety (7-64%). Moreover, these estimates were made prior to the COVID-19 pandemic. It is likely that rates have increased (Racine et al., 2021), making it an especially timely endeavor to understand factors impacting the experience of anxiety and depression in youth with JIA to tailor assessment, prevention, and treatment efforts.

One risk factor that is a frequently presenting clinical phenomenon that has received minimal attention in the pediatric and pain literatures is perfectionism (Randall, Gray, et al., 2018). Perfectionism is a multidimensional personality trait that involves striving for flawlessness, setting exceedingly high standards, making overly critical self-evaluations, and feeling pressure to meet standards imposed by others (Frost et al., 1990; Hewitt & Flett, 1991). The present study utilized Hewitt and Flett's (1991) model, which proposes three dimensions: self-oriented perfectionism (SOP; a self-imposed pursuit of exceedingly high standards and self-scrutiny when that is not actualized), socially prescribed perfectionism (SPP; the belief that other people demand perfection from

oneself), and other-oriented perfectionism (OOP; demanding perfection from other people). Some researchers (e.g., O'Connor et al., 2009; Piercy et al., 2020) have further separated the dimension of SOP to reflect the presence of adaptive (i.e., SOP-strivings) and maladaptive (i.e., SOP-critical) components. Perfectionistic traits are posited to emerge during childhood in response to the child's own characteristics (e.g., temperament, ability) and their broader family and sociocultural environment (e.g., expectations, contingencies, social learning, social reactions, attachment) (Flett et al., 2002; Smith et al., 2022).

Perfectionism is a well-known risk factor for anxiety and depression in youth (e.g., Affrunti & Woodruff-Borden, 2014; Morris & Lomax, 2014) that has begun to receive attention in the context of health conditions (Behrens, 2017; Flett et al., 2011; Hadjistavropoulos et al., 2007; Molnar et al., 2016). The few studies exploring dimensions of perfectionism in pediatric populations have found them to be positively associated with externalizing and internalizing symptoms in youth with inflammatory bowel disease (IBD; Piercy et al., 2020); and somatization, catastrophizing, and fear of pain in chronic pain populations (Randall, Smith, et al., 2018). Moreover, dimensions of parent perfectionism have been modestly associated with youth and parent catastrophizing, youth pain-related fear, and youth functional disability (Randall, Smith, et al., 2018).

There has been a recent call to better understand the mechanisms in which youth and parent perfectionism contribute to anxiety and depression in pediatric pain (Randall, Gray, et al., 2018). Randall, Gray, et al. (2018) theorized that the coexistence of perfectionism and pain is not a coincidence. Rather, perfectionism in youth and their

parents likely amplifies challenges (such as internalizing symptoms) in the context of pain by undermining coping and recovery efforts. More specifically, perfectionism and pain are thought to be related through biological (i.e., via increased stress and subsequent alterations in pain processing and inflammatory processes), psychological (i.e., via cognitive and behavioral correlates that can precipitate, maintain, or exacerbate pain), and social (i.e., via greater interpersonal challenges and school problems) processes (Randall, Gray, et al., 2018). This is particularly relevant for youth with JIA, who uniquely face an idiopathic, variable, chronic, and invisible disease. The relationships between perfectionism and internalizing symptoms and their mechanisms (e.g., disease- and pain-specific cognitions) may be unique. The Stress and Coping Cyclical Amplification Model of Perfectionism in Illness (SCCAMPI) proposed by Molnar and colleagues (2016) suggests that individuals high in perfectionism may be susceptible to amplified stress, maladaptive coping, and worse health-related outcomes through various inter- and intra-personal pathways which may mediate or explain the experience of amplified stress.

One pathway describes how individuals high in trait perfectionism may engage in negative self-evaluations when they are unable to meet their impossibly high standards (Molnar et al., 2016). Although a chronic disease such as JIA may encourage some individuals to become more flexible with their goals, individuals high in SOP (given its emphasis on self-scrutiny when perfection is not actualized) may be unwilling to re-adjust their goals to account for their pain and disease flares. As such, youth with JIA and their parents who are high in SOP may have a heightened awareness of the limits imposed by JIA (e.g., more catastrophic thoughts and pain-related fears), but their unwillingness to adjust their goals may exacerbate negative self-evaluations thus leading

to more internalizing symptoms in youth.

Another pathway suggests that individuals with perfectionism maintain their self-image by hiding their unappealing characteristics (such as a diagnosis of JIA and its symptoms) through self-concealment (Molnar et al., 2016). The 'invisibility' of JIA pain highlights the need for youth to communicate their experiences and advocate for adaptations; however, this may also make it easier for youth high in both SOP and SPP (because they want to achieve perfection and convey this to others), and with parents high in OOP (because they feel perfection is expected of them) to conceal their symptoms. Although in the short-term there may be benefits to this (e.g., appearing "normal"), these may have undue long-term consequences for the individual's health and well-being (Larson & Chastain, 1990). As such, self-concealment may mediate the relationship between the youth's own perfectionism (or the parent's OOP) and internalizing symptoms.

Taken together, despite advances in recognizing that perfectionism contributes to one's mental health, little is known about the mechanisms of this relationship, especially as it relates to parent/child dyads and the context of JIA. The objective of this study is to examine the relationships between youth and parent perfectionism and mental health outcomes in youth with JIA. It was hypothesized that:

(H1) Youth and parent SOP would be associated with increased anxiety and depression in youth, and these relationships would be mediated by negative self-evaluations (as measured by pain catastrophizing and fear of pain in youth and parents).

(H2) Youth SOP and SPP and parent OOP would be associated with heightened anxiety and depression in youth, and these relationships would be mediated by the

youth's self-concealment.

3.3. Methods

3.3.1. Study Design

This dyadic (youth/parent), Internet-based, cross-sectional research was approved by the IWK Health's Research Ethics Board. Consistent with patient-oriented research, this study was conducted in consultation with a leader in patient engagement (IJ), and in partnership with Cassie and Friends, a Canadian parent-led organization for families of children with rheumatic diseases (www.cassieandfriends.ca). A panel of three diverse youth and parent partners became involved during the initial phases of this study (Brandelli, Jordan, et al., 2022) and provided input on study conceptualization through to dissemination. This study was preregistered prior to collecting and analyzing the data (https://osf.io/wnxb8), and the deidentified data and syntax are openly available through Open Science Framework (OSF; https://osf.io/svn8d/?view_only=d2c8e3e9e6544b76be10c137b134a841).

3.3.2. Participants

Participants included English-speaking youth between 13-18 years old with a diagnosis of JIA and one of their parents or caregivers (herein referred to as parents). Purposeful recruitment occurred worldwide between November 2021 and April 2023 and predominantly took place through online and social media platforms (e.g., arthritis and pain communities, advertisements over Facebook and Instagram, blog posts). Additional strategies included recruitment through previous studies, posters at rheumatology and pain clinics, research registries, and industry partnerships.

Details about the power analysis are in the preregistered plan on OSF. The aim

was to recruit 319 dyads or to terminate data collection by Spring 2023 given funding timelines, whichever came first. Repeated interference by sham respondents (e.g., disingenuous and software automated responses) slowed data collection considerably. Recruitment efforts produced less than the required sample size; although, post-hoc power analyses demonstrated most models retained at least 70% power to detect indirect effects. Two hundred and six dyads consented online, 33 of whom were ineligible given their diagnosis or age and 17 of whom stopped after providing consent, resulting in a final sample size of 156 unique dyads.

Missing data was complex, as not all dyads had data for both members. Most parents completed the full survey (n = 129; 82.7%), some provided partial data (n = 11; 7.0%), and the rest did not complete the survey at all (n = 16; 10.3%). Most children completed the full survey (n = 122; 78.2%), some provided partial data (n = 7; 4.5%), and the remainder did not complete the survey at all (n = 27; 17.3%). In total, 104 dyads (66%) had complete data (i.e., both parents and children completed all measures). All viable data, including partial data, was analyzed when appropriate.

3.3.3. Measures

Demographics and Medical Variables

Youth and parents reported on ethnicity (Canadian Institute for Health Information, 2022) and other demographic (e.g., age, sex, gender), medical (e.g., diagnosis, disease status), and pain-related (Birnie, Hundert, et al., 2019) variables.

Perfectionism

Youth Perfectionism. The 22-item Child and Adolescent Perfectionism Scale (CAPS; Flett et al., 2016) assessed two dimensions of perfectionism on a scale of 1

(*False – not at all true of me*) to 5 (*very true of me*): SOP (12 items; e.g., *"I try to be perfect in everything I do"*) and SPP (10 items; e.g., *"My family expects me to be perfect"*). In psychometric studies the CAPS has demonstrated good internal consistency, concurrent validity, and test-retest reliability (Flett et al., 2016); however, debate exists regarding the number of dimensions captured in the measure (O'Connor et al., 2009).

Parent Perfectionism. Parent perfectionism was assessed using three subscales from the Big Three Perfectionism Scale (BTPS; Smith et al., 2016) on a scale of 1 (*strongly disagree*) to 7 (*strongly agree*): SOP (5 items; e.g., *"I have a strong need to be perfect"*), SPP (4 items; e.g., "*People expect too much from me"*), and OOP (5 items; e.g., *"I expect those close to me to be perfect*"). Research has demonstrated good internal consistency and preliminary evidence for convergent and divergent validity (Smith et al., 2016).

Pain Catastrophizing

Youth Pain Catastrophizing. The Pain Catastrophizing Scale for Children (PCS-C; Crombez et al., 2003; Crombez et al., 2012) measures pain-related maladaptive thinking patterns. Thirteen items are administered on a scale from 0 (*not at all*) to 4 (*extremely*) totaling three dimensions of pain catastrophizing (helplessness, magnification, and rumination; e.g., *"When I am in pain, I become afraid that the pain will get worse"*). In past research, internal consistency has been good (Crombez et al., 2003).

Parent Pain Catastrophizing. Parent catastrophizing was assessed using the Pain Catastrophizing Scale for Parents (PCS-P; Crombez et al., 2012). The scale uses the same 13-items as the PCS-C, with modified language (e.g., "*When my child is in pain, I*

become afraid the pain will get worse"). The initial validation study reported strong construct validity and internal reliability (Goubert et al., 2006).

Fear of Pain

Youth Fear of Pain. The Fear of Pain Questionnaire Child - Short Form (FOPQC-SF; Heathcote et al., 2020) measures pain-related fear and two sub-dimensions: fear and avoidance. Ten items (e.g., *"Pain causes my heart to beat fast or race"*) were rated on a scale from 0 (*strongly disagree*) to 4 (*strongly agree*). The validation study showed strong construct validity, criterion validity, test-retest reliability; and good internal consistency (Heathcote et al., 2020).

Parent Fear of Pain. Parent pain-related fears were measured using the 21-item Parent Fear of Pain Questionnaire (PFOPQ; Simons et al., 2015). This scale compiles four dimensions of pain-related fear in parents: fear, avoidance, school, and movement. Items (e.g., *"My child's feelings of pain are scary for me"*) were rated on a scale from 0 (*strongly disagree*) to 4 (*strongly agree*). Excellent internal consistency, construct- and criterion-related validity have been observed (Simons et al., 2015).

Youth Self-Concealment

Youth self-concealment was assessed using the 5-item concealment subscale of the Health-Related Felt Stigma and Concealment Questionnaire (FSC-Q; Laird et al., 2020). While developed for youth with abdominal symptoms, the instructions were modified to suit a JIA population (i.e., *""Symptoms" refers to any arthritis symptoms, including: aches, pain, stiffness, swelling").* Items (e.g., *I try not to let other people know when I'm having symptoms*) were rated on a scale of 1 (*strongly disagree*) to 5 (*strongly agree*). This subscale demonstrated good internal consistency and construct validity in

the validation study (Laird et al., 2020).

Youth Internalizing Symptoms

Internalizing symptoms were assessed with the 25-item youth self-report version of the Revised Child Anxiety and Depression Scale Short Version (RCADS-25; Ebesutani et al., 2012). Youth responded to 15 items about feelings of anxiety (e.g., "*I worry what other people think of me"*) and 10 items about feelings of depression (e.g., *"I feel sad or empty"*) on a scale from 0 (*never*) to 3 (*always*). This scale, validated on healthy and clinical samples of youth, displays good internal consistency and acceptable concurrent validity (Ebesutani et al., 2012).

3.3.4. Procedures

Participants self-selected into this study. After one dyad member verified their eligibility through a screening questionnaire, youth and parents were emailed a unique survey link where they independently provided informed consent and completed a 45-minute battery of questionnaires online. Questions were mandatory; however, participants had the option of selecting "*prefer not to answer*" for all items which was treated as missing data. Completed participants were offered a $15 CAD online gift card and completed dyads were also offered the option of entering a draw to win one of two pairs of $250 CAD gift cards.

To ensure the validity of the data collected, safeguards were put in place to protect against sham responses (Teitcher et al., 2015). This included, but was not limited to, a screening questionnaire, attention checks, "spam trap" questions, captchas, passwords, and the prevention of multiple submissions from the same Internet Protocol address.

3.3.5. Analytic Plan

Youth-parent dyads were paired, and total scores were calculated for each scale (items were averaged such that higher values indicate greater levels of the variable in question). Descriptive statistics and bivariate correlations were used to describe measures of pain, perfectionism, mediators, and outcomes. A Full Information Maximum Likelihood approach was used to account for missing data.

Given discrepancy in the literature (Flett et al., 2016; O'Connor et al., 2009), an exploratory factor analysis of the CAPS was planned to determine whether the three-factor/14-item structure (i.e., SOP-strivings, SOP-critical, SPP) was superior to the two-factor/22-item structure (i.e., SOP, SPP) in these data. Using the jmv() package in R (https://www.r-project.org/) with maximum likelihood estimation and oblique rotation, the number of factors was extracted using parallel analysis, logical interpretability, and factor loadings of >.40.

Path analysis using the lavaan() package (Rosseel, 2012) in R was used to test hypotheses. Analyses were informed by actor-partner interdependence modelling with distinguishable dyads (Ledermann et al., 2011), exploring associations between variables at the individual (i.e., actor and partner effects) and partner (i.e., interactions between the actor and the partner effects) levels (Cook & Kenny, 2016). Models used maximum likelihood robust estimation and robust estimates of standard errors. Indirect effects were calculated using bootstrapping with 5000 resamples. Preregistered hypotheses were tested in 6 path models (Figures 3.1-3.3), wherein standardized paths, covariances, and R^2 values are reported.

3.4. Results

3.4.1. Exploratory Factor Analysis

Results of the parallel analysis suggested the presence of two factors in both the 14-item and 22-item versions of the CAPS. A logical pattern of factors (SOP and SPP) mapped to those proposed by Flett et al. (2016) instead of the 3-factor version proposed by O'Connor et al. (2009). Factor loadings for the selected two-factor/22-item model (ranging from .40 to .83) are presented in Supplementary File 1.

3.4.2. Sample Characteristics

The sample consisted of 129 adolescents with JIA and 140 parents. Participant demographics are found in Table 3.1. The sample consisted of largely female youth (67.9%) and parents (94.9%) with mean ages of 15.29 (*SD* = 1.62) and 45.24 (*SD* = 4.87). Over half of the sample was currently experiencing active disease and chronic pain (i.e., pain more days than not over the past 3 months). The usual pain severity experienced by youth was 4.96 (*SD* = 2.23) on the 11-point Numeric Rating Scale. Internal consistencies ranged from good to excellent (.84-.94; Table 3.2). Weak to moderate positive correlations were generally observed within and between youth and parent measures of perfectionism, pain catastrophizing, fear of pain, and anxiety and depression. The strongest correlations were between pain catastrophizing and fear of pain in youth (r = .66) and parents (r = .71). Two notable exceptions include the lack of correlation between dimensions of parent perfectionism and youth anxiety and depression, and the weak/absent correlations between youth self-concealment and youth perfectionism.

3.4.3. Hypothesis 1: Negative Self-Evaluations Mediating the Relationships between Youth/Parent Self-Oriented Perfectionism and Youth Anxiety and

Depression

Four path analyses were completed to test the above-mentioned hypothesis by varying the mediator (pain catastrophizing and fear of pain) and outcome (anxiety and depression) one at a time across analyses (Figure 3.1). Across all models, there were significant, positive covariances between youth and parent SOP, pain catastrophizing, and fear of pain. That is, youth and parents were more similar to each other on these traits than would be expected due to chance.

