Duquesne University Duquesne Scholarship Collection

Electronic Theses and Dissertations

Summer 8-11-2018

Psychological and Family Variables Associated with the Treatment of Pediatric Idiopathic Pain

Rachael Hoffman

Follow this and additional works at: https://dsc.duq.edu/etd



Part of the Pain Management Commons

Recommended Citation

Hoffman, R. (2018). Psychological and Family Variables Associated with the Treatment of Pediatric Idiopathic Pain (Doctoral dissertation, Duquesne University). Retrieved from https://dsc.duq.edu/etd/1473

This Immediate Access is brought to you for free and open access by Duquesne Scholarship Collection. It has been accepted for inclusion in Electronic Theses and Dissertations by an authorized administrator of Duquesne Scholarship Collection. For more information, please contact phillipsg@duq.edu.

PSYCHOLOGICAL AND FAMILY VARIABLES ASSOCIATED WITH THE TREATMENT OF PEDIATRIC IDIOPATHIC PAIN

A Dissertation

Submitted to the School of Education

Duquesne University

In partial fulfillment of the requirement for the degree of Doctor of Philosophy

By

Rachael Hoffman, MSEd

August 2018

Copyright by

Rachael Hoffman, MSEd

PSYCHOLOGICAL AND FAMILY VARIABLES ASSOCIATED WITH THE TREATMENT OF PEDIATRIC IDIOPATHIC PAIN

By

Rachael Hoffman, MSEd

Approved May 1, 2018

Ara J. Schmitt, PhD Associate Professor of Department of Counseling, Psychology, and Special Education (Committee Chair) Kara M. McGoey, PhD Professor of Department of Counseling, Psychology, and Special Education (Committee Member)

Carol S. Parke, PhD Associate Professor of Department of Foundations and Leadership (Committee Member)

ABSTRACT

PSYCHOLOGICAL AND FAMILY VARIABLES ASSOCIATED WITH THE TREATMENT OF PEDIATRIC IDIOPATHIC PAIN

By

Rachael Hoffman, MSEd

August 2018

Dissertation supervised by Ara J. Schmitt, PhD.

The present study examined variability in treatment outcomes of children with idiopathic chronic pain. Specifically, the biopsychosocial model was used as a framework to consider the way that symptoms of depression and anxiety as well as parent response to pain contributed to the change in a child's confidence in participating in daily activities following treatment in an inpatient chronic pain treatment program.

ACKNOWLEDGMENTS

I would like to express my sincere gratitude to my dissertation chair, Dr. Ara Schmitt, for his support throughout my doctoral training and particularly in the completion of my dissertation. His guidance, patience, and good humor were key in carrying out this project. I am also grateful to other members of my committee, including Dr. Kara McGoey and Dr. Carol Parke, for their insightful feedback and encouragement.

I would also like to express utmost appreciation to the many caring clinicians at The Children's Institute. This research would not have been possible without access to data that had been systematically collected and recorded over several years. Furthermore, the support of the behavioral health staff as I pursued this project is unparalleled. I am fortunate and grateful for ongoing training under Dr. Jessica Walters, Dr. Paula Cerrone, and Dr. Heather Ufberg, and I thank each of them for offering their knowledge and encouragement. Additionally, my sincere thanks go to my colleagues and friends, Becky Keenan and Nicole Shea, for offering the feedback, motivation, and structure that enabled me to make continual progress.

I am incredibly grateful to my parents for inspiring my love of learning and desire to help others. I also would like to thank my brother, who continues to motivate me to work harder and happier in every aspect of my life. Finally, I would like to thank my husband for being a continuous source of support and joy in this endeavor and others. Without the confidence and love offered by my family, this project would not have been possible.