In model 1, standardized regression coefficients confirmed that youth SOP was positively related to youth catastrophizing, and parent SOP was positively related to parent catastrophizing, each accounting for 6% of the variance. This model predicted 39% of the variance in youth anxiety; however, not all paths were statistically significant. Specifically, youth SOP directly and indirectly predicted anxiety through youth catastrophizing (*a1b1* = 0.13, 95% CI [0.03, 0.24]). Parent catastrophizing, however, was not related to anxiety, nor was parent SOP (directly or indirectly; *a2b2* = -0.03, 95% CI [-0.08, 0.02]).

These results were largely maintained in model 2 wherein the mediator was replaced with fear of pain scores. Youth SOP accounted for 3% of the variance in fear of pain, and parent SOP accounted for 6% of the variance in parent fear of pain. The whole model accounted for 40% of the variance in youth anxiety scores. Youth SOP and fear of pain each directly contributed to anxiety; however, no indirect effect occurred (*a1b1* = 0.08, 95% CI [-.03, .17]). Conversely, parent SOP and fear of pain were not related to anxiety and no indirect effect occurred (*a2b2* = -0.02, 95% CI [-0.07, 0.03]).

Model 3, which examined pain catastrophizing as the mediator and youth

depression scores as the outcome, was a slightly stronger model. As in model 1, youth and parent SOP each accounted for 6% of the variance in their respective catastrophizing scores. This model predicted 40% of the variance in youth depression scores, which was explained through significant positive relationships between youth SOP and depression and youth catastrophizing and depression, a significant negative relationship between parent SOP and depression, and an indirect effect of youth SOP through youth catastrophizing (*a1b1* = 0.12, 95% CI [.03, .22]). Parent catastrophizing was not related to depression, and no indirect effect occurred (*a2b2* = 0.03, 95% CI [-.01, .10]).

Similarly, in model 4 which included fear of pain as the mediator, youth and parent SOP contributed to 3% and 6% of their respective fear of pain scores. The model predicted 42% of the variance in youth depression scores, which was positively predicted by youth SOP and youth fear of pain, and negatively by parent SOP. Parent fear of pain was not related to youth depression scores, and the indirect effects of youth (*a1b1* = 0.06, 95% CI [-.03, .14]) and parent (*a2b2* = 0.02, 95% CI [-.02, .07]) SOP through fear of pain were not significant.[1]

3.4.4. Hypothesis 2: Self-Concealment Mediating the Relationships between Youth/Parent Perfectionism and Youth Anxiety and Depression

Two path analyses tested the above-mentioned hypothesis with youth anxiety and depression scores as the outcomes. Given the moderately large covariances (*β*s from .20 to .49) observed between predictors, each model was followed up with exploratory mediations to understand the relationships for each predictor independently, as

[1] Partner effects were explored between the predictors and mediators in each of these models, though these paths were not preregistered. None of these pathways were significant.

collinearity might be obscuring interesting patterns in these data.

In model 5 (Figure 3.2), neither youth SOP, youth SPP, or parent OOP were significantly related to youth self-concealment when entered in as simultaneous predictors, though they collectively accounted for 5% of the variance. This model predicted 22% of the variance in youth anxiety; however, only paths from youth SOP and self-concealment to anxiety were significant. Youth SOP, youth SPP, and parent OOP did not indirectly contribute (*a1b1* = -0.01, 95% CI [-0.07, 0.04]; *a2b1* = 0.05, 95% CI [-0.01, 0.11]; *a3b1* = 0.02, 95% CI [-0.03, 0.08], respectively). In exploratory mediations, one perfectionism variable was entered at a time. Youth self-concealment remained a significant predictor of anxiety across all models; however, only youth SPP was significantly related to self-concealment. Direct effects were observed for both youth SOP and youth SPP in predicting youth anxiety, and an indirect effect emerged for youth SPP through self-concealment, *ab* = 0.05, 95% CI [0.01, 0.11]. No other results were significant, including the indirect effects for youth SOP (*ab* = .03, 95% CI [-0.02, 0.08]) and parent OOP (*ab* = 0.04, 95% CI [-0.02, 0.10]).

In model 6 (Figure 3.3), the same patterns were observed. While perfectionism accounted for 5% of the variance in youth self-concealment, and the model predicted 20% of the variance in youth depression, only paths for youth SPP and self-concealment in predicting depression were significant. Youth SOP, youth SPP, and parent OOP did not indirectly contribute to depression via youth self-concealment (*a1b1* = -0.01, 95% CI [-0.07, 0.04]; *a2b1* = 0.05, 95% CI [-0.01, 0.11]; *a3b1* = 0.02, 95% CI [-0.03, 0.08], respectively). Exploratory mediations confirmed a significant relationship between youth self-concealment and depression in each model. Youth SPP directly predicted self-

concealment and depression, and indirectly predicted depression through self-concealment (*ab* = 0.05, 95% CI [0.004, 0.11]). No other significant pathways emerged, including indirect effects with youth SOP (*ab* = 0.03, 95% CI [-0.02, 0.08]) and parent OOP (*ab* = 0.04, 95% CI [-0.02, 0.11]) as predictors.

3.5. Discussion

Finding ways to reduce internalizing symptoms in youth with JIA is of utmost importance. This involves identifying and curtailing risk factors, and enhancing protective and promotive factors (Zimmerman, 2013). While some disease-specific risk factors have been identified (e.g., disease activity, disease burden, pain; see Fair et al., 2019), other risk factors, such as perfectionism, that have been identified in the broader literature (Morris & Lomax, 2014), have not yet been explored amongst youth with JIA.

Using the two-factor CAPS measure, significant correlations were observed between youth (but not parent) dimensions of perfectionism and self-reported depression and anxiety symptoms. Hypotheses regarding the role of parent/youth perfectionism were partially supported in predicting these outcomes, suggesting the value of considering youth and parent perfectionism in the mental health of youth with JIA.

In hypothesis 1, not only was it found that youth and parent SOP were related (as expected based on the social learning model; Smith et al., 2022), SOP was associated with more negative self-evaluations (i.e., pain-related fears and catastrophic thoughts) in youth and parents, and greater SOP in youth predicted more internalizing symptoms (in part explained through more catastrophic thoughts about their pain). Interestingly, SOP in parents was predictive of fewer symptoms of depression in youth, although this was not explained through negative self-evaluations.

The findings observed for youth are in keeping with the broader literature. The relationships between SOP, pain catastrophizing, and fear of pain have been seen in youth with chronic pain (Randall, Smith, et al., 2018), and the relationships between dimensions of SOP and internalizing symptoms have been seen in both youth with IBD (Piercy et al., 2020) and nonclinical samples (Hewitt et al., 2002). The novel finding that youth SOP predicted internalizing symptoms in youth with JIA, in part through pain catastrophizing, suggests some support for the SCCAMPI model (i.e., that perfectionism may increase negative self-evaluations), and importantly a mechanism by which youth SOP may contribute to internalizing symptoms in youth with JIA. Although both pain catastrophizing and fear of pain (which are highly correlated; e.g., Simons et al., 2015) were used to capture the construct of negative self-evaluations, the lack of indirect effect observed for the latter may suggest that its assessment of fears may involve less of an evaluative component, thus serving as a weaker proxy variable. This difference is especially interesting given the high intercorrelations observed between these variables, which may have otherwise been suggestive of construct overlap.

The relationships observed between parent SOP and pain catastrophizing have also been seen in other studies (Randall, Smith, et al., 2018), as have the mixed relationships between parent SOP and youth internalizing symptoms (Cook & Kearney, 2009; Piercy et al., 2020) (i.e., while positive relationships have been observed, they are weaker and less robust in complex analyses). The negative relationship between parent SOP and youth depression was an intriguing finding, suggesting parent SOP in the context of JIA may be a protective factor. It may be that parents high in SOP have high standards for themselves pertaining to meeting their child's physical and psychosocial

needs, thus protecting against the symptoms of depression that youth with JIA may experience. Additional research is required to further explore this relationship. Finally, although the hypothesized indirect effects of parent SOP on youth internalizing symptoms through pain-related fears and catastrophizing were not supported, it is possible that other unmeasured mediators may be relevant (e.g., adolescent perceived pressure from parents; Randall et al., 2015), or that parent perfectionism may effect youth in unique ways (of relevance, the SCCAMPI model is not inherently dyadic and was developed based on adult health literature without regard for parents' contributions). Future research might explore how models of perfectionism in the contexts of illness (e.g., Molnar et al., 2016) and development (e.g., Affrunti & Woodruff-Borden, 2014) can be combined to better explain perfectionism in pediatric populations.

Hypothesis 2 explored the role of youth SOP and SPP and parent OOP in predicting anxiety and depression through increased youth self-concealment. While the full model was not supported, given the reduced sample size and the strong correlations amongst predictors, exploratory mediations tested each predictor separately to minimize loss of statistical power due to collinearity. Youth SOP and SPP (but not parent OOP) directly predicted youth internalizing symptoms in these models; however, only youth SPP predicted self-concealment and indirectly predicted internalizing symptoms through self-concealment. Logically, SPP is most relevant, as when youth perceive that others demand perfection of them, they may uphold their façade of perfection even in the context of JIA pain, thereby contributing to internalizing symptoms. This is consistent with findings from a recent qualitative study, demonstrating the presence of pain-related stigma and concealment in JIA populations (Wakefield et al., 2023). Similar results have

been observed in healthy adults (Williams & Cropley, 2014) and are consistent with longitudinal research on university students suggesting that perfectionistic concerns (akin to SPP) lead to increased efforts to present oneself as perfect, rather than the reverse (Mackinnon & Sherry, 2012). Comparatively, youth high in SOP may not experience that same pressure to conceal their imperfections, and youth with parents high in OOP may not perceive their parents' perfectionism as targeted towards them (as the items assessing OOP in the BTPS were not specifically about their child, as has been seen with other measures being adapted based on the dyad member in question; Mackinnon et al., 2012) .

Together, these findings support the consideration of youth and parent perfectionism in understanding the mental health of youth with JIA, particularly when pain catastrophizing and self-concealment are observed. Although further research is warranted, directly assessing for and targeting trait perfectionism, alongside other concerns such as pain and internalizing symptoms, may be beneficial (Flett & Hewitt, 2014; Morris & Lomax, 2014; Randall, Gray, et al., 2018). Clinically treating perfectionism can yield benefits for internalizing symptoms, and if left out may dampen the effects of prevention and treatment efforts (Galloway et al., 2022). Interestingly, perfectionism may have a silver-lining in facilitating the uptake of treatments. Randall and colleagues (2020) explored the role of pre-treatment perfectionism in youth participating in intensive interdisciplinary pain treatment. While perfectionism was associated with worse outcomes pre-treatment, these youth also had lower pain catastrophizing and pain severity scores post-treatment. Similarly, Piercy and colleagues (2020) found that higher SOP-striving in youth with IBD was associated with greater disease self-management.

The strengths of this study include exploration of a relatively novel risk factor both in the pediatric pain and JIA literatures, use of parent/youth data, preregistration of a priori hypotheses, open data, and exploration of the mechanisms by which perfectionism impacts youth mental health. Study findings should nevertheless be interpreted in the context of their limitations. Limitations include the use of an Internet survey design, self-report data (e.g., it is possible one does not know their disease characteristics or diagnosis), low completion rate, potential sampling bias, and threats to data validity given the increase in online fraudulent activity (Zhang et al., 2022). Best available evidence was nevertheless used to prevent, manage, and screen out fraudulent responses (Teitcher et al., 2015). Given the challenges with recruitment, the sample size was lower than anticipated, preventing the exploration of covariates (e.g., age, sex, disease activity, proximity to parents) and potentially limiting the ability to detect significance for the parent pathways. A further limitation was the types of scales used. Had the 3-factor model of the CAPS measure fit these data, the SOP-critical dimension would have been the hypothesized predictor. As such, it is possible that non-significant results were observed given the inclusion of items measuring both SOP-critical and SOP-striving. The use of proxy measures to assess "negative self-evaluations" (i.e., pain catastrophizing and fear of pain) is also a limitation. In the absence of a better measure, however, both constructs may be interpreted as a negative self-evaluation, given the maladaptive and intrusive cognitions and fears about pain involved, which may prevent one from reaching their excessively high standards. The limited sample size prevented the exploration of all pathways in the SCCAMPI model. While other mediators may be interesting to explore (e.g., there may be similarities between pain and stress), this study prioritized

understanding how the intrapsychic and interpersonal dynamics of individuals with perfectionism may present in unique ways in youth with JIA and their caregivers, thus contributing to worse outcomes. Finally, given the cross-sectional design temporal precedence could not be established. Although perfectionism is conceptualized as a trait that emerges early in childhood, longitudinal designs are needed to confirm causality.

Future research would benefit from overcoming the abovementioned limitations. Further exploration of the SCCAMPI is also warranted (e.g., application to pediatric populations, assessing other pathways of perceived control and social support, measurement of other mediators such as coping and stress, measurement of other outcomes such as pain and functioning). Finally, while perfectionism is generally observed to be a risk factor in these data, the possibility that some domains of perfectionism (i.e., SOP-strivings, SOP in parents) may have adaptive components warranting further exploration.

In conclusion, this study identified various direct and indirect relationships between youth and parent perfectionism and anxiety and depression in youth with JIA. While future research would benefit from further exploring these relationships with larger samples and longitudinal designs, screening for perfectionistic tendencies in youth with JIA and their parents may be beneficial.