Table of Contents

| ABSTRACT | vii |
|---|-----|
| Chapter I: INTRODUCTION | 1 |
| Theoretical Basis | 2 |
| Relevant Literature | 3 |
| Problem Statement | 6 |
| Summary | 8 |
| Chapter II: LITERATURE REVIEW | 9 |
| Idiopathic Chronic Pain in Children | 9 |
| Epidemiology | 10 |
| Biopsychosocial Model | 11 |
| Biological Factors | 14 |
| Psychological Factors | 17 |
| Social Factors | 22 |
| Consequences of Idiopathic Chronic Pain | 25 |
| Treatment | 28 |
| Summary | 31 |
| Chapter III: METHOD | 32 |
| Participants | 32 |
| Measures | 33 |
| Research Design | 35 |
| Procedures | 36 |
| Data Analysis | 37 |
| Summary | 40 |
| Chapter IV: RESULTS | 41 |
| Descriptive Statistics | 41 |
| Statistical Analyses | 41 |
| Chapter V: DISCUSSION | 52 |
| Research Question 1 | 53 |
| Research Question 2 | 55 |
| Research Question 3 | 56 |
| Research Question 4 | 57 |

| Research Question 5 | 58 |
|-------------------------------------|----|
| Implications | 59 |
| Limitations | 62 |
| Recommendations for Future Research | 63 |
| Conclusion | 64 |
| References | 65 |

Chapter I: INTRODUCTION

Chronic pain is a common problem among children that can be associated with significant impacts across biological, psychological, and social functioning. Up to 88% of children present with chronic pain complaints (King et al., 2011). In many of these children, the pain is medically unexplained, or characterized as idiopathic pain. In fact, 44.4% of adolescents are estimated to report chronic idiopathic pain (Hoftun, Romundstad, Zwart, & Rygg, 2011). These children are at high risk for a variety of difficulties in social and psychological functioning, including their ability to complete activities of daily living, participate in school, and cope with stress. Moreover, idiopathic chronic pain in a child can substantially affect the family system due to elevated healthcare utilization, financial burden, and overall increased stress.

Significance of the Problem

A child's subjective experience with idiopathic chronic pain is often characterized by difficulty completing tasks that were previously simple, symptoms of anxiety and depression, and significant decreases in participation in activities and school (e.g., Aasland et al., 1997; Eccleston et al., 2004; Wasan, Sullivan, & Clark, 2010). These negative sequelae can be typically be understood within the context of functional disability, or the "impact of disease or trauma on a patient's daily functioning" (Walker & Greene, 1991: p. 39). Idiopathic chronic pain can result in substantial functional disability due to its physical, emotional, and social consequences (Konijnenberg et al., 2005).

Children with idiopathic chronic pain often also undergo numerous medical appointments and evaluations to rule out the possibility of various disorders and diseases that can cause pain before it is determined that their pain is idiopathic. As a result, families often experience

1

significant financial burden due to numerous doctor and therapy appointments leading up to and following the diagnosis of an idiopathic pain syndrome (Ho et al., 2008; Sleed, Eccleston, Beecham, Knapp, & Jordan, 2005). As compared to pain disorders with a discernable medical origin, families of children with idiopathic chronic pain have three times more direct and indirect (e.g., missed work) costs (Sleed et al., 2005). Similarly, in the United States, the cost of outpatient visits to the families of children seen at an outpatient multidisciplinary pain clinic over three months averaged \$1,761.06. Additionally, patients and their families were noted to spend an average of 28.54 hours at appointments over three months, which resulted in multiple days of missed school for patients and missed work for parents (Ho et al., 2008).

Theoretical Basis

Understanding the predictors and consequences of pediatric idiopathic chronic pain requires consideration of the biological, psychological, and social factors that can contribute to the onset and trajectory of pain. Thus, the biopsychosocial model, which examines each of these aspects, best applies as a framework to appreciate the contributions and interaction of these factors to better inform treatment. The biopsychosocial model originated in the work of George Engel (1977), as he advocated for a more comprehensive understanding of the multiple systems that can influence the presentation and resolution of medical problems. Along with traditional biomedical considerations, he asserted that the patient's behavioral and psychological presentation, along with their life experiences and present circumstances, must be considered for appropriate diagnosis and treatment. Additionally, he noted the potential impact of the patient's self-perception as well as his or her perception of disease and the social role of being "ill." Thus, biological, psychological, and social factors should be considered as part of the case conceptualization and treatment. Application of biopsychosocial model has been widely

supported in addressing idiopathic chronic pain, as many have noted the potentially invalidating and negative effect of focusing solely on biological phenomena in cases where biomedical explanations are insufficient to explain or treat a child's symptoms (e.g., Carter & Threlkeld, 2012; Gatchel, Peng, Peters, Fuchs, & Turk, 2007; Kozlowska et al., 2008). The present study focused specifically on the psychological and social components of the biopsychosocial framework. In particular, the psychological symptoms of depression and anxiety were examined along with the social factors of parent response to pain in terms of treatment efficacy.