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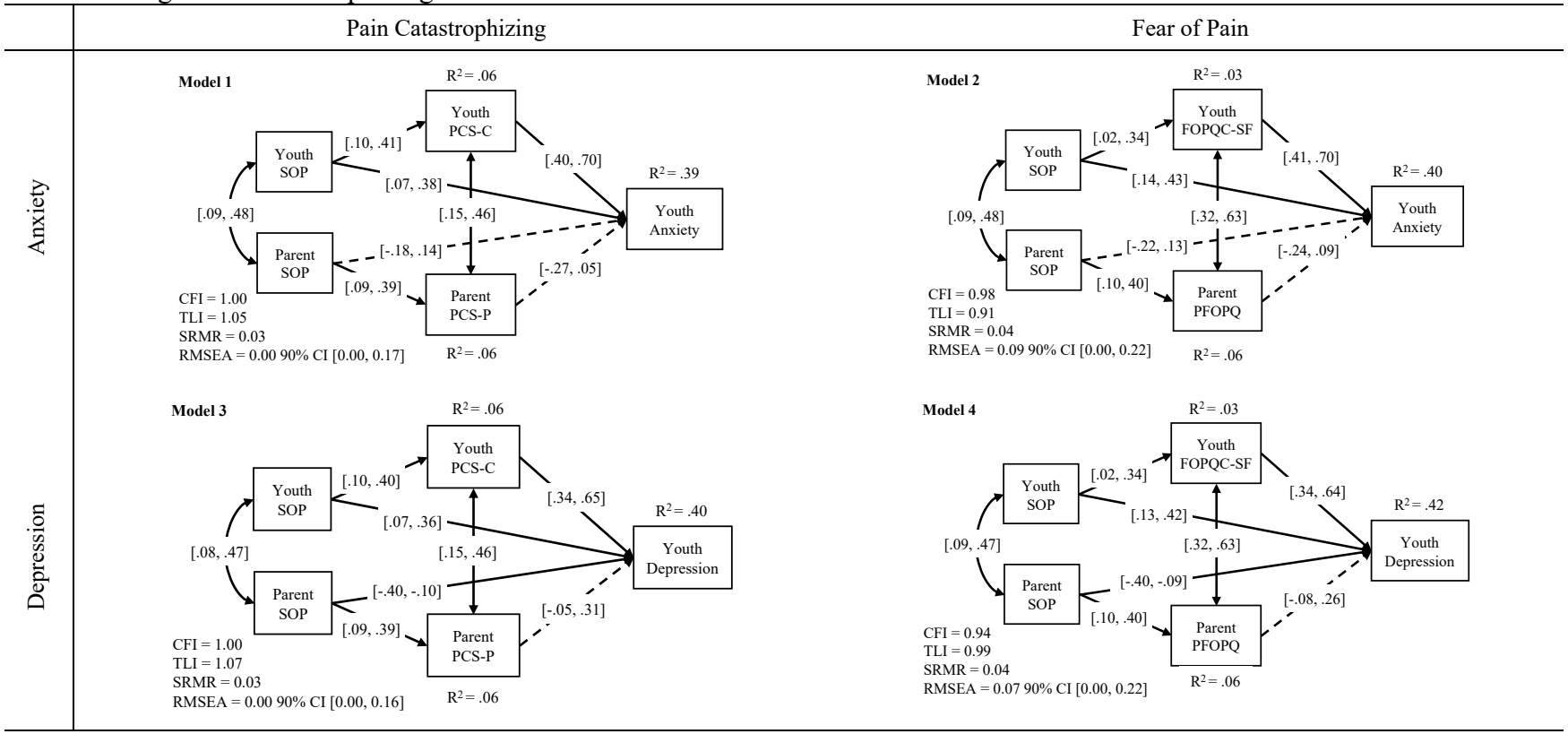
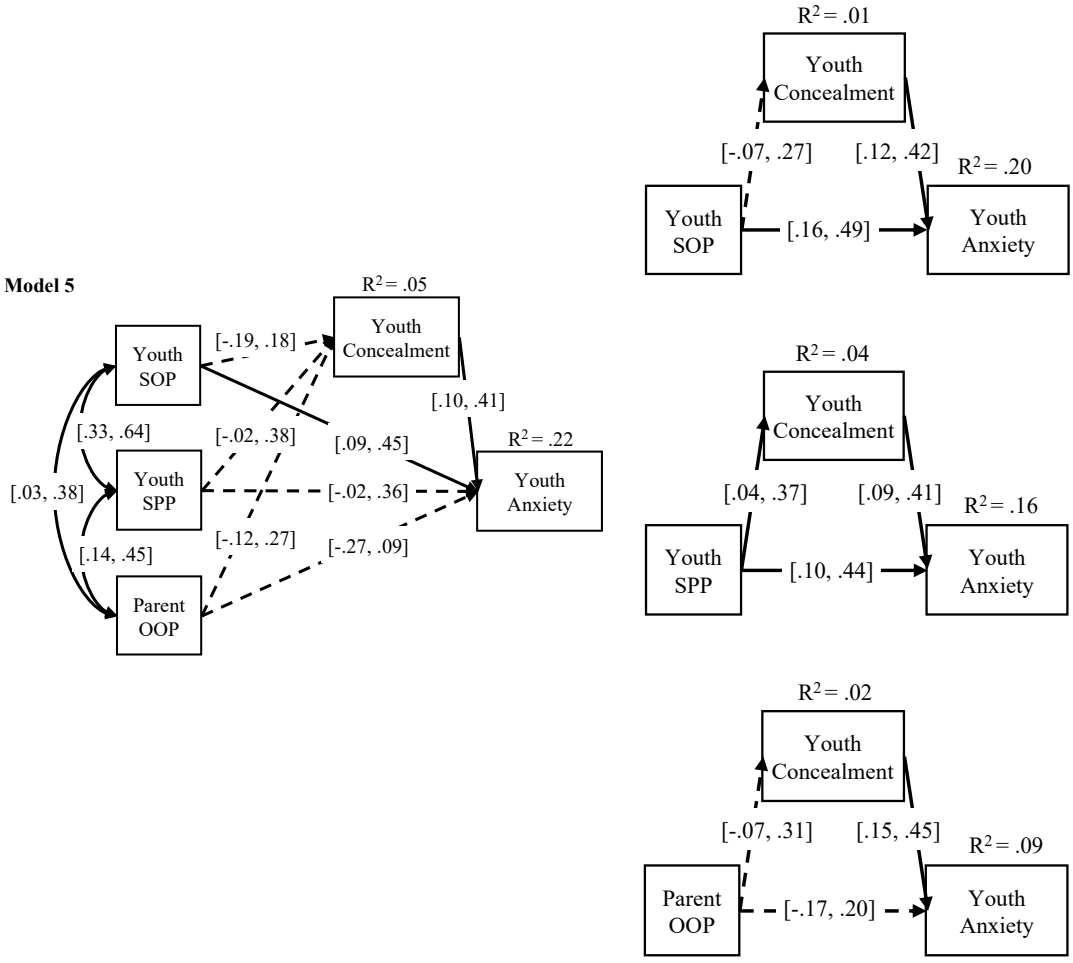
3.9. Figures

Figure 3.1. Path Analyses for Hypothesis 1 Predicting the Effects of Youth/Parent Self-Oriented Perfectionism on Youth Mental Health Through Pain Catastrophizing and Fear of Pain



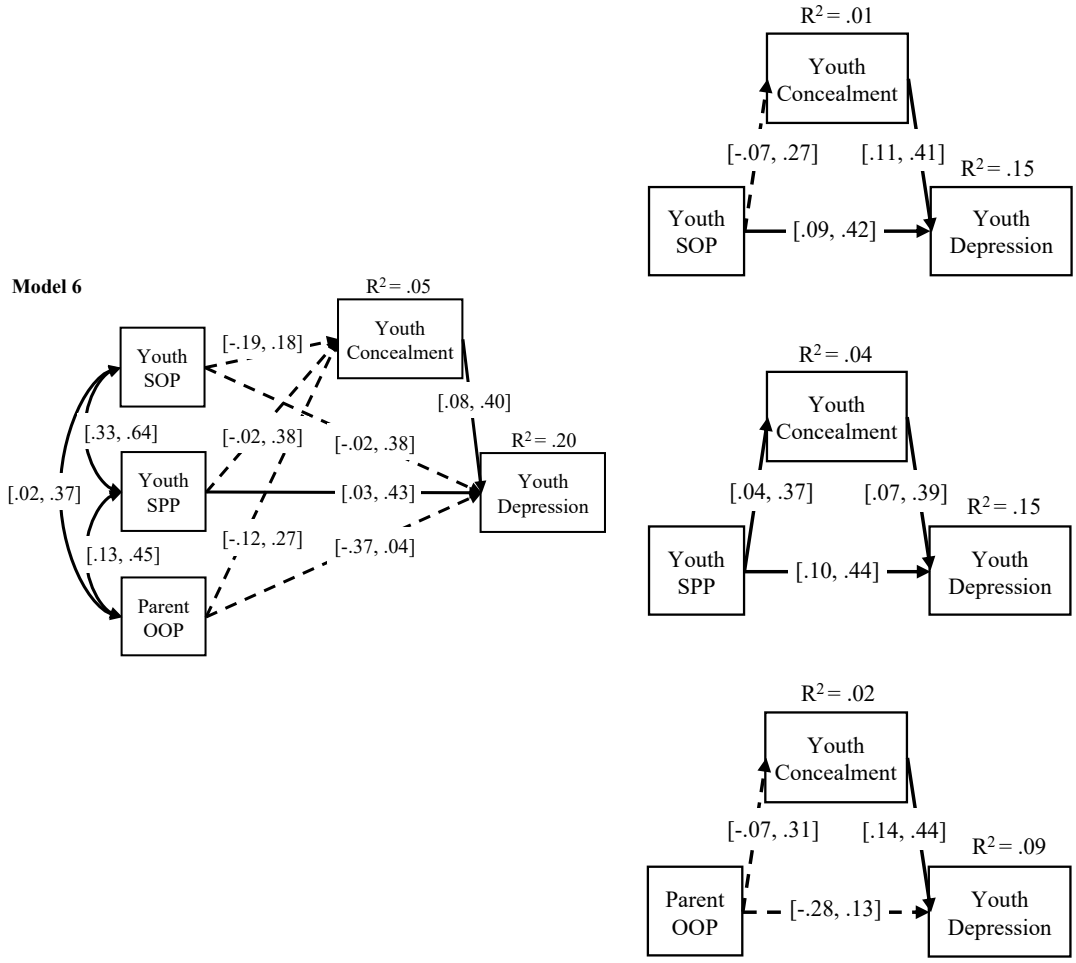
Note. The 95% confidence intervals for standardized coefficients are reported. Solid lines represent $p < .05$. Arrows pointing towards the factor represent residual variance. CFI = Robust Comparative Fit Index; FOPQC-SF = Fear of Pain Questionnaire Child – Short Form; PCS-C = Pain Catastrophizing Scale for Children; PCS-P = Pain Catastrophizing Scale for Parents; PFOPQ = Parent Fear of Pain Questionnaire; RMSEA = Root Mean Square Error of Approximation; SOP = Self-Oriented Perfectionism; SRMR = Standardized Root Mean Square Residual; TLI = Robust Tucker-Lewis Index.

Figure 3.2. Path Analyses for Hypothesis 2 and Exploratory Mediations with each Predictor Predicting the Effects of Perfectionism on Youth Anxiety Through Self Concealment



Note. The 95% confidence intervals for standardized coefficients are reported. Solid lines represent $p < .05$. Arrows pointing towards the factor represent residual variance. OOP = Other-Oriented Perfectionism; SOP = Self-Oriented Perfectionism; SPP = Socially Prescribed Perfectionism.

Figure 3.3. Path Analyses for Hypothesis 2 and Exploratory Mediations with each Predictor Predicting the Effects of Perfectionism on Youth Depression Through Self Concealment



Note. The 95% confidence intervals for standardized coefficients are reported. Solid lines represent $p < .05$. Arrows pointing towards the factor represent residual variance. OOP = Other-Oriented Perfectionism; SOP = Self-Oriented Perfectionism; SPP = Socially Prescribed Perfectionism.

3.10. Tables

Table 3.1. Descriptive and Medical Variables

Demographics and Medical Variables	Parent (n=140) *N* (%) or *M* ± *SD* (Min, Max)	Youth (n=129) *N* (%) or *M* ± *SD* (Min, Max)
Participant Demographics		
Age (Years)	45.24 ± 4.87 (33, 57) [d]	15.29 ± 1.62 (13, 18) [a]
Sex (Female) [a]	--	110 (70.5)
Gender [a]		
Mother/Girl	148 (94.9)	106 (67.9)
Father/Boy	8 (5.1)	46 (29.5)
Other (transgender, nonbinary, gender fluid)	--	4 (2.5)
Ethnicity [b]		
Aboriginal	7 (4.5)	9 (5.8)
Black	3 (1.9)	3 (1.9)
East/Southeast Asian	3 (1.8)	3 (1.9)
South Asian	4 (2.6)	4 (2.6)
White	123 (78.8)	110 (70.5)
Other (Jewish, West Asian, Latin American)	5 (3.2)	4 (2.6)
Prefer not to answer	2 (1.3)	1 (0.6)
Country of Residence		
Canada	96 (68.6)	95 (74.2)
United Kingdom	24 (17.1)	19 (14.8)
USA	16 (11.4)	11 (8.6)
Other (Ireland, South Africa, Australia)	4 (2.8)	3 (2.4)
Income (in CAD)		
<$50,000	16 (11.5)	--
$50,000 – 99,999	42 (30.0)	--
$100,000 – 149,999	29 (20.7)	--
>$150,000	36 (25.7)	--
Prefer not to answer	17 (12.1)	--
Youth Medical Characteristics		
Diagnosis [a e]		
Polyarticular Arthritis	--	37 (23.9)
Enthesitis-Related Arthritis	--	27 (17.4)
Oligoarticular Arthritis	--	26 (16.8)
Systemic Arthritis	--	17 (11.0)
Psoriatic Arthritis	--	10 (6.5)
Undifferentiated Arthritis or Unknown [c]	--	38 (24.5)
Age at Diagnosis [a f]		8.09 ± 4.72 (0, 16)
Current Disease Activity (Active/Flare)	87 (62.1)	75 (59.1) [g]
Pain Severity – Current VAS (0-10)	3.45 ± 3.04 (0, 10)	2.80 ± 2.90 (0, 10) [g]
Pain Severity – Usual VAS (0-10)	4.93 ± 2.15 (0, 10)	4.96 ± 2.23 (0, 10) [h]
Pain Frequency – Past month		
Not at all	27 (19.3)	26 (20.5) [g]
1-14 days	38 (27.2)	37 (29.1) [g]
15-28 days	26 (18.6)	14 (11.0) [g]
Daily	49 (35.0)	50 (39.4) [g]
Pain – Currently experiencing chronic pain	50 (35.7)	50 (39.4) [h]

Note. Parent data are used for parent demographics, and parent report data were used for youth medical characteristics. Percent was calculated based on number of participants who completed the question rather than the total N. [a] Data from parents/youth were combined to achieve an N=156. Data that matched were used when possible. If not possible, youth data was used before parent data for demographic information, and parent data was used before youth data for medical information. [b] Participants could select more than one response. [c] Three participants indicated also having a diagnosis of autoimmune arthritis, juvenile dermatomyositis, and scleroderma. [d] n=139. [e] n=155. [f] n=152. [g] n=127. [h] n=125.

Table 3.2. Descriptive Statistics and Correlations for Study Variables

	Variable	1	2	3	4	5	6	7	8	9	10	11
1	Youth Anxiety (self-report)	--										
2	Youth Depression (self-report)	.73***	--									
3	Youth Self-Oriented Perfectionism	.35***	.28**	--								
4	Youth Socially Prescribed Perfectionism	.32***	.32***	.49***	--							
5	Parent Self-Oriented Perfectionism	.11	-.07	.29**	.14	--						
6	Parent Other-Oriented Perfectionism	.05	-.04	.20*	.30***	.63***	--					
7	Youth Pain Catastrophizing	.57***	.55***	.25**	.27**	.17	.12	--				
8	Parent Pain Catastrophizing	.08	.21*	.05	-.03	.27**	.24**	.31***	--			
9	Youth Fear of Pain	.55***	.54***	.14	.15	.17	.12	.66***	.33***	--		
10	Parent Fear of Pain	.15	.23*	-.04	.05	.30***	.33***	.31***	.71***	.47***	--	
11	Youth Self-Concealment	.30***	.28**	.10	.20*	-.03	.13	.22*	.06	.30***	.20*	--
	n	126	126	126	126	139	139	126	138	126	138	126
	M	12.85	11.56	3.36	2.51	2.58	1.97	1.49	1.61	1.66	1.19	3.32
	SD	8.23	6.82	0.82	0.86	0.90	0.67	1.01	0.88	0.90	0.74	1.08
	Alpha	.89	.91	.91	.90	.88	.84	.95	.94	.89	.94	.90

Note. **Correlation is significant at the <.001 level. **Correlation is significant at the .01 level. *Correlation is significant at the .05 level (2 tailed).

3.11. Supplementary Materials

Supplementary File 1

Exploratory Factor Analysis of the original Two-Factor Child and Adolescent Perfectionism Scale (CAPS)

	SOP	SPP
1. I try to be perfect in everything I do	**0.75**	-0.04
2. I want to be the best at everything I do	**0.68**	-0.09
4. I feel that I have to do my best all the time	**0.62**	0.13
6. I always try for the top score on a test	**0.67**	-0.14
7. It really bothers me when I don't do my best all the time	**0.73**	0.00
9. I don't always try to be the best*	**0.40**	-0.10
11. I get mad at myself when I make a mistake	**0.53**	0.10
14. I get upset if there is even one mistake in my work	**0.74**	-0.01
16. When I do something, it has to be perfect	**0.78**	0.10
18. I do not have to be the best at everything I do*	**0.59**	0.11
20. Even when I pass, I feel that I have failed if I didn't get one of the highest marks in the class	**0.70**	0.01
22. I can't stand to be less than perfect	**0.82**	0.02
3. My parents don't always expect me to be perfect in everything I do*	-0.15	**0.46**
5. There are people in my life who expect me to be perfect	-0.13	**0.82**
8. My family expects me to be perfect	0.03	**0.66**
10. People expect more from me than I am able to give	-0.10	**0.67**
12. Other people think I have failed if I do not do my very best all the time	0.04	**0.75**
13. Other people always expect me to be perfect	0.01	**0.83**
15. People around me expect me to be great at everything	0.10	**0.80**
17. My teachers expect my work to be perfect	0.26	**0.50**
19. I am always expected to do better than others	0.31	**0.53**
21. I feel that people ask too much of me	0.03	**0.61**

Note. *Reverse coded. Extraction method = maximum likelihood; Rotation method = Oblimin with Kaiser normalization; Factor loadings >.4 are bolded. SOP-Striving = self-oriented perfectionism – Striving; SPP = socially prescribed perfectionism; SOP-Critical = self-oriented perfectionism - Critical. The items are from "The child–adolescent perfectionism scale: Development, validation, and association with adjustment," by G. L. Flett, P. L. Hewitt, D. J. Boucher, L. A. Davidson, and Y. Munro, 1997. Copyright 1997 by G. L. Flett, P. L. Hewitt, D. J. Boucher, L. A. Davidson, and Y. Munro.

CHAPTER 4: EXPLORING PAIN ADAPTATION IN YOUTH WITH JUVENILE IDIOPATHIC ARTHRITIS: IDENTIFYING YOUTH AND PARENT RESILIENCE RESOURCES AND MECHANISMS

The manuscript based on this study is detailed below. Yvonne Brandelli, under the supervision of Drs. Christine Chambers and Sean Mackinnon, was responsible for developing the research question, methodology, and analytic approach; and obtaining ethical approval. She developed the study protocol and data collection procedures, recruited participants, led the data analysis and interpretation with the support of her supervisors and co-authors, and made the data publicly available after publication. Ms. Brandelli wrote the initial draft of this manuscript and received and incorporated feedback from the study co-authors. The manuscript was submitted to Arthritis, Care, and Research on April 4th, 2024. The current reference for this manuscript is:

Brandelli, Y. N., Mackinnon, S. P., Chambers, C. T., Parker, J. A., Huber, A. M., Stinson, J. N., Johnson, S. A., & Wilson, J. P. (Submitted). Exploring pain adaptation in youth with juvenile idiopathic arthritis: Identifying youth and parent resilience resources and mechanisms. *Arthritis, Care, and Research.*

4.1. Abstract

Objective: Although juvenile idiopathic arthritis (JIA) is often associated with pain, this experience does not necessitate negative outcomes (e.g., depression, functional impairment). Little research has explored youth and parent resilience resources (i.e., stable traits) and mechanisms (i.e., dynamic processes) in this context, and studies have focused on their contributions independently rather than collectively. This study, informed by the Ecological Resilience-Risk Model in Pediatric Pain, sought to: 1) explore the relationships amongst youth and parent resilience resources and mechanisms; and 2) identify the relative importance (RI; i.e., independent contributions when entered simultaneously) of evidence-based youth and parent resources and mechanisms in contributing to recovery, sustainability, and growth outcomes.

Methods: Youth (13-18 years) with JIA and their parents (156 dyads) completed a battery of online questionnaires assessing resilience resources (optimism, resilience), mechanisms (psychological flexibility, pain acceptance, self-efficacy), recovery/sustainability (pain intensity, functional disability, health-related quality of life), and growth (benefit finding) outcomes.

Results: Analyses demonstrated significant positive correlations across within-person resources and mechanisms, and weaker correlations across within-dyad resources and mechanisms. Although the RI of predictors varied by outcome, youth pain acceptance was the most robust predictor across models (RI = .03 - .15). Parent optimism and psychosocial self-efficacy were also important. Some effect sizes shrank close to zero once adjusting for other variables in the analyses, suggesting construct overlap.

Conclusions: While additional research is needed to further understand resilience, results highlight the importance of fostering pain acceptance in youth and incorporating parents in psychosocial interventions to optimize living with JIA.

4.2. Significance and Innovations

- This study assessed the relative importance of youth and parent resilience resources and mechanisms to advance knowledge as it pertains to JIA pain.
- Most youth resilience resources and mechanisms were significantly related to one another, as were most parent resilience resources and mechanisms. Relationships between youth and parent resilience resources and mechanisms were less likely to be significant.
- Across surrogate markers of pain adaptation, youth pain acceptance was one of the most robust predictors. Parent contributions such as optimism and psychosocial self-efficacy also played an important role.
- To promote resilience in the context of JIA, results highlight the importance of fostering youth pain acceptance and incorporating parents in the psychosocial interventions provided.

4.3. Introduction

The hallmark experience of juvenile idiopathic arthritis (JIA) is pain (Canadian Paediatric Society, 2009) which has been identified as a top research priority for families (Correll et al., 2020). To date, research has focused on negative outcomes associated with JIA pain (e.g., internalizing symptoms, lower health-related quality of life, impaired social functioning; Brandelli et al., 2023); however, the experience of pain and the presence of risk factors does not guarantee that youth with JIA will endure the abovementioned negative outcomes. There is individual variation in pain experiences (Stinson et al., 2011), which is likely due to the presence of promotive and/or protective factors (i.e., factors that have a positive and direct influence on outcomes regardless of the presence of risk factors, and factors that can dampen the presence of risk factors, respectively; Zimmerman, 2013).

The study of promotive and protective factors is encompassed within the resilience literature. Although a complex, systemic, and dynamic process without a universal definition, resilience can be conceptualized as the capacity of a dynamic system to adapt successfully to disturbances (such as a diagnosis of JIA) that threaten system function, viability, or development (Masten, 2014). There is a growing need within the JIA and pediatric pain literature to further this field of study (Cousins, Kalapurakkel, et al., 2015), particularly as the aim of many treatments is to manage pain and prevent irreversible damage rather than "cure" the disease (Beukelman et al., 2011). Thus, by shifting emphasis to understanding and promoting the conditions necessary for resilience, youth can be protected from unfavorable outcomes and learn to optimize living in the face of adversity.

Although there is no unified outcome of resilience, Sturgeon and Zautra (2010) theorized that pain adaptation can be measured in terms of one's recovery (i.e., resumed functioning; psychological, physical, or academic well-being), sustainability (i.e., perseverance with valued activities), and growth (i.e., new learning or a better understanding of one's capabilities). These are nevertheless surrogate markers of adaptation (Rosenberg & Yi-Frazier, 2016), as it is a process that depends on the individual and their context, and it is unclear which of the many outcomes are necessary or sufficient to determine successful adaptation. Cousins, Kalapurakkel, and colleagues (2015) tailored this model for pediatric populations, placing greater emphasis on the ecological system. Specifically, these outcomes are the result of an interaction between resilience resources and risk factors (i.e., stable traits such as optimism) and resilience and risk mechanisms (i.e., dynamic processes such as pain acceptance) that occur within and between the individual, their family/social environment, and their culture and time.

There is preliminary support for components of the Ecological Resilience-Risk Model in Pediatric Chronic Pain in the broader literature. In terms of resources, trait optimism (i.e., having favorable expectations for the future) predicts improved health-related quality of life (HRQoL) directly in youth with abdominal pain (Tomlinson et al., 2021) and through reduced fear and catastrophizing in youth with chronic pain (Cousins, Cohen, et al., 2015). Trait resilience (i.e., a general disposition of bouncing back) is associated with reduced disease severity, pain, and disability, and greater HRQoL (Gmuca et al., 2019). There is also preliminary support for other resources, including mindfulness (Wright et al., 2021), positive affect (Beeckman et al., 2020), and positive peer relationships (Forgeron et al., 2011).

By way of mechanisms, psychological flexibility, or the ability to be present-focused and engaged in values-based action, is associated with less daily activity avoidance in youth with chronic pain (Beeckman, Simons, et al., 2019; Beeckman et al., 2020), and in parents is positively associated with youth HRQoL in some (S. Lee et al., 2020) but not all (Wright et al., 2021) studies. Support also exists for pain acceptance as a resilience mechanism. In the context of pediatric pain rehabilitation programs, increases in acceptance are predictive of a decrease in depressive symptoms, catastrophizing, and functional disability (Weiss et al., 2013). More broadly, youth pain acceptance is positively associated with HRQoL (S. Lee et al., 2020; Wright et al., 2021) and negatively associated with pain intensity (S. Lee et al., 2020), and parent pain acceptance is indirectly associated with decreases in pain interference and increases in mobility through youth pain acceptance (Feinstein et al., 2018). Finally, self-efficacy, or one's belief in their ability to function effectively in the presence of pain or disease, also contributes to pain acceptance (S. Lee et al., 2020), psychological flexibility (S. Lee et al., 2020), HRQoL (S. Lee et al., 2020), reduced pain intensity (S. Lee et al., 2020), reduced disability (Kalapurakkel et al., 2015), and fewer depressive symptoms (Kalapurakkel et al., 2015).

Despite this literature, these constructs have only been minimally applied to the context of JIA. Hynes et al. (2019) systematically reviewed the risk and resilience resources and mechanisms in the JIA literature. By way of resources, they found that family dysfunction is associated with lower hope in the context of youth with JIA (Connelly, 2005), and that child perceived social support is associated with better HRQoL (Seid et al., 2014). By way of mechanisms, self-efficacy is associated with

greater HRQoL (Seid et al., 2014) and less functional disability (Barlow et al., 2000, 2001; Sawyer et al., 2004); child psychological flexibility is associated with improved quality of life (QoL) and HRQoL (Feinstein et al., 2011), improved psychosocial health (Beeckman, Hughes, et al., 2019), and reductions in negative affect (Beeckman, Hughes, et al., 2019); child pain acceptance is related to improved QoL (Feinstein et al., 2011), better psychosocial and physical health (Beeckman, Hughes, et al., 2019), and less negative affect and disability (Beeckman, Hughes, et al., 2019); and parent reports of child pain coping (problem solving) and self-efficacy are related to less functional disability (Sawyer et al., 2004).

Given this scant literature, numerous variables remain to be explored (e.g., parent optimism, trait resilience) (Cousins, Kalapurakkel, et al., 2015; Hynes et al., 2019). Moreover, much of the literature has used small samples, relied on proxy reports, and emphasized outcomes of HRQoL (Hynes et al., 2019). Studies have largely focused on resources and mechanisms independently, neglecting to focus on their relationships with one another and the broader sociocultural environment (Hynes et al., 2019; Knafl et al., 2015). As such, there is a need to identify the resilience resources and mechanisms that are relevant to this population in a holistic manner to better understand what to emphasize to optimize living with JIA (Cousins, Kalapurakkel, et al., 2015).

The aims of this study were to: 1) explore the relevance of, and relationships between, youth and parent resilience resources and mechanisms that have been identified in the broader literature (i.e., optimism, trait resilience, psychological flexibility, pain acceptance, and self-efficacy) in the context of JIA pain; and 2) explore their relative importance (RI; i.e., their independent contributions while simultaneously accounting for

other resources and mechanisms) in contributing to recovery/sustainability (i.e., pain intensity, functioning, HRQoL) and growth (i.e., benefit finding) outcomes. A priori hypotheses were that: 1) there would be significant, positive relationships amongst the resilience resources and mechanisms; and 2) resilience resources and mechanisms would predict positive adaptation in the presence of JIA pain; however, no a-priori predictions were made regarding which constructs would emerge as most important in the analyses.

4.4. Patients and Methods

4.4.1. Study Design

The data used for the current study were part of a larger dataset. Another study with a different research question (https://osf.io/79rwp?view_only=d2c8e3e9e6544b76be10c137b134a841), variables, and analyses has been submitted for publication elsewhere. Data and syntax for the present study are openly available through Open Science Framework (OSF; https://osf.io/8g29d/?view_only=e4b2e29701db4031a0ed1f17f8715249). This cross-sectional, Internet-based study was approved by the IWK Research Ethics Board (ref. 1026950) and complies with the Declaration of Helsinki.

Following best practice in patient engagement, a leader in the field (IJ) co-developed the patient partnership plan for this study. In addition to partnering with Cassie and Friends, a parent-led organization for families of children with rheumatic diseases (www.cassieandfriends.ca), two parents and one youth with JIA provided consultation, support, and feedback on this study from conceptualization through to dissemination. Partners were compensated following the Solutions for Kids in Pain compensation guidelines (https://kidsinpain.ca/wp-content/uploads/2021/03/SKIP-Patient-Partner-

Compensation-Guidelines-approved-Feb-10-2020-1.pdf).

4.4.2. Participants

Youth (13-18 years old) with a diagnosis of JIA and a parent/caregiver were recruited through online and social media platforms (e.g., arthritis and pain communities, Facebook advertisements, blog posts), previous studies, posters at rheumatology and pain clinics, the IWK Health research registry, and industry partnerships. Recruitment took place towards the end of the COVID-19 pandemic, between November 2021 and April 2023.

Of the 206 youth and parent dyads who consented online, 33 were ineligible given their diagnosis or age, and 17 stopped after providing consent. The final sample size was 156 unique dyads. Missing data was complex given the study design. Parents generally filled out the entire survey (n = 129, 82.7%), with a small number providing partial data or not completing the survey at all (n = 11, 7.0% and n = 16, 10.3%, respectively). Youth also generally filled out the entire survey (n = 122, 78.2%) with the minority providing partial or no data (n = 7, 4.5% and n = 27; 17.3%, respectively). All data, including partial data, was analyzed when possible. A sensitivity analysis was conducted in G*power using the final sample of 156 dyads, an alpha of .05 and power of .80. With 12 predictors, there is sufficient power to detect an overall R^2 of .12. When considering power for individual predictors, there is sufficient power to detect an f^2 of 0.051 or ΔR^2 = 0.05.

4.4.3. Measures & Procedures

Participants self-selected into this study. After completing an eligibility screening questionnaire, youth and parents were emailed unique survey links that contained a

consent form and a 45-minute battery of validated questionnaires. Questions probing background information measured ethnicity via fixed categories and open-ended responses (Canadian Institute for Health Information, 2022), demographic and medical variables (e.g., age, gender, diagnosis), and pain variables (Birnie, Hundert, et al., 2019). Resilience resources were assessed via measures of optimism (Ey et al., 2005; Scheier et al., 1994) and trait resilience in youth (Smith et al., 2008). Resilience mechanisms were assessed via measures of psychological flexibility (Greco et al., 2008; Timmers et al., 2019), pain acceptance (Gauntlett-Gilbert et al., 2019; Smith et al., 2015), and arthritis-specific self-efficacy (Barlow et al., 2000, 2001). Pain adaptation outcomes were assessed via the following youth-reported recovery, sustainability, and growth outcomes: usual pain intensity (Birnie, Hundert, et al., 2019), functioning (Walker & Greene, 1991), generic and rheumatology specific HRQoL (Varni et al., 2002) and benefit finding (Phipps et al., 2007). Table 4.1 outlines the list of measures, including their definitions, scaling, and psychometric properties. Items were averaged to create total scores with higher scores reflecting greater endorsement of the construct. Responses were mandatory; however, participants could select "*prefer not to answer*" (treated as missing data). Upon completion, participants received a $15 CAD online gift card and dyads were entered into a draw to win one of two pairs of $250 CAD gift cards.

4.4.4. Analyses

To ensure data validity, in addition to screening participants during the data collection stage (e.g., screening questionnaire, passwords, the prevention of multiple submissions from the same Internet Protocol address; Teitcher et al., 2015) data was also screened prior to analyses (e.g., review of attention checks, "spam trap" questions,

captchas).

Analyses were completed using the psych() and lavaan() packages in R (https://www.r-project.org/). Youth-parent dyads were paired, and total scores were calculated. Assumptions of normality were met. Regressions were run in structural equation modelling software and a Full Information Maximum Likelihood approach was used for missing data.

Descriptive statistics and bivariate correlations (Aim 1) were used to describe measures of pain, resilience resources and mechanisms, and outcomes. To address Aim 2, a series of five multiple regressions were tested through structural equation models using each of the 12 resources and mechanisms as predictors. RI was calculated with the Pratt Index (Pratt, 1987), the product of the bivariate correlation and standardized regression coefficients. This method partitions the total R^2 across all variables to quantify the relative importance of each predictor variable in a way that sums to the total R^2 value (e.g., if the total R^2 is 0.10 and a single RI value is 0.05, that predictor accounts for 5% of the variance in the outcome and 50% of the total R^2 value). Standardized correlation (r) and regression coefficients (β), their respective p-values, confidence intervals (CI), the total variance predicted by each model, and the Pratt Relative Importance Index (RI) are reported. As our study has 80% power to detect ΔR^2 of 0.05 or larger, and because the effect size observed in much of the psychological literature is .21 or $R^2 = .044$ (Richard et al., 2003), RI values at or above .05 will generally be considered important. Given the large number of coefficients (r, β, RI), each of the 12 predictors will be classified into one of three categories for ease of exposition. Predictors coded as "Important" will have a RI ≥.05, and at least one statistically significant coefficient. Predictors coded as "Not

Important" will have a RI ranging between -.05 and .05, and no statistically significant coefficients. Predictors coded as "Inconclusive" will incorporate all other cases (i.e., RI < .05 with varying patterns in the statistical significance of coefficients). Definitive conclusions regarding "Inconclusive" predictors cannot be made as discrepancies may reflect the complexity of these constructs or a lack of statistical power.

Note that this scheme may oversimplify the results in exchange for ease of interpretation (Thomas et al., 1998). More complex patterns that might be observed include: (a) the *r* coefficient is significant but the β coefficient is not (suggestive of construct overlap), (b) the *r* coefficient is insignificant but the β coefficient is significant (suggesting that outcome-irrelevant variance has been removed by the other included predictors), or (c) a predictor has a positive *r* coefficient but a negative β coefficient (or vice versa) which is akin to a suppressor variable (Thomas et al., 1998) which may enhance the predictive ability of other predictors in the model by accounting for some of their outcome-irrelevant variance. Such results will be described in-text to reflect these nuances.

4.5. Results

4.5.1. Descriptive Statistics

One hundred and fifty-six dyads participated, including 129 adolescents with JIA and 140 parents. Adolescents were generally female (67.9%) with a mean age of 15.29 (*SD* = 1.62 years). Similarly, 95% of parents were mothers, with a mean age of 45.24 (*SD* = 4.87 years). Youth were on average diagnosed at age 8.09 (*SD* = 4.72 years), most of whom had received a diagnosis of polyarticular (23.9%) or enthesitis-related (17.4%) arthritis. Over half of the sample was currently experiencing active disease by both youth

and parent report. Internal consistencies of measures ranged from adequate to excellent (.76 - .95). See Tables 4.2 and 4.3 for demographics and study variables.