Relevant Literature

Biological Considerations

Multiple biological factors are associated with idiopathic chronic pain in children. Gender has been noted as an important factor, with prevalence of idiopathic chronic pain being significantly higher in females (e.g., Eccleston, Crombez, Scotford, Clinch, & Connell, 2004; Hoftun, Romundstad, Zwart, & Rygg, 2011; King et al., 2011; Schechter et al., 2010; Sherry & Malleson, 2002). In fact, estimated ratios of females to males have been as high as 4:1 in patients with idiopathic musculoskeletal pain (Sherry & Malleson, 2002). Age has also been identified as a relevant consideration, as rates of idiopathic chronic pain in children have been noted to increase with age (Hoftun et al., 2011; King et al., 2011). Moreover, a reciprocal relationship exists between chronic pain and sleep, with increased pain associated with sleep problems, and sleep problems associated with increased pain (Puzino & Mindell, 2015). An association has also been noted between familial chronic pain and child chronic pain, with children with chronic pain more often having a "pain model" in their family (Aasland, Flatö, & Vandvik, 1997). The association between family chronic pain and child chronic pain has been

considered in the context of physiological characteristics, such as variation in pain thresholds, and dynamics such as social modeling (Schechter et al., 2010).

Psychological Considerations

Anxiety and depression have been found to be particularly prevalent in children with idiopathic chronic pain. Researchers have estimated the prevalence of anxiety disorders among children with chronic pain to be 12-18% (Knook et al., 2011). Depressive symptomology is also common, with 70.7% of children with chronic pain displaying at least mildly elevated symptoms, and 8% displaying severely elevated symptoms. Notably, particular coping strategies, such as the tendency toward internalizing and/or catastrophizing, have been found to be predictive of depression and anxiety in children with chronic pain. However, seeking social support tends to have an inverse relationship with depression and anxiety symptoms and utilizing behavioral distraction tends to have an inverse relationship with depression (Eccleston et al., 2004).

Social Considerations

Parent and peer dynamics also relate to the manifestation and trajectory of idiopathic chronic pain. The way a parent responds to a child's expression of pain is associated with the level of functional disability and somatic symptoms the child displays. Protective responding has been found to relate to increased functional disability as well as non-specific somatic symptoms. Minimization is noted to be positively correlated with somatic symptoms, while encouragement and monitoring has been associated with increased functional disability (Claar, Simons, & Logan, 2008). Furthermore, it has been suggested that the association between family chronic pain and child chronic pain may be in part due to social modeling (Schechter et al., 2010). Peer models may also be present, and adolescents with chronic pain have been found to

be more likely to have significant others who are also chronic pain sufferers than peers without chronic pain are (Merlijn et al., 2003).

Treatment Considerations

Treatment of idiopathic chronic pain typically uses a multi-disciplinary approach, with a primary goal of managing pain and restoring functional skills, and secondary goal of pain reduction. Patients are typically initially referred by a physician for outpatient physical therapy, occupational therapy, and/or psychotherapy. Exact programming can vary depending on how severely the patient's functioning has been impacted, and which domains of functioning appear to be in need of treatment. For many children with idiopathic chronic pain, a combination of physical, occupational, and psychological treatment is indicated. Physical therapy and occupational therapy address regaining physical strength and the ability to complete daily activities. Psychotherapy typically integrates cognitive behavioral therapy techniques with pain management strategies and includes psychoeducation for the parent and patient, identification of stressors and distortions, development of appropriately coping and pain management strategies. Biofeedback is also sometimes used as part of treatment, potentially in conjunction with techniques such as relaxation training, to increase the patient's awareness of their physiological responses to stress as well as to pain, and to better regulate their body's responses through the use of appropriate coping strategies (Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010; Sherry, 2011). If a patient completes outpatient therapies without substantial improvement in functionality, the patient may then be referred for intensive inpatient treatment. As part of the inpatient treatment model used in the present evaluation, patients are admitted to a rehabilitation hospital for two weeks or more, with daily physical, occupational, and psychotherapy (Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010; Sherry, 2011).