4.5.2. Associations Between Youth and Parent Resilience Resources, Mechanisms, and Outcomes

Pearson correlation coefficients are presented in Figure 4.1. As hypothesized, correlations between the 12 resilience resources and mechanisms were generally positive (except for youth psychological inflexibility, an inverse score) and significant. Correlations between within-youth resources and mechanisms were all significant (except for symptom self-efficacy/pain acceptance), ranging from moderate (r = -.28, p <.05) to strong (r = .71, p < .001). Correlations between within-parent resources and mechanisms were also positive and significant (except for psychological flexibility/symptom self-efficacy), ranging from weak (r = .20, p < .05) to strong (r = .82, p < .001). Non-significant results were more likely to occur across dyad members.

A similar pattern was observed between resources/mechanisms and outcomes, wherein every youth predictor was significantly related to the outcomes of usual pain, functional disability, and HRQoL (benefit finding was not significantly related to pain acceptance or activity self-efficacy); however, parent predictors were less strongly, if at all, related.

4.5.3. Relative Importance of Resilience Resources and Mechanisms in Predicting Outcomes

Results of the five multiple regression analyses with the relative contributions of the 12 resilience resources and mechanisms across recovery, sustainability, and growth outcomes can be seen in Table 4.4. A summary of the findings and their coding is

depicted in Figure 4.2.

In Model 1, 29% of the variance in usual pain intensity was accounted for by the included predictors. Youth pain acceptance (RI = .12) was the most robust contributor to reduced pain intensity. Other important contributors included parent psychosocial self-efficacy (RI = .07) and parent optimism (RI = .05). Youth optimism was also important (RI = .05), but became nonsignificant once adjusting for all other variables. In Model 2, 42% of the variance in youth functional disability was accounted for, with youth activity self-efficacy (RI = .23), youth pain acceptance (RI = .11), and parent psychosocial self-efficacy (RI = .07) as the most robust and significant contributors. In Model 3, 59% of the variance in generic HRQoL was explained by the predictors. Youth activity self-efficacy (RI = .19), youth psychological flexibility (RI = .10), and youth pain acceptance (RI = .09) were the most important and robust predictors. Youth optimism (RI = .08) was also important, although it lost predictive ability with the inclusion of other variables. In Model 4, the predictors accounted for 47% of the variance in rheumatology specific HRQoL, with youth pain acceptance (RI = .15) as the most robust contributor. Youth activity and symptom self-efficacy (RI = .14 and .08, respectively), and parent psychosocial self-efficacy (RI = .06) were important correlates that lost predictive ability in the regression model. Finally, 28% of the variance in benefit finding was explained by predictors (Model 5). Youth optimism was the only robust contributor (RI = .12). Youth emotion self-efficacy and parent symptom self-efficacy were also important (RI = .05 each), albeit to a less robust degree.

4.6. Discussion

Resilience is a complex process involving an interaction of risk and resilience

resources and mechanisms at the individual, familial, and cultural levels resulting in diverse outcomes pertaining to pain adaptation (Cousins, Kalapurakkel, et al., 2015). This study applied a novel approach to this construct in the context of JIA pain to explore the synergy between evidence-based youth and parent resilience resources and mechanisms and how they collectively interact and contribute to proxy measures of pain adaptation.

Significant correlations were observed between resilience resources and mechanisms, especially within individuals. This supports the notion that these variables conceptually align as resilience (rather than risk) resources and mechanisms. Moreover, given their moderate to high correlations, this also emphasizes the need to determine which variables are the most important predictors of key outcomes (Aim 2). As an example, large correlations were observed between youth optimism, trait resilience, and psychological flexibility (r = .62 - .69) which suggests construct overlap, and logically makes sense given their definitions (i.e., holding positive expectations for the future conceptually aligns with being able and willing to bounce back even when undesirable events occur and with maintaining the capacity to be present focused).

It was hypothesized the resilience resources and mechanisms would predict positive adaptation in the context of JIA pain across five surrogate markers of recovery, sustainability, and growth. While this was the case, the variables that were most relevant differed slightly based on the outcome in question. This is consistent with the literature emphasizing that the process of resilience, as well as the outcomes that are considered necessary and sufficient, are dependent on the individual and their context (Rosenberg & Yi-Frazier, 2016).

In Model 1, pain acceptance contributed the most to usual pain intensity. This was

closely followed by parent psychosocial self-efficacy and parent optimism which became more significant with the addition of other predictors, and youth optimism which became less significant. A recent systematic review conducted by our team explored the psychosocial factors associated with JIA pain (Brandelli et al., 2023). Although pain acceptance and optimism had not been identified, parent psychosocial self-efficacy emerged as a significant protective factor in 3/5 associations. Other psychosocial factors associated with reduced pain (albeit in fewer associations) included other domains of youth and parent self-efficacy, youth coping via distraction and positive self-statements, and select family factors (e.g., family achievement, activities, and cohesion). Interestingly, some of these factors were also significantly correlated with reduced pain intensity in this study; however, when other variables were included, their RI and predictive ability were less stable.

In Model 2, the RI of activity self-efficacy was double that of any other variable in predicting reduced functional disability. This is logical, as feeling capable of engaging in activities influences one's efforts and actions. Youth pain acceptance and parent psychosocial self-efficacy were also robust contributors to this model. While trait resilience, youth and parent psychological flexibility, and youth optimism were also relevant, their role was inconclusive in the regression. These findings are largely consistent with the literature showing support for these predictors independently (Feinstein et al., 2018; Gmuca et al., 2019; Kalapurakkel et al., 2015); although, there is mixed literature with regards to youth pain acceptance (Beeckman, Hughes, et al., 2019; Feinstein et al., 2011; Weiss et al., 2013) and psychological flexibility (Beeckman, Simons, et al., 2019; Beeckman et al., 2020; Feinstein et al., 2011), and limited evidence

exploring the synergy of these predictors. As such, to improve functional ability in youth with JIA, these findings emphasize the importance of youth prioritizing values-based actions and believing they can engage in activities in the presence of pain, and parents perceiving they are capable of psychosocially supporting their child in these endeavors.

Models 3 and 4 explored predictors of generic and rheumatology specific HRQoL. Both scores are comprised of subscales, including physical, emotional, social, and school functioning; and pain and hurt, daily activities, treatments, worry, and communication. Given the array of functional areas addressed, unsurprisingly many of the youth predictors (e.g., optimism, pain acceptance, psychological flexibility, self-efficacy) have independently predicted improvements in HRQoL in the literature (Beeckman, Hughes, et al., 2019; Cousins, Cohen, et al., 2015; Feinstein et al., 2011; S. Lee et al., 2020; Seid et al., 2014; Tomlinson et al., 2021; Wright et al., 2021), which was also seen in Models 3 and 4 to an extent. Interestingly, many youth predictors lost significance in these regression models. Activity-related self-efficacy, psychological flexibility, and pain acceptance were the most robust predictors, followed by optimism, symptom self-efficacy, and parent psychosocial self-efficacy which together remained significant and explained the greatest portion of variance.

In Model 5, the ability of youth to identify positive consequences of their arthritis was best predicted by youth optimism. Youth emotion self-efficacy and parent symptom self-efficacy were also important; but demonstrated collinearity. While benefit finding is a construct that has not received much attention in the JIA literature, it has been associated with optimism and self-esteem in pediatric oncology patients (Phipps et al., 2007). Interestingly, in pediatric chronic pain patients it has demonstrated an inverse

effect, wherein it was associated with reduced QoL and greater post-traumatic stress disorder, anxiety, and depression symptoms, which authors posited was due to the complex nature of living with chronic pain (Soltani et al., 2018). Although an inverse relationship between benefit finding and QoL was not seen in this study (all correlations were non-significant), it is possible that unlike a diagnosis of chronic pain which has been associated with a perceived lack of physician understanding (Meldrum et al., 2009), a diagnosis of JIA may facilitate the process of resilience. Given these discrepancies it will be critical for future research to further explore this outcome.

Despite these differences across models, key patterns emerged. There were generally large *r* coefficients which shrunk after controlling for other variables (suggesting construct overlap) with fewer instances of suppressor variables or *b* coefficients growing with the addition of other predictors. Youth pain acceptance was the most robust predictor across outcomes and was always among the top four contributors in terms of RI. Thus, a willingness to permit pain to be present and persist with valued activities is a key mechanism of change in fostering resilience, regardless of the outcome in question. This maps to the existing literature demonstrating its importance as a main effect or mediator (Beeckman, Hughes, et al., 2019; S. Lee et al., 2020; Weiss et al., 2013; Wright et al., 2021), with the added notion that it maintains its unique contributions even in the presence of other resources and mechanisms. Comparatively, some predictors consistently demonstrated weaker relationships to outcomes, including youth trait resilience (consistently demonstrated construct overlap); and parent pain acceptance and psychological flexibility (largely insignificant and classified as “Not Important”). Parent psychological flexibility and symptom self-efficacy were likely less important as parents

overall made weaker contributions to youth outcomes. It may be that a parent's capacity to stay present focused, engaged in values-based actions, and hopeful in their ability to manage their child's symptoms is developmentally less relevant for adolescents as they begin to increase independence and self-management. While unimportant in these analyses, they are likely more relevant for the parent's own adaptation. Moreover, at least one parent resilience resource or mechanism (often psychosocial self-efficacy) was significant across most models, indicating that parents have some influence on child outcomes.

These results generally support the Ecological Resilience-Risk Model in Pediatric Chronic Pain (Cousins, Kalapurakkel, et al., 2015) in the context of JIA, particularly for within youth resources and mechanisms. Moreover this study addressed numerous gaps in the literature (Hynes et al., 2019). Not only did this study identify the role of novel resilience resources and mechanisms in this population, it illuminated select constructs that hold greater weight in terms of one's pain adaptation, and demonstrated the ways in which many of these resources and mechanisms co-exist. Clinically, harnessing resilience to promote adaptation in the face of adversity aligns well with Acceptance and Commitment Therapy (Pielech et al., 2017). This study has demonstrated support for numerous protective and/or promotive factors; however, the factors most relevant for clinicians to focus on will largely depend on client goals.

Limitations of this study include the use of an Internet survey design, which resulted in reliance on self-report data for diagnosis and disease characteristics, low retention, and missing data. Akin to much of the existing literature, this study was also limited by its sample size. As such, covariates such as age, gender, and diagnosis

(Tomlinson et al., 2021); and other evidence-based resilience resources and mechanisms (e.g., trait mindfulness, positive affect) were not incorporated.

In addition to promoting data sharing and multi-site collaborations to increase sample sizes, there is a need to develop and validate scales to measure other resilience resources and mechanisms (e.g., committed action, self-regulation, sense of self; Cousins, Kalapurakkel, et al., 2015); incorporate perspectives from others within the child's network (e.g., siblings; Hynes et al., 2019); and statistically account for the biological, developmental, social, and cultural milieu to identify how these factors might interact with those identified in this study. Furthermore, more complex methodological and statistical approaches (e.g., longitudinal designs, profile analyses, network analyses, simultaneously mapping multiple latent variables as resources and mechanisms to a broader resilience factor) and the use of qualitative and/or mixed methods approaches (Hynes et al., 2019) would enrich our understanding of resilience in youth with JIA. Finally, there is value in rigorously testing the effects of strengths-based interventions (Cousins, Kalapurakkel, et al., 2015), particularly those incorporating the identified predictors.

Taken together, this study explored the relationships between and predictive ability of various youth and parent resilience resources and mechanisms in predicting pain adaptation in the context of JIA. In addition to demonstrating how predictors depend on the surrogate marker being used, an important pattern emerged wherein pain acceptance was one of the most robust predictors across outcomes, and parent contributions such as optimism and psychosocial self-efficacy also played an important role. These findings further support the Ecological Resilience-Risk Model in Pediatric

Chronic Pain and have important implications for the processes interventions should emphasize when helping youth and parents adjust to living with JIA.

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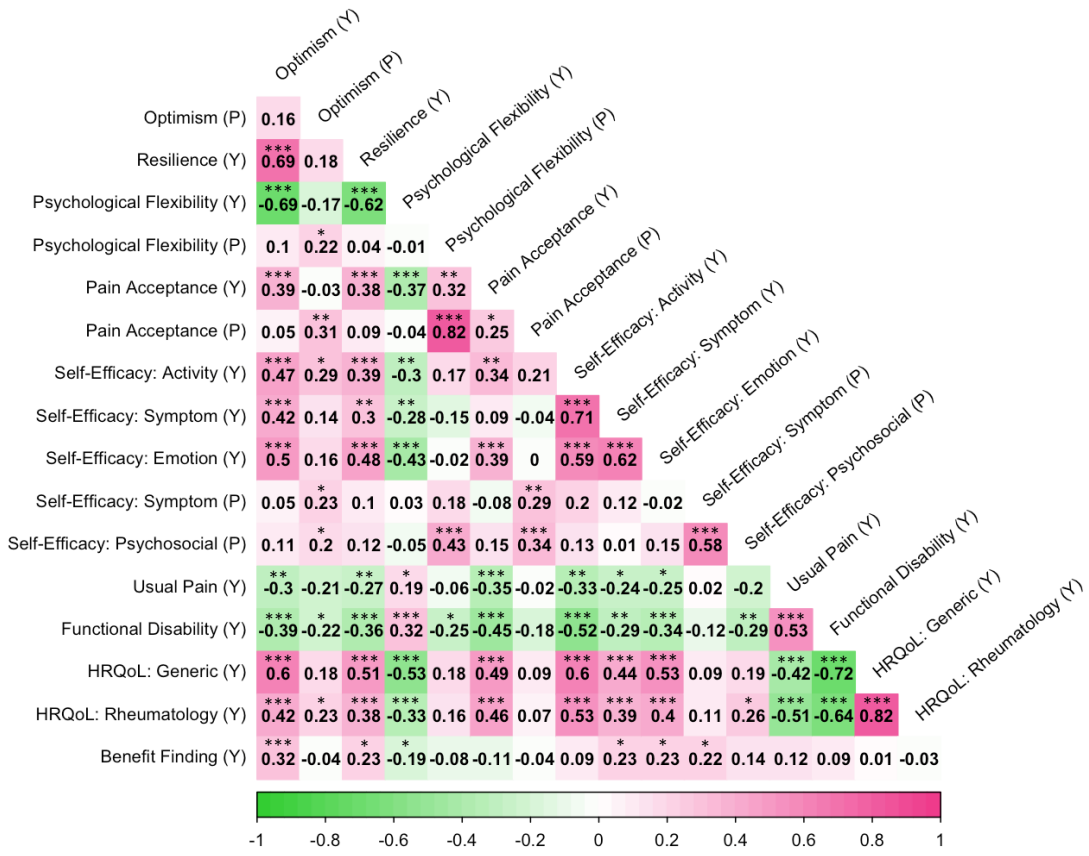
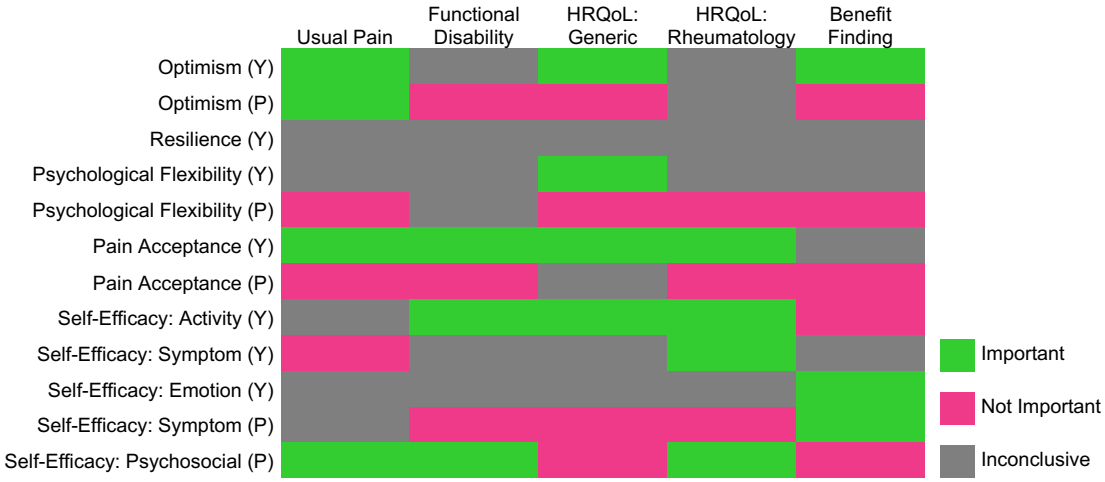
4.10. Figures

Figure 4.1. Bivariate Correlations for Study Variables



Note. Pearson correlation coefficient is significant at the ***.001 level, **.01 level, or *.05 level (2 tailed). HRQoL = Health-related quality of life; P = Parent; Y = Youth.