Problem Statement

Existing research has identified associations between various biological, psychological, and social factors and pediatric idiopathic chronic pain. However, the ways in which constellations of biological, psychological, and social factors influence treatment outcomes among children with idiopathic chronic pain have not been thoroughly explored. The purpose of this study was to investigate the ways in which parent and child factors are related to child benefit from an inpatient chronic pain treatment program. Although existing literature provides information regarding multiple antecedents and consequences associated with idiopathic chronic pain, little information is available about the ways in which those factors affect a child's potential benefit from treatment. Thus, the following research questions were developed to provide further insight on ways in which parent response to pain as well as the patient's symptoms of depression and anxiety may impact the patient's confidence in their ability to manage their pain.

Research Question 1

What relationships exist among patient levels of depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels? This exploratory analysis examined the relationships among variables. It was hypothesized that significant correlations would exist among levels of depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels.

Research Question 2

While controlling for initial levels of coping, do levels of post-treatment coping differ between children with elevated levels of depression and children with normal levels of depression? Research has found that children with chronic pain who display elevated

depressive symptoms tend to exhibit greater deficits across domains, including social and adaptive functioning, along with greater overall functional disability (Gauntlett-Gilbert & Eccleston, 2007), which indicates difficulties coping with pain in order to participate in daily activities. Therefore, it was hypothesized that while controlling for initial coping levels, children with normal levels of depression would display significantly higher levels of coping after treatment than children with elevated levels of depression.

Research Question 3

While controlling for initial levels of coping and depression, do levels of post-treatment coping differ between children with elevated levels of anxiety and children with normal levels of anxiety? Previous research has found that, after controlling for the effects of depression, anxiety alone does not account for variability in social functioning, adaptive skills, or functional disability (Gauntlett-Gilbert & Eccleston, 2007). Therefore, it was hypothesized that while controlling for initial coping levels and levels of depression, there would not be a significant difference in levels of coping after treatment between children with normal levels of anxiety and children with elevated anxiety.

Research Question 4

While controlling for initial levels of coping, do parent response behaviors predict levels of post-treatment coping? Previous research has suggested that a parent's tendency to distract, minimize, or protect in response to a child's chronic pain is associated with variability in the degree of functional disability displayed by the child. Specifically, high levels of protective behaviors have been associated with greater functional disability and somatic symptoms (e.g., Claar, Simons, & Logan, 2008; Connelly et al., 2010). Thus, it was hypothesized that after accounting for the effects of initial coping levels, parent response behaviors, including

distraction, minimization, and protection, would predict post-treatment coping. Research

Question 5

While controlling for initial levels of coping and depression, do parent response behaviors predict levels of post-treatment coping? Previous research has suggested that a significant interaction exists between parent response style and anxiety symptoms on functional disability. Specifically, an interaction between parent protective behavior and anxiety has been found to be associated with higher levels of disability. However, a significant interaction has not been identified between parent protective behavior and depressive symptoms (Claar, Simons, and Logan, 2008). Therefore, it was hypothesized that while controlling for initial coping levels and depression, parent response behaviors would continue to predict post-treatment coping levels.

Summary

In this chapter, the reader was provided with an overview of the characteristics and consequences of pediatric idiopathic chronic pain. An introduction to the biopsychosocial model was also provided within the context of its application to chronic pain conditions. Finally, research questions for the present study were outlined, which examined whether children and adolescents with high levels of anxiety and/or depression are less likely to benefit from inpatient treatment than children with typical levels of anxiety and depression are and how parent response to pain affects a child's benefit from inpatient treatment.