Figure 4.2. Summary of the Significance of Each Predictor between Correlational and Regression Analyses Across Models



Note. HRQoL = Health related quality of life; P = Parent; Y = Youth. "Important" = Relative importance (RI) ≥ +.05 and at least one of the standardized coefficients are significant. "Not Important" = RI values between -.05 and +.05 and neither standardized coefficient is significant. "Inconclusive" = All other cases where RI < +.05 but at least one of the two coefficients is statistically significant.

4.11. Tables

Table 4.1. Description, Scoring, and Psychometric Support for Study Measures

Domain	Concept/Definition	Scale	Scoring and Psychometric Support
Resilience Resources	Dispositional Optimism: A set of positive expectations regarding the future.	Youth Life Orientation Test (YLOT; Ey et al., 2005)	Twelve items were rated on a scale from 0 (*Not true for me*) to 3 (*True for me*). Six items measuring pessimistic expectations were reverse coded. This scale has good internal consistency, test-retest reliability, and convergent validity (Ey et al., 2005).
		Life Orientation Test Revised (LOT-R; Scheier et al., 1994)	Six items (and four fillers) were rated on a scale from 0 (*Strongly disagree*) to 4 (*Strongly agree*). Half of the items were reverse scored. Adequate predictive and discriminant validity, and good internal consistency have been observed (Scheier et al., 1994).
	Trait Resilience: The ability to bounce back or recover from stress.	Brief Resilience Scale (BRS; Smith et al., 2008)	Six items assessing resilience were rated on a scale from 1 (*Strongly disagree)* to 5 (*Strongly agree*). Select items were reverse scored. While not specifically developed for adolescents, it has been used in many adolescent populations and demonstrated acceptable internal consistency and appropriate concurrant, discriminant, and criterion validity (Smith et al., 2023).
Resilience Mechanisms	Psychological Flexibility: The capacity to stay present focused and engaged in values-based action.	Avoidance and Fusion Questionnaire for Youth (AFQ-Y8; Greco et al., 2008)	Eight items were rated on a scale from 0 (*Not at all true*) to 4 (*Very true*). They were averaged to create a total score of psychological *inflexibility*. This scale has demonstrated excellent internal consistency and good convergent and construct validity (Greco et al., 2008).
		Parental Psychological Flexibility Questionnaire (PPFQ-10;	Ten items were rated on a scale from 0 (*Never true*) to 6 (*Always true*). Select items were reverse scored. This scale has good internal consistency

Domain	Concept/Definition	Scale	Scoring and Psychometric Support
		Timmers et al., 2019)	and construct validity (Timmers et al., 2019).
	Pain Acceptance: A willingness to permit pain to be present without trying to stop or reduce it (pain willingness) and a willingness to persist with important activities (activity engagement).	Chronic Pain Acceptance Questionnaire for Adolescents (CPAQ – A8; Gauntlett-Gilbert et al., 2019)	Eight items assessing pain willingness (reverse scored) and activity engagement were rated on a scale from 0 (*Never true*) to 4 (*Always true*) and averaged to create a total score. Good internal consistency, validity, and sensitivity to treatment have been seen with this abbreviated scale (Gauntlett-Gilbert et al., 2019).
		Parent Pain Acceptance Questionnaire (PPAQ; Smith et al., 2015)	Fifteen items assessing acceptance of pain-related thoughts and feelings (reverse scored) and activity engagement were rated from 0 (*Never true*) to 4 (*Always true*) and averaged to create a total score. This scale has strong internal consistency and construct validity (Smith et al., 2015).
	Arthritis Self-Efficacy: The perceived ability to carry out the courses of action that produce desired attainments in various domains of life with JIA (i.e., activities, symptoms, and emotions in youth; and symptoms and psychosocial domains in parents).	Children's Arthritis Self-Efficacy Scale (CASE; Barlow et al., 2001)	Four items assessing activity, three items assessing symptom, and three items assessing emotion self-efficacy were rated on a scale from 1 (*Not at all sure*) to 5 (*Very sure*). The scale has good to excellent internal consistencies and strong concurrent and construct validity (Barlow et al., 2001).
		Parent's Arthritis Self-Efficacy Scale (PASE; Barlow et al., 2000)	Seven items assessing parent's belief in their ability to manage their child's symptoms and seven items assessing their certainty in managing psychosocial components were rated from 0 (*Very uncertain*) to 7 (*Very certain*). This was not 1 to 7 due to an administration error. Excellent internal consistency and good concurrent and construct validity were seen in the validation study (Barlow et al., 2000).
Recovery and Sustainability	Pain Intensity: A sensory component of the pain	11-point Numeric Rating	Youth rated their current and usual pain intensity on a scale

Domain	Concept/Definition	Scale	Scoring and Psychometric Support
Outcomes	experience, assessing one's perceived intensity of their usual pain.	Scale (NRS-11)	from 0 (*No hurt)* to 10 (*The worst hurt you could ever imagine*). A recent review demonstrated sufficient reliability and criterion validity for this scale and strongly recommended its use amongst adolescent populations (Birnie et al., 2019).
	Functional Disability: An assessment of the impact of a disease on one's daily functioning.	Functional Disability Inventory (FDI; Walker & Greene, 1991)	Fifteen items assessing difficulty engaging in activities were rated by youth on a scale from 0 (*No trouble*) to 4 (*Impossible*). In the validation study, there was strong support for the internal consistency and construct, concurrent, and predictive validity of this measure (Walker & Greene, 1991).
	Health-Related Quality of Life (HRQoL): A multidimensional assessment of one's physical, mental, and social functioning (Generic Core Total Score) and rheumatology specific physical and psychosocial health (Rheumatology Total Score).	The Pediatric Quality of Life Inventory (PedsQL) 4.0 Generic Core Scales and 3.0 Rheumatology Module; Varni et al., 2002)	Youth rated 23 items pertaining to their general HRQoL and 22 items pertaining to their rheumatology specific HRQoL from 0 (*Never*) to 4 (*Almost always*). Items were reverse scored, transformed to a 0-100 scale and averaged to create 2 total scores. Good to excellent internal consistency has been seen for total scores, and both construct validity and responsiveness have been demonstrated (Varni et al., 2002).
Growth Outcomes	Benefit Finding: An acknowledgement of positive changes or benefits in the presence of an event such as an illness or trauma.	Benefit Finding Scale for Children (BFSC; Phipps et al., 2007)	Ten items were rated from 0 (*Not true for me*) to 4 (*Very true for me*). Instructions were modified to, "Having had my *arthritis…*" as opposed to "*illness*". This scale has excellent internal consistency and some evidence of construct validity (Phipps et al., 2007).

Table 4.2. Descriptive and Medical Variables

Demographics and Medical Variables	Parent (n=140) *N* (%) or *M* ± *SD* (Min, Max)	Youth (n=129) *N* (%) or *M* ± *SD* (Min, Max)
Participant Demographics		
Age (Years)	45.24 ± 4.87 (33, 57) [d]	15.29 ± 1.62 (13, 18) [a]
Sex (Female) [a]		110 (70.5)
Gender [a]		
Mother/Girl	148 (94.9)	106 (67.9)
Father/Boy	8 (5.1)	46 (29.5)
Other (transgender, nonbinary, gender fluid)	--	4 (2.5)
Ethnicity [b]		
White	123 (78.8)	110 (70.5)
Aboriginal	7 (4.5)	9 (5.8)
South Asian	4 (2.6)	4 (2.6)
Black	3 (1.9)	3 (1.9)
East/Southeast Asian	3 (1.8)	3 (1.9)
Other (Jewish, West Asian, Latin American)	5 (3.2)	4 (2.6)
Prefer not to answer	2 (1.3)	1 (0.6)
Country of Residence		
Canada	96 (68.6)	95 (74.2)
United Kingdom	24 (17.1)	19 (14.8)
USA	16 (11.4)	11 (8.6)
Other (Ireland, South Africa, Australia)	4 (2.8)	3 (2.4)
Income (in CAD)		
<$50,000	16 (11.5)	--
$50,000 – 99,999	42 (30.0)	--
$100,000 – 149,999	29 (20.7)	--
>$150,000	36 (25.7)	--
Prefer not to answer	17 (12.1)	--
Youth Medical Characteristics		
Diagnosis [a e]		
Oligoarticular Arthritis	--	26 (16.8)
Polyarticular Arthritis	--	37 (23.9)
Enthesitis-Related Arthritis	--	27 (17.4)
Psoriatic Arthritis	--	10 (6.5)
Systemic Arthritis	--	17 (11.0)
Undifferentiated or Unknown [c]	--	38 (24.5)
Age at Diagnosis [a f]		8.09 ± 4.72 (0, 16)
Current Disease Activity (Active/Flare)	87 (62.1)	75 (59.1) [g]

Note. Parent report data were used for medical characteristics. Percent was calculated based on number of participants who completed the question rather than the total N. [a]Data from parents/youth were combined to achieve an N=156. If parent/youth data did not match, youth data was used before parent data for demographic information, and parent data was used before youth data for medical information. [b]Participants could select more than one response. [c]Three participants indicated also having a diagnosis of autoimmune arthritis, dermatomyositis, and scleroderma. [d]n=139. [e]n=155. [f]n=152. [g]n=127.

Table 4.3. Questionnaire Data

Questionnaire & Theoretical Range of Scores	*N*	*M*	*SD*	Min, Max	α
Optimism (Y): YLOT; 0-3	124	1.73	0.72	0.00, 3.00	.92
Optimism (P): LOT-R; 0-4	131	2.39	0.71	0.83, 4.00	.83
Resilience (Y): BRS; 1-5	122	3.16	0.78	1.00, 4.83	.84
Psychological Flexibility (Y): AFQ-Y8; 0-4	125	1.29	0.87	0.00, 4.00	.87
Psychological Flexibility (P): PPFQ-10; 0-6	137	4.02	0.94	1.30, 5.80	.86
Pain Acceptance (Y): CPAQ-A8; 0-4	124	2.48	0.67	0.88, 4.00	.76
Pain Acceptance (P): PPAQ; 0-4	137	2.29	0.66	0.00, 3.60	.83
Self-Efficacy (Y): CASE; 1-5					
Activity	120	2.89	1.14	1.00, 5.00	.90
Symptom	119	2.76	1.00	1.00, 5.00	.85
Emotion	120	3.05	1.22	1.00, 5.00	.87
Self-Efficacy (P): PASE; 0-7					
Symptom	125	3.05	1.42	0.00, 6.57	.89
Psychosocial	126	4.43	1.51	0.40, 7.00	.93
Pain Intensity (Y): NRAS-11; 0-10	125	4.96	2.23	0.00, 10.00	--
Functional Disability (Y): FDI; 0-4	126	1.20	0.82	0.00, 3.40	.94
HRQoL (Y): PedsQL Generic 4.0; 0-100	122	60.01	21.21	16.30, 100.00	.95
HRQoL (Y): PedsQL Rheumatology 3.0; 0-100	122	63.06	20.39	9.09, 100.00	.94
Benefit Finding (Y): BFSC; 0-4	120	2.28	0.80	0.00, 4.00	.89

Note. P = Parent; Y = Youth.

Table 4.4. Contributions of Resilience Resources and Mechanisms to the Outcomes of Usual Pain Intensity, Functional Disability, Generic and Rheumatology Specific Health-Related Quality of Life, and Benefit Finding through Multiple Regressions

	r	*p* (*r*)	*β*	*p* (*β*)	95% CI (*β*)	R^2	RI
Usual Pain Intensity	--	--	--	--	--	.29	--
Pain Acceptance (Y)	-0.34	**<.001**	-0.34	**<.001**	-0.52, -0.16	--	.12†
Self-Efficacy: Psychosocial (P)	-0.20	.071	-0.35	**.010**	-0.61, -0.08	--	.07†
Optimism (Y)	-0.31	**.002**	-0.16	.229	-0.42, 0.10	--	.05†
Optimism (P)	-0.20	.072	-0.22	**.019**	-0.41, -0.04	--	.05†
Self-Efficacy: Activity (Y)	-0.30	**.007**	-0.14	.355	-0.44, 0.16	--	.04
Self-Efficacy: Symptom (Y)	-0.22	.055	-0.14	.322	-0.41, 0.13	--	.03
Resilience (Y)	-0.28	**.002**	-0.10	.402	-0.32, 0.13	--	.03
Self-Efficacy: Symptom (P)	0.00	.999	0.25	**.037**	0.02, 0.49	--	.00
Pain Acceptance (P)	-0.02	.852	0.03	.876	-0.37, 0.43	--	-.00
Psychological Flexibility (P)	-0.04	.759	0.22	.338	-0.23, 0.67	--	-.01
Psychological Flexibility (Y)	0.19	**.042**	-0.14	.176	-0.35, 0.06	--	-.03
Self-Efficacy: Emotion (Y)	-0.23	**.017**	0.22	.053	-0.00, 0.45	--	-.05
Functional Disability	--	--	--	--	--	.42	--
Self-Efficacy: Activity (Y)	-0.52	**<.001**	-0.44	**<.001**	-0.68, -0.19	--	.23†
Pain Acceptance (Y)	-0.45	**<.001**	-0.25	**.007**	-0.43, -0.07	--	.11†
Self-Efficacy: Psychosocial (P)	-0.29	**.005**	-0.23	**.029**	-0.44, -0.02	--	.07†
Resilience (Y)	-0.36	**<.001**	-0.09	.468	-0.31, 0.14	--	.03
Psychological Flexibility (Y)	0.32	**<.001**	0.07	.563	-0.17, 0.31	--	.02
Psychological Flexibility (P)	-0.25	**.027**	-0.09	.625	-0.44, 0.27	--	.02
Optimism (P)	-0.21	.060	-0.08	.388	-0.28, 0.11	--	.02
Optimism (Y)	-0.39	**<.001**	-0.02	.845	-0.26, 0.21	--	.01
Self-Efficacy: Symptom (Y)	-0.28	**.006**	0.02	.875	-0.21, 0.25	--	-.01
Self-Efficacy: Symptom (P)	-0.14	.179	0.07	.549	-0.16, 0.31	--	-.01
Pain Acceptance (P)	-0.19	.082	0.14	.408	-0.19, 0.46	--	-.03
Self-Efficacy: Emotion (Y)	-0.34	**<.001**	0.14	.259	-0.10, 0.37	--	-.05
HRQoL: Generic	--	--	--	--	--	.59	--
Self-Efficacy: Activity (Y)	0.60	**<.001**	0.32	**.005**	0.09, 0.55	--	.19†
Psychological Flexibility (Y)	-0.53	**<.001**	-0.20	**.050**	-0.40, -0.00	--	.10†
Pain Acceptance (Y)	0.48	**<.001**	0.18	**.015**	0.04, 0.33	--	.09†
Optimism (Y)	0.61	**<.001**	0.14	.240	-0.09, 0.37	--	.08†
Psychological Flexibility (P)	0.18	.138	0.27	.064	-0.02, 0.56	--	.05
Self-Efficacy: Emotion (Y)	0.54	**<.001**	0.08	.433	-0.12, 0.27	--	.04
Resilience (Y)	0.52	**<.001**	0.06	.479	-0.10, 0.22	--	.03
Self-Efficacy: Symptom (Y)	0.44	**<.001**	0.04	.749	-0.18, 0.26	--	.02
Self-Efficacy: Symptom (P)	0.13	.221	0.08	.407	-0.11, 0.27	--	.01
Self-Efficacy: Psychosocial (P)	0.19	.104	0.01	.906	-0.19, 0.22	--	.00
Optimism (P)	0.17	.116	0.01	.904	-0.15, 0.17	--	.00
Pain Acceptance (P)	0.10	.381	-0.29	**.045**	-0.57, -0.01	--	-.03
HRQoL: Rheumatology	--	--	--	--	--	.47	--
Pain Acceptance (Y)	0.45	**<.001**	0.34	**<.001**	0.19, 0.49	--	.15†
Self-Efficacy: Activity (Y)	0.53	**<.001**	0.26	.064	-0.01, 0.53	--	.14†
Self-Efficacy: Symptom (Y)	0.40	**<.001**	0.21	.127	-0.06, 0.47	--	.08†
Self-Efficacy: Psychosocial (P)	0.28	**.018**	0.21	.067	-0.02, 0.44	--	.06†

	r	*p* (*r*)	*β*	*p* (*β*)	95% CI (*β*)	R^2	RI
Optimism (P)	0.25	**.027**	0.18	**.040**	0.01, 0.35	--	.04
Resilience (Y)	0.38	**<.001**	0.09	.309	-0.08, 0.26	--	.03
Psychological Flexibility (P)	0.16	.202	0.14	.437	-0.21, 0.49	--	.02
Optimism (Y)	0.43	**<.001**	0.04	.671	-0.16, 0.25	--	.02
Psychological Flexibility (Y)	-0.33	**<.001**	-0.01	.910	-0.21, 0.19	--	.00
Self-Efficacy: Symptom (P)	0.15	.190	-0.02	.853	-0.24, 0.20	--	-.00
Pain Acceptance (P)	0.09	.485	-0.30	.080	-0.63, 0.04	--	-.03
Self-Efficacy: Emotion (Y)	0.40	**<.001**	-0.14	.257	-0.38, 0.10	--	-.06
Benefit Finding	--	--	--	--	--	.28	--
Optimism (Y)	0.32	**<.001**	0.36	**.006**	0.10, 0.62	--	.12†
Self-Efficacy: Emotion (Y)	0.23	**.017**	0.22	.057	-0.01, 0.45	--	.05†
Self-Efficacy: Symptom (P)	0.22	**.038**	0.21	.063	-0.01, 0.43	--	.05†
Pain Acceptance (Y)	-0.12	.172	-0.29	**<.001**	-0.44, -0.13	--	.03
Self-Efficacy: Symptom (Y)	0.24	**.012**	0.08	.572	-0.19, 0.35	--	.02
Self-Efficacy: Psychosocial (P)	0.14	.135	0.07	.537	-0.16, 0.30	--	.01
Psychological Flexibility (P)	-0.10	.336	-0.07	.695	-0.40, 0.27	--	.01
Optimism (P)	-0.03	.775	-0.15	.125	-0.34, 0.04	--	.00
Psychological Flexibility (Y)	-0.20	**.024**	-0.02	.881	-0.23, 0.20	--	.00
Resilience (Y)	0.23	**.013**	0.01	.928	-0.22, 0.25	--	.00
Pain Acceptance (P)	-0.08	.486	0.03	.834	-0.28, 0.34	--	-.00
Self-Efficacy: Activity (Y)	0.10	.313	-0.17	.203	-0.43, 0.09	--	-.02

Note. Relative importance was calculated as the product of the reported standardized correlation and regression coefficients. CI = Confidence Interval; HRQoL = Health-related quality of life; P = Parent; RI = Relative Importance; Y = Youth. † Variables classified as "Important" based on the size of the RI value and pattern of statistical significance. Minor inconsistencies in RI values are due to rounding; calculations were done with higher precision than two decimal places.

CHAPTER 5: DISCUSSION

The final chapter of this dissertation summarizes and discusses the key findings of the included studies (Chapters 2-4). Theoretical and clinical implications are presented, followed by a discussion of the key strengths and limitations of this dissertation and directions for future research.

5.1. Summary and Discussion of Key Findings

The objective of this dissertation was to further our understanding of how to promote resilience and pain adaptation in the context of JIA pain. This was accomplished via three interrelated studies using different informants and methodologies. The findings and significance of these studies are presented below.

Given the vast and discrepant landscape of information exploring the relationship between psychosocial factors and JIA pain, the broad goals of Chapter 2 were to synthesize the literature, address discrepancies, and generate research directions and hypotheses for future study. Specifically, the aim was to synthesize the literature to determine what psychosocial factors in both individuals with JIA and others in their environment (e.g., parents) are associated with and predictive of different measures of JIA pain. Using the JBI methodology for systematic reviews (Aromataris & Munn, 2020), 61 studies reporting on 516 unique associations were included. While many psychosocial factors have been explored, slightly under half had a statistically significant relationship with outcomes. Specifically, worse JIA pain was consistently found to be associated with greater perceptions of pain unpleasantness and more beliefs that pain causes harm, disability, and loss of control. This suggests the importance of interventions targeting pain beliefs in conjunction with pain neuroscience education. In other words,

providing youth and caregivers with education around how chronic pain works and can be affected by biopsychosocial experiences, including one's beliefs about pain. JIA pain was also found to generally be associated with lower youth and parent self-efficacy and worse school and social functioning, which are key domains to address in clinic appointments and psychosocial interventions. Positive self-statements and behavioral distraction were the most robust adaptive coping styles across studies, whereas catastrophizing was the most robust maladaptive coping style. Finally, JIA pain was consistently related to lower HRQoL and well-being, and more internalizing symptoms in children (and to a lesser degree parents). The prognostic relationships generally explored the relationships between pain and either well-being or internalizing symptoms; however, most demonstrated bidirectional relationships (e.g., either greater pain leading to worse well-being, or worse well-being leading to greater pain). This finding is nevertheless in line with current theoretical models. In addition to identifying important targets for clinical intervention, this review also provoked broader reflections on the current state of the literature. Specifically, most of the studies were of moderate quality due to the lack of covariates and the smaller sample sizes (*Mdn* = 85 participants, range = 11 – 1906, *IQR* = 99). Most associations were correlational, identified risk (versus resilience) resources or mechanisms, and neglected parent-factors. Moreover, a small but noteworthy number utilized proxy reports to measure the child's pain despite recommendations against when possible (e.g., Eccleston et al., 2021). As such, findings from Chapter 2 laid the groundwork for Chapters 3 and 4. In addition to addressing the broader gaps in the literature (e.g., including parent factors, using larger samples and self-report measures of pain), Chapter 3 assessed youth and parent perfectionism, a novel resource that had not

been identified in this literature, and Chapter 4 utilized the few resilience resources and mechanisms identified in this study as predictors.

Building off of these findings, Chapter 3 used a micro approach to explore the role of perfectionism - a resource that had not been explored in relation to JIA pain (Chapter 2) or any other recovery, sustainability, or growth outcomes in JIA (Hynes et al., 2019). Perfectionism is a multidimensional construct that was conceptualized in terms of three dimensions (Hewitt & Flett, 1991): SOP, SPP, and OOP. Based on the SCCAMPI model of perfectionism in health (Molnar et al., 2016), a priori preregistered hypotheses were that youth and parent trait perfectionism would serve as a risk factor, increasing symptoms of depression and anxiety in youth with JIA through two pathways. The first hypothesis was that youth and parents high in SOP would be more likely to engage in maladaptive cognitions regarding the limits that JIA pain may impose, which in turn would impact the youth's mental health. The second hypothesis was that youth SOP, SPP, and parent OOP would predict greater internalizing symptoms in youth through increased self-concealment. In other words, youth and parents high in perfectionism would encourage youth to conceal their pain and symptoms thus negatively affecting their mental health. Addressing limitations observed in Chapter 2, data was collected from 156 youth with JIA (13 to 18 years old) and a caregiver through a large-scale online study. Findings suggested that youth perfectionism serves as a risk factor for internalizing symptoms in youth with JIA and may manifest through pain catastrophizing and self-concealment. Parent perfectionism was negatively associated with youth symptoms of depression, suggesting that while it may largely be a maladaptive trait, in this context it may have adaptive components. Parents high in SOP may go above and beyond in

ensuring their child is supported in managing the physical and psychosocial sequalae of a JIA diagnosis, though this is speculative. Although the role of parent perfectionism was largely not significant in these models, this may be due to smaller effect sizes for parent-child effects (e.g., Randall, Smith, et al., 2018), and thus lower statistical power than correlations observed within youth. While this was a preliminary exploration of the role of youth and parent perfectionism in the context of JIA, results suggest that at minimum, knowledge of the clinical presentation of perfectionism and an awareness of these relationships may be useful for HCP. At an intervention level, this may be an important target alongside other concerns such as pain and internalizing symptoms, with some research suggesting that ignoring perfectionism can lessen the effects of other therapeutic targets (Galloway et al., 2022). Taken together, this study addressed important gaps in the literature identified in Chapter 2, exploring this novel resource in youth and parents, and identifying its relationship to the psychological health of youth with JIA and the mechanisms in which it may act.

Building off the methodological gaps and incorporating some of the evidence-based resilience resources and mechanisms identified in Chapter 2, Chapter 4 approached the study of resilience in the context of JIA pain from a macro approach. A significant limitation of this literature to date is that it has largely focused on resources and mechanisms independently rather than understanding the competing effects of multiple variables analyzed together (Hynes et al., 2019; Knafl et al., 2015) which is necessary to understand what to emphasize in order to optimize living with JIA. As such, the aims of this study were to explore youth and parent resources/mechanisms that have been identified as relevant in the broader pediatric pain literature (i.e., optimism, trait

resilience, psychological flexibility, pain acceptance, self-efficacy), and explore their RI in contributing to diverse outcomes related to recovery/sustainability (i.e., pain intensity, functional disability, generic and rheumatology specific HRQoL), and growth (i.e., benefit finding). As hypothesized, significant positive relationships were observed between resilience resources and mechanisms within individuals. Relationships were weaker and less often significant across dyad members (e.g., youth and parent optimism). Using the Pratt Index (i.e., the product of standardized correlation and regression coefficients) to calculate the RI, alongside results from the bivariate correlations and multiple regressions, the importance of each predictor was determined. Results varied depending on the specific outcome measure being used. Clinically, the strong correlations observed suggest that by targeting one resource or mechanism, it is possible that improvements may be seen in other domains. This research nevertheless underscores the importance of working with clients to establish their treatment goals (e.g., increasing functioning versus improving HRQoL) as that will determine which resilience resources or mechanisms are most important to emphasize. Despite these differences, key patterns emerged. Notably, youth pain acceptance was the most robust predictor (rated as "Important" across 4/5 outcomes) and was always among the top four contributors in terms of RI. This finding accentuates its salience above and beyond other resources and mechanisms. This has important implications for working with youth with JIA clinically, as helping youth to allow the pain to be present and to continue to persist with valued activities appears to be a key mechanism change in fostering resilience, regardless of the outcome. Another pattern that was observed was that across 4/5 different surrogate markers of resilience, at least one parent resilience resource or mechanism (most often

psychosocial self-efficacy) was classified as “Important”. This suggests that supporting parent resilience (e.g., through involvement in interventions) in the context of JIA continues to have positive impacts on even older adolescents.

Throughout these studies (Chapters 2-4), findings add to the growing body of literature about the risk and resilience resources and mechanisms that contribute to the pain adaptation of youth with JIA. Across these studies, support emerged for various youth-specific risk (SOP; SPP; pain unpleasantness; beliefs that pain causes harm, disability, and loss of control; impaired school and social functioning; coping via catastrophizing; pain-related fears; self-concealment; internalizing symptoms) and resilience (pain acceptance; self-efficacy; coping via positive self-statements and behavioral distraction; optimism; trait resilience; psychological flexibility; HRQoL; well-being) resources and mechanisms. Support also emerged for parent-specific risk (SOP; coping via catastrophizing, fears of pain, internalizing symptoms) and resilience (e.g., self-efficacy, optimism) factors, albeit to a lesser degree. While the studies utilized different surrogate markers of youth pain adaptation given the diverse possibility of measures (i.e., Studies 1 and 3 used pain as a proxy for recovery/sustainability; Study 2 used internalizing symptoms as a proxy for recovery/sustainability; Study 3 used functioning and HRQoL as proxies for recovery/sustainability and benefit finding as a proxy for growth), similarities were nevertheless seen. For example, coping via catastrophizing was seen as a risk mechanism in relation to both pain (Chapter 2) and internalizing symptoms (Chapter 3); and youth and parent self-efficacy were significant resilience mechanisms in relation to pain (Chapters 2 and 4), functional disability (Chapter 4), HRQoL (Chapter 4), and benefit finding (Chapter 4). Although there is a

need for further research in this area with larger samples and more complex analyses, together these studies elevate the state of the literature pertaining to promoting resilience and pain adaptation in youth with JIA and their caregivers. These findings have important theoretical and clinical implications, as well as numerous strengths and limitations that can be used to guide future research, all of which are described below.

5.2. Theoretical Implications

The Ecological Resilience-Risk Model in Pediatric Chronic Pain (Cousins, Kalapurakkel, et al., 2015) served as the foundational model for this dissertation. This model builds off the prominent risk-resilience model in adult chronic pain (Sturgeon & Zautra, 2013) incorporating components of ecological systems theory (Bronfenbrenner, 1979) and resilience theory (e.g., Zimmerman, 2013). Specifically, this model describes the dynamic and systemic process that is resilience. Resilience resources and risk factors (stable traits) interact with resilience and risk mechanisms (dynamic processes) within and between the individual, family/social context, culture, and time, to impact pain adaptation, as defined by outcomes that can be categorized into one's recovery or sustainability (e.g., physical health, psychological health, functioning) or growth (e.g., benefit finding). As such, the studies that comprise this thesis are founded on, and are contributing to, theoretically-driven science.

Each study mapped directly to this model, providing an empirical basis to further our knowledge regarding the diverse interactions and pathways that exist, at times weaving in other models as appropriate. Chapter 2 synthesized the literature regarding the known youth and parent psychosocial factors (resilience resources, resilience mechanisms, risk factors, risk mechanisms) that are related to JIA pain (i.e., a recovery

and sustainability outcome similar to assessing one's physical health; Palit et al., 2021). Chapter 3 narrowed in on this model, exploring the role of youth and parent perfectionism in the context of JIA pain. This was explored at a multivariate level, wherein dimensions of perfectionism (largely risk factors) predicted more internalizing symptoms in youth with JIA (recovery and sustainability outcomes assessing psychosocial health), in part through pain catastrophizing and youth self-concealment of health-related symptoms (risk mechanisms). Intriguingly, one variation in this pattern was that parent SOP, while a risk factor in some contexts (e.g., associated with greater catastrophizing and pain-related fears in parents), was also in part a resilience resource in that it was associated with fewer depression symptoms in youth with JIA. Finally, Chapter 4 zoomed out on this model, exploring the RI of evidence-based resilience resources and mechanisms as identified by the broader pediatric pain literature (Brandelli et al., 2023; Hynes et al., 2019) at a multivariable level in contributing to usual pain, functioning, HRQoL (recovery and sustainability outcomes), and benefit finding (growth). Results highlighted the salience of youth pain acceptance (resilience mechanism) followed by youth optimism (resilience resource) and domains of youth and parent self-efficacy (resilience mechanism) as key contributors to this model.

Taken together, these studies demonstrate support for the Ecological Resilience-Risk Model in Pediatric Chronic Pain in its application to the context of youth with JIA and their parents, with particular support for within-youth resources and mechanisms. Nevertheless, some gaps in this model have also been identified. As Hynes and colleagues (2019) first pointed out, the differentiation of mechanisms and outcomes is at times unclear (e.g., self-management may be perceived as a resilience mechanism,

however may also suggest adaptation to JIA and thus a sustainability outcome). To take this one step further, the operationalization of resources and mechanisms also stands to be clarified. Although resources, such as perfectionism, have the tendency to be pre-existing trait-like factors, there remains a potential for prevention and/or new learning (e.g., see Flett and Hewitt [2014] for ways to prevent perfectionism in youth). As such, a reconceptualization of these pathways wherein both existing and new resources pertaining to internal, external, and existential sources are accounted for may help further the literature in this field (Rosenberg & Yi-Frazier, 2016). Finally, although the broader culture, time, and developmental stage are incorporated into this model, there is a need for them to be more central and thoroughly integrated (e.g., Palit et al., 2021; Riggenbach et al., 2019) into empirical studies. As an example, in Chapter 3 it was found that youth SPP predicted youth depression, in part through youth self-concealment of JIA pain/symptoms. While this is interesting in and of itself, greater knowledge may be ascertained by quantitatively capturing the dynamic sociocultural and developmental milieu in which this relationship may occur or become stronger (e.g., perhaps their age, whether they personally know others with JIA, their socioeconomic status, and/or ability to afford treatments moderates the relationship between youth SPP and self-concealment; or perhaps whether their environment allows them to access timely mental health support may moderate the relationship between self-concealment and depression symptoms). A final consideration in advancing this model would be to move beyond largely a psychosocial conceptualization of resilience. There is value in integrating historical (e.g., early life events) and biological (e.g., sleep, biomarkers, immune functioning) markers that may also affect the dynamic process of pain adaptation (Palit et al., 2021).