Chapter II: LITERATURE REVIEW

Idiopathic Chronic Pain in Children

Whether from a fall from a bicycle or an injury at softball practice, children commonly experience pain on multiple occasions across childhood. In some instances, pain is severe enough to merit a visit to the hospital or doctor's office. When pain is the reason for referral to a medical practice, a clinician will gather various details regarding the location, qualities, history, and intensity of the pain, as well as exacerbations or alleviations to it. A physical examination is typically conducted, and personal and family history is examined along with any physiological or psychological symptoms occurring in conjunction with the pain. At that point, various explanations for the pain may be explored, with differential diagnoses varying widely depending upon the nature of the pain (Sherry, 2011). Often, children undergo extensive medical evaluations to identify potential organic causes of their pain. However, in some cases, the pain was not necessarily preceded by an identifiable injury, or the pain continues to occur considerably longer than expected without a medical reason. After thorough evaluation, if an organic cause cannot be identified, the child is commonly diagnosed with an idiopathic pain syndrome.

Multiple diagnostic terms may be used to describe idiopathic chronic pain, with little distinction among the sub-diagnoses. As Weissmann and Uziel (2016) note, Amplified Musculoskeletal Pain Syndrome (AMPS) is a term used to characterize chronic pain syndromes of "unconfirmed etiology." Thus, AMPS is a disorder category that can be used to describe multiple syndromes. Complex Regional Pain Syndrome (CRPS), Reflex Sympathetic Dystrophy (RSD), Reflex Neurovascular Dystrophy (RND), Causalgia, and localized idiopathic pain are terms within this category and are often used somewhat interchangeably. Juvenile fibromyalgia

is also sometimes considered as a diffuse amplified pain syndrome. It has been suggested that in these conditions, pain signals are amplified, causing significantly more pain than would be expected given a particular stimulus, thus resulting in functional disability. A minor injury may precede the condition, but in some cases, it develops without any evidence of physiological damage. Additionally, conditions such as Juvenile Rheumatoid Arthritis (JRA) and Sickle Cell Disease that cause pain and deconditioning (i.e., loss of muscle tone and endurance) associated with decrease in physical activity often require similar treatment techniques for management of symptoms (Schechter, Palermo, Walco, & Berde, 2010).

Epidemiology

Chronic pain is a common complaint among children. A meta-analysis of 41 studies found significant variation in the prevalence of unexplained chronic pain among children and adolescents (King et al., 2011). Prevalence estimates can be found in Table 1.

Table 1Pain Prevalence, adapted from King et al. (2011)

| Pain type | Prevalence range |
|---------------------------|------------------|
| Headaches | 8% to 82.9% |
| Back Pain | 13.5% to 24% |
| Musculoskeletal/limb pain | 3.9% to 40% |
| Multiple locations | 3.6% to 48.8% |
| Other/general pain | 5% to 88% |

Furthermore, large scale research found that, among 7373 adolescents, 44.4% reported chronic idiopathic pain, defined as pain occurring at least weekly for 3 or more months without a known

medical cause. Musculoskeletal pain was the most common, with pain most frequently focused in the neck/shoulder region. 8.5% of the adolescents reported diffuse idiopathic pain (Hoftun, Romundstad, Zwart, & Rygg, 2011).

Biopsychosocial Model

In the late 1970s, George Engel outlined a new model for use in medicine. He criticized the "adherence to a model of disease no longer adequate for the scientific tasks and social responsibilities of either medicine or psychiatry" and noted the pitfalls of reducing medical problems to solely biological factors (Engel, 1977: p. 129). The biomedical model of the time relied on the premise that disease must be entirely explained through biochemical mechanisms. While he acknowledges the importance of the biomedical framework in propelling scientific and technological advances, he cites the exclusion of psychological and social as a significant shortcoming in need of resolution. The resolution for which Engel advocated is the biopsychosocial model.

The biopsychosocial model, as characterized by Engel, involves the following principles:

- Biochemical deviation should be considered as "necessary but not a sufficient condition for the occurrence of the human experience of the disease, the illness" (Engel, 1977: p. 131). He expresses that biological abnormalities must be considered as factors in a complex, multifaceted interaction that leads to the patient's presentation.
- The behavioral and psychosocial data provided by the patient must be adequately collected and interpreted, including use of appropriate clinical tools and skills (e.g., clinical interviewing).

- The interaction of the patient's life experiences and circumstance must be considered
 alongside psychological and biological factors to understand the onset and trajectory of
 disease.
- The clinician must evaluate how the patient views him/herself and how the patient
 believes that others perceive him/her. In particular, whether or not the patient accepts the
 social role of being ill should be considered, as such acceptance or denial may influence
 symptom presentation and reporting of symptoms.
- Treatment of disease must address biological, psychological, and social variables.
- The clinician must consider psychological and social impacts of patient-clinician interactions and ways in which the relationship with the patient can influence treatment outcomes.

Furthermore, Engel advocates that the clinician's role is to analyze the various factors contributing to a patient's presentation and to understand what aspects have led the patient to "seek medical help, adopt the sick role, and accept the status of patienthood" (Engel, 1977: p. 133).

At present, the influence of the biopsychosocial model has become increasingly evident in many medical systems and practices. It has been particularly relevant to the study, assessment, and treatment of idiopathic chronic pain. When a patient presents with pain, health service providers must analyze the nature and source of pain to better characterize it and develop evidence-based treatments. However, in the case of children with idiopathic pain, it has been suggested that patients are best served by focusing on the child's experience of the pain and "avoiding the deleterious polarization of the pain as either physical or psychogenic in origin" (Kozlowska et al., 2008, p.1). Carter and Threlkeld (2012) discuss pediatric chronic pain

conditions in terms of a spectrum, from "clearly medically delineated" (e.g., Sickle Cell Disease, Juvenile Rheumatoid Arthritis) to those that are medically unexplained (e.g., Chronic Regional Pain Syndrome, Juvenile Fibromyalgia). However, these authors argue that, rather than focusing on differentiating based on medical explanation, a patient is better served by a biopsychosocial model that considers the "multidimensional nature of pain in which biological, psychological, individual, social and environmental variables are interactive in the development, maintenance, and subjective experience of pain and disability" (Carter & Threlkeld, 2012, p.2). This perspective is congruent with the biopsychosocial model of chronic pain, which is an integrative perspective considering the innate and environmental contributors to the experience and maintenance of pain (Gatchel, Peng, Peters, Fuchs, & Turk, 2007).

For the present research, biological factors contributed to the characteristics of the sample population, as all participants underwent physical evaluations prior to admission to the inpatient treatment program to rule out the possibility of a known organic cause for the pain and were found to meet criteria for an idiopathic chronic pain diagnosis (e.g., Reflex Neurovascular Dystrophy, Complex Regional Pain Syndrome, etc.). Psychological and social factors were of particular relevance to examining the data, including analysis of the psychological factors of patient anxious and depressive symptomology as well as the social factors of parent response to pain. The biopsychosocial model and its application to the present research can be explored through consideration of the biological, psychological, and social aspects of pain and the ways in which they interact.

Biological Factors

Pain

Pain can be defined in a variety of ways, often varying based on the philosophical, occupational, or theoretical perspective of the individual defining the term. The International Association for the Study of Pain (IASP) defines pain as "an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage (Johan & Loeser, 2012). Turk and Okifuji (2010) further note distinctions among acute and chronic pain, noting that while traditional definitions have focused on the length of time the pain persists, consideration of both time and physical pathology is important. These authors conceptualize acute pain as, "Pain elicited by the injury of body tissues and activation of nociceptive transducers at the site of local tissue damage...In general, the state of acute pain lasts for a relatively limited time and remits when the underlying pathology resolves." (Turk & Okifuji, 2010, p. 14). Conversely, chronic pain "may be elicited by an injury or disease but is likely to be perpetuated by factors that are both pathogenetically and physical remote from the originating cause. Chronic pain extends for a long period of time and/or represents low levels of underlying pathology that does not explain the presence and extent of pain" (Turk & Okifuji, 2010, p. 14). The authors further note that idiopathic chronic pain may occur in part due to changes within the nerves and sensitivity of the nervous system. Furthermore, they note that genetic and psychosocial factors may affect an individual's propensity to develop chronic pain. They conclude, "Just as the brain is modified by experience, especially in early life, the brain may alter the way noxious information is processed to reduce or augment its impact on subjective awareness." (Turk & Okifuji, 2010, p. 14).