A secondary theoretical model explored in this dissertation (Chapter 3) was the SCCAMPI (Molnar et al., 2016). It is worth noting that findings are generally in keeping with this model, which proposes that perfectionism in the context of chronic illness creates a situational discrepancy which through various intrapsychic and interpersonal pathways (i.e., lack of perceived control, negative self-evaluations, reduced social support, and greater self-concealment) can escalate stress and lead to ineffective coping, ultimately leading to worse adjustment and health outcomes. While certain dimensions of the model were not directly assessed (e.g., stress, coping, physical symptoms), the model was nevertheless supported wherein youth high in trait perfectionism were at an elevated risk of experiencing poorer adjustment/psychosocial outcomes by way of negative self-evaluations and self-concealment. It would be interesting for research to build off this and explore how the physiological experience of stress and poor psychosocial health may affect physical symptoms as well. A significant limitation of this model is its individualistic focus, which inherently limits its applicability to pediatric populations. This model would benefit from integrating developmental considerations to broaden its pertinence. This includes developing age-appropriate measures to assess the intrapsychic and interpersonal pathways, and incorporating ways in which parent and youth perfectionism may bidirectionally impact the intrapsychic and interpersonal processes that can amplify stress and coping in the family, subsequently leading to worse adjustment and health outcomes.

5.3. Clinical Implications

These studies have served to advance our knowledge regarding the diverse resilience and risk factors involved in the pain adaptation of youth with JIA. Given that

many individuals are involved in the care of youth with JIA, it is important that implications are tailored to each party. At each stage of this research, the empirical findings were integrated with the clinical expertise of the research team (e.g., psychologists, researchers, nurse-clinician scientists, pediatric rheumatologists) and with the lived experience of the panel of patient, family, and community partners involved with this research to enhance the meaningfulness of these findings for everyone involved in the child's care. As such, implications for rheumatologists and clinic staff; psychologists and mental health professionals; and patient, family, and community partners will be discussed.

For rheumatologists and clinic staff, the key implications from this set of studies are 1) the importance of assessing for and treating JIA pain (Chapter 2), 2) having an awareness of factors that contribute to the successful pain adaptation of youth with JIA and their caregivers (Chapters 3 and 4), and 3) recognizing when there may be value in referring youth and families for more targeted psychosocial support in addressing pain, perfectionism, or supporting resilience (Chapters 2-4). When it comes to pain assessment, some literature has found that HCP lack training and confidence, and are reluctant to engage in pain discussions (R. R. Lee et al., 2020), which is a stark disconnect from patient priorities (Correll et al., 2020) and the current evidence base (Connelly et al., 2011; Stinson & Prescott, 2021). Chapter 2 systematically reviewed the literature and identified a comprehensive set of psychosocial correlates that have been associated with various dimensions of JIA pain. Moreover, this study highlights some of the gaps in pain assessment in this literature which may extend to clinical practice (e.g., the use of unvalidated and single item pain measures, the reliance on proxy report measures of pain

when self-report measures may be ascertained). As such, this study along with resources such as the chapter, "Pain and Its Assessment" in the Textbook of Pediatric Rheumatology (Stinson & Prescott, 2021) are excellent resources that may help HCP heighten their own knowledge of, and subsequently the assessment and management of, pain in the context of JIA. For the second key implication, Chapters 3 and 4 identified resilience and risk factors that are related to the successful pain adaptation of youth with JIA. Although some of the findings are novel and further research is required (e.g., Chapter 3), an awareness of their associations and general presentations is nevertheless important. For example, if a clinician observes perfectionistic tendencies in a youth or their caregivers (e.g., setting extremely high standards for themselves/their child, focused on pleasing others) this may signal to them that there may be other challenges now or in the future that may benefit from intervention from a mental health professional, such as pain catastrophizing, concealment of symptoms, or internalizing symptoms. Similarly, youth pain acceptance and parent psychosocial self-efficacy emerged as largely relevant resilience resources (Chapter 4). Although comprehensively addressing these may be beyond the scope of the clinic staff, weaving in components of pain acceptance (e.g., discussing the importance of continuing to pursue valued activities perhaps in tailored ways) or parent psychosocial self-efficacy (e.g., checking in with parents regarding their beliefs in their abilities) are a few examples of how study findings can be incorporated. Finally, findings from this dissertation highlight times when rheumatologists and clinic staff may find value in referring youth and parents for further support, either to interdisciplinary pain programs (Liossi et al., 2019) or mental health professionals.

For mental health professionals, these studies provide empirical support for 1) the thorough assessment of JIA pain (Chapter 2), and 2) relevant psychosocial constructs that may necessitate the focus, refinement, or design of psychosocial interventions to promote resilience in youth with JIA and their families (Chapters 2-4). Chapter 2 provided a synthesized account of psychosocial factors related to various metrics of JIA pain, supporting the interdisciplinary approach to pain management. Regardless of whether these psychosocial factors cause, are caused by, or are indirectly related to pain, they are important to consider in the comprehensive assessment of pain and its impacts on the lives of youth with JIA and their families. The second key takeaway is that, generally, results are too heterogeneous and samples are too small to draw conclusions, though a small literature regarding the provision of psychosocial interventions in this population does exist (Cohen et al., 2017). Thus, there is a need for further research in this domain, and the findings of Chapter 2 along with empirically supported resilience and risk resources and mechanisms identified in Chapters 3 and 4 (e.g., youth and parent perfectionism, self-efficacy, pain acceptance, etc.) are fodder for the focus, refinement, or development of psychosocial interventions in this context. Regardless of the theoretical framework and therapeutic modalities of the clinician, adopting a resilience lens requires a shift in emphasis from diseases and disorders to client strengths and values. The focus of intervention, however, is dependent on the client's presenting concern and goals (e.g., pain management, learning skills to handle specific thoughts/feelings/emotions) and based on the results of these studies would benefit from the inclusion of parents when possible.

Finally, there are important implications for the broader JIA community, inclusive of youth with JIA and their caregivers. For researchers and community partners alike, as youth with health conditions and their families are important consumers of health information over the internet and social media (Hamm et al., 2014; Silver et al., 2019; Tonsaker et al., 2014; van Pelt et al., 2015), translating and disseminating this knowledge through social media and community networks is of utmost importance. The key implications of this research for youth with JIA and their families are: 1) having a greater awareness of their own experiences with the resilience or risk factors identified throughout this research and how that may map to their ability to adapt to JIA and continue to lead a values-based life; and 2) through increased awareness of their own experiences and needs, acquire greater knowledge around how to continue to hone in on their strengths and lead a values-based life through self-management, peer-led, or professionally-led interventions. As an example, in the iterative process of sharing and interpreting study results with the youth and parent partners involved with this research, one partner shared that they, in the past, have had the thought that they cannot control the pain or do anything about it. Only upon hearing how these beliefs have been associated with worse pain experiences (Chapter 2) did they make that connection in their own lives. They elaborated on how they were feeling hopeless and would have benefited from more knowledge about the neuroscience of pain (e.g., appreciating how thoughts and feelings can also impact their pain experiences). Thus, being able to recognize these patterns, and similarly these strengths (e.g., their own acceptance and self-efficacy), is a key implication for youth and parents. With this knowledge, this historically vulnerable medical population (given the novel integration of shared-decision making in pediatrics;

Wyatt et al., 2015) has greater knowledge and empowerment to seek out support as necessary, be it through resources within their community (e.g., Cassie + Friends, JIA at the National Rheumatoid Arthritis Society), peer support initiatives (e.g., Cassie + Friends, 2022; Stinson et al., 2016), evidence-based self-management interventions (e.g., Stinson et al., 2020), or professionally-led interventions as described above.

5.4. Key Strengths and Limitations

While each study contains its own set of strengths and limitations, this dissertation collectively has numerous strengths. From a methodological perspective, the inclusion of multiple methods (i.e., a systematic review, online data collection) adds to the validity of findings, and the inclusion of multiple informants (i.e., parents and children) is critical given that children do not exist in isolation (Palermo & Chambers, 2005) and parents are highly involved in the promotion of their child's resilience (e.g., Cousins, Kalapurakkel, et al., 2015).

From a conceptual lens, a shift in thinking has been adopted within the last decade, wherein resilience is conceptualized as a dynamic and systemic process that is seen as a complex network of differentially inter-related resources and mechanisms that interact within and across systems (Rutter, 2012; Ungar, 2018). In other words, resources and mechanisms do not exist in isolation from one another, rather it is this dynamic network that creates resilience (or risk) for an individual (Ungar, 2011). Although much of the existing literature exploring resilience in the context of JIA pain reflects this thinking (i.e., resilience resources and mechanisms are explored as main effects; Hynes et al., 2019); this dissertation made a concerted effort to explore the relationships that exist

between resources and mechanisms. While there is more work to be done in this domain, this also stands out as an important strength of this dissertation.

An additional strength is the exploration of resilience and risk as it pertains to a singular population; youth with JIA. While there may be some commonalities between the experience of those with JIA and other chronic pain populations, there are also differences given the inherent subjectivity of pain and to some degree, resilience. As an example, Wakefield and colleagues (2022; 2023) qualitatively explored the construct of diagnostic certainty in youth with JIA. They found that the diagnostic certainty experienced by youth with JIA, compared to chronic pain samples, allowed for greater support among medical providers and within families, and less stigma regarding their controllability of pain (i.e., the perception held by others that the patient is to blame for their pain). Understandably, this may lead to unique experiences among this population as it relates to domains such as perfectionism (Chapter 3), pain acceptance and self-efficacy (Chapter 4) and more broadly their experiences with pain adaptation. Furthermore, studying a singular population allows for more targeted dissemination messages. As an example, MacKenzie et al. (2021) explored the uptake of a knowledge translation tool for parents that provided strategies to manage vaccination pain before and after a vaccination. They found that participants were 9.76 times more likely to report using a strategy if they found the information relevant to themselves and their child. Although a different population, this finding also alludes to the benefits from a knowledge mobilization perspective of working with one community.

Two final strengths of this study are the adoption of best-practices in patient-engagement and open science. The inclusion of patient partners throughout the design

through dissemination conferred important advantages (e.g., the development of relevant research objectives and questions, user-friendly questionnaires and appropriate recruitment strategies, consumer-focused interpretation of data, and enhanced dissemination of findings; Brett et al., 2014). Moreover, through the open science practices of preregistering studies and hypotheses, making data available to the public for replication or additional study, and using open access options when publishing, this dissertation is not only contributing to easily accessible knowledge, it promotes the rigour of scientific inquiry, minimizes participant burden, and enhances the accessibility of data for further investigation in this niche field of study.

Nevertheless, there are important limitations to this dissertation that must also be addressed. The primary limitations of this research were in response to the COVID-19 pandemic. As this set of studies was in development prior to the COVID-19 pandemic, much of this work had to be reconceptualized to meet the limitations that ensued (e.g., in-person data collection was indeterminately terminated). While pivoting to online data collection conferred some advantages (e.g., access to larger samples, greater ease of participation), it also brought about unanticipated disadvantages. Throughout data collection for Chapters 3 and 4, the online survey was accessed to varying degrees by 421 sham respondents (e.g., disingenuous and software automated responses) across four distinct rounds of recruitment. This necessitated the closure and application of novel strategies to prevent, identify, and manage sham respondents on multiple occasions (e.g., consultations with ethics, removal of Twitter as a recruitment strategy, adjustment of advertisements to remove compensation, verification of participation via telephone calls, implementation of a screening questionnaire, use of "spam trap" questions, etc.).

Although not a novel phenomenon (Jones et al., 2021; Teitcher et al., 2015), online fraudulent activity such as this increased drastically over the course of the pandemic (Zhang et al., 2022). Despite best efforts and an extended recruitment period, the ideal sample size of 351 dyads was unable to be obtained given these challenges. As such, Chapters 3 and 4 were reconceptualized a second time to account for a smaller sample size. While important and significant findings were nevertheless identified, this ultimately precluded the observation of smaller effects (particularly regarding the role of parents) and of more synergistic effects between youth and parent resources and mechanisms. A final limitation of this dissertation, akin to much of the existing literature (e.g., Chapter 2), was the cross-sectional and correlational design of Chapters 3 and 4. Although the theoretical underpinnings of certain variables (e.g., perfectionism, optimism) suggests them to be more trait-like and as such may exist prior to the outcomes in question, this design ultimately does not allow for causal conclusions to be drawn.

Despite these limitations, these studies are important starting points for future research. Not only is it hoped that future research can make use of the learnings around online data collection in the post-pandemic era, it is hoped that the openly available data collected for this dissertation will also be used to further advance this science, ideally in tandem with other samples to increase the sample size and allow for more complex relationships to be analyzed.

5.5. Future Research Directions

Future research directions specific to each study are described throughout each chapter. In addition to building off the abovementioned theoretical/clinical implications and the limitations of this dissertation, there remain important directions for research in

three broad domains. As Ungar (2005) eloquently described, “a thicker definition of resilience reveals a seamless set of negotiations between individuals who take initiative, and an environment with crisscrossing resources that impact one on the other in endless and unpredictable combinations”. While this dissertation has illuminated just how complex resilience theory and research is, there is an important need to continue to delve into these unique crisscrossing resources and combinations within the context of JIA pain to continue to advance this field. Findings from this research suggest that this may be accomplished through qualitative or mixed-method methodologies, and/or more complex and longitudinal designs. Additionally, research is needed to continue to explore the clinical application of resilience theory to practice.

Given the subjective experiences of pain and pain adaptation, and the complex and undefined process of resilience (Hynes et al., 2019), this points to the value of qualitative or mixed method methodologies to help expand our understanding. As an example, while quantitative research is largely concerned with mapping out resources and mechanisms to help promote the resilience of individuals facing adversity, the lay-person’s experience and definition of resilience may not map to these models so fluently. As an example, Cox et al. (2022) used interpretive phenomenological analysis to ascertain the experiences of parents whose adolescents had been diagnosed with complex regional pain syndrome. Parents in this study experienced resilience in a different tone, describing how resilience to them was the incongruence between their own private distress and their perceived obligation to display socially desirable resilience behaviors. This is echoed by a recent column in *The Rheumatologist* (Kumar, 2023), wherein it was reflected that resilience is a laudatory and romanticized term that can convey judgment

and maintain inequity and exclusion to an individual who is merely "yearning to recover a sense of agency in a world that is often uncaring to personal circumstances". As such, there is value in furthering our understanding of resilience through qualitative and mixed-method approaches that describe the lived experiences of resilience amongst those experiencing adversity and help map those to our theoretical frameworks and models.

Moreover, as resilience is a never-ending process, taking a complex methodological and statistical approach to this field of study is also strongly encouraged. While there is initial value in looking at novel resources or mechanisms through univariate approaches (e.g., mindfulness, self-concept, self-esteem, self-compassion, motivation, spirituality, parent modelling, relationship with HCP, etc.), at its core, resilience is more complex than this. Similarly, while exploring resilience at a singular point in time with regards to a singular outcome may have some generalizability regarding one's overall ability to adapt, there is also potential for this to change based on innumerable life circumstances. As such, future research should incorporate multi-informant assessments (e.g., parents, siblings, teachers, etc.), longitudinal/ecological momentary assessment designs (e.g., throughout first few years of living with JIA or at key developmental periods such as the transition to adolescence), and/or more complex and interactive statistical approaches (e.g., network analyses, moderations, or mediations to explore protective and promotive relationships) to truly understand the interactions and patterns that comprise one's ability to adapt to their experiences with adversity.

Finally, there is a need for future research to test the value of strengths-based approaches in the clinical interventions provided to those with JIA to explore whether they provide desirable and sustained effects (Cousins, Kalapurakkel, et al., 2015).

5.6. Concluding Remarks

This multi-method, multi-informant approach to the study of resilience in the context of JIA pain has made important advancements in the fields of JIA, resilience, and importantly, pain. In 2021, the lancet commission on pediatric pain emphasized that for meaningful and lasting change to occur in the field, we need to make pain matter, make pain visible, make pain understood, and make pain better (Eccleston et al., 2021). This dissertation has addressed each of these targets. By considering how the psychosocial context can exacerbate and/or mitigate the experience of JIA pain and subsequently one's pain adaptation, this dissertation is inherently helping to bring discourse to the field and make JIA pain, an experience that is both invisible and rarely considered in childhood, matter. This dissertation helped to make JIA pain understood by using a multi-method and multidimensional assessment of JIA pain to further our understanding of the complexities of this experience (Chapters 2-4). These studies also helped to make JIA pain visible by synthesizing the known psychosocial correlates of JIA pain (Chapter 2), emphasizing the need for multidimensional pain self-assessments when appropriate and available (Chapter 2), and considering the ways in which JIA pain is a multidimensional experience in terms of the experience of pain itself, its broader impacts, and the need to promote resilience and pain adaptation (Chapters 2-4). Finally, the empirical findings from each study in this dissertation will contribute to the evidence base that serves the goal of making pain better. This is being implemented through open access publications, the translation and dissemination of findings to the JIA community (e.g., via blog posts and webinars), and ideally improvements to the interventions offered to youth with JIA to help them to optimize living in the face of adversity.

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