Gender

Females are significantly more likely to present with idiopathic chronic pain than males are (e.g., Eccleston, Crombez, Scotford, Clinch, & Connell, 2004; Hoftun, Romundstad, Zwart, & Rygg, 2011; King et al., 2011; Schechter et al., 2010; Sherry & Malleson, 2002). Regarding idiopathic musculoskeletal pain, estimated ratio of females to males have been as high as 4:1 (Sherry & Malleson, 2002). Furthermore, in a large-scale study in Norway, twice as many girls endorsed headache/migraine pain as boys, and three times as many girls endorsed abdominal pain. The gender difference among these youth was most pronounced at age 16 (Hoftun et al., 2011).

Age

Generally, rates of idiopathic chronic pain appear to increase with age. A meta-analysis of 41 studies indicated that headaches, back pain, and musculoskeletal/ limb pain were more common in older children, while abdominal pain was more common in younger children (King et al., 2011). Age differences for multiple pains and other/general pain in that investigation were unclear. Hoftun et al. (2011) examined a population of 7373 children between the ages of 13-18 in Norway as part of the 2008 Nord-Trøndelag Health "HUNT" Study, in which questionnaires regarding health-related issues were completed by in the school setting. Data used for Hoftun et al.'s research came from a pain questionnaire, which asked if the child had had idiopathic pain during the last three months, and, if so, where and how often the pain occurred, and a subjective disability index. As discussed previously, results revealed that 44.4% of children reported pain in any location at least once a week during the last three months. Notably, the research found that, in general, pain prevalence was higher in older adolescents, with the exception of lower extremity pain, which was more common in younger (13-15 year old) adolescents than in older (16-18 year old) adolescents. Sherry and Malleson (2002) note that the average age of onset for

idiopathic musculoskeletal is 12 years. Though it can present in children who are younger, they caution against diagnosis before age 7 years.

Health Impacts. Sleep is a particular area commonly affected by chronic pain. A reciprocal relationship between sleep and chronic pain has been noted, with increased pain leading to sleep difficulties, and sleep difficulties exacerbating pain (Puzino & Mindell, 2015). Hoftun et al. (2011) found that 33% of children with idiopathic pain in at least one location reported that their pain caused difficulties falling asleep and/or disturbed their sleep. Additionally, approximately 50% of children with idiopathic chronic pain have difficulty sitting for long periods of time, and 60% have difficulty in daily activities during leisure time (Hoftun et al., 2011).

Parent pathology. As compared to those with medically explained pain, children with idiopathic chronic pain are significantly more likely to have a familial "pain model" (Aasland, Flatö, & Vandvik, 1997). It has been suggested that a combination of physiological characteristics, such as variation in pain thresholds, and psychological characteristics, such as social modeling, likely contribute to this pattern (Schechter et al., 2010). In a sample of young adults, Lester, Lefebvre, and Keefe (1994) found that a greater family history of pain, as characterized through number of family members reported to have migraine headaches, low back pain, or arthritis was associated with experiencing pain in multiple locations as well as with increased functional problems as the result of pain (e.g., interference with daily activities and job and/or school work). In addition to increased history of chronic pain, parents of children with chronic pain often display a high degree of emotional distress. On a depression and anxiety screener, (Christopher Eccleston et al., 2004) found that 40% of parents of children with chronic

pain were above the cutoff for depressive disorders, and 62% were above the cutoff for anxiety disorders.

Psychological Factors

Psychological characteristics, particularly a predisposition to anxiety and depressive symptoms accompanied by exposure to significant stress, are often considered as potential antecedents of idiopathic pain. Children with frequent complaints of pain are more likely to display symptoms of psychopathology (Campo, Comer, Jansen-McWilliams, Gardner, & Kelleher, 2002). Furthermore, as compared to children with juvenile chronic arthritis (JCA), children with idiopathic musculoskeletal pain in particular report more school stress and display more persistent psychiatric conditions over time (Aasland et al., 1997).

Psychopathology. Anxiety and depression tend to be particularly common in the chronic pain population (Wasan, Sullivan, & Clark, 2010). Significantly higher rates of anxiety in children with idiopathic chronic pain have been identified as compared to non-clinical groups (Christopher Eccleston et al., 2004). Various anxiety disorders are common; Aasland et al. (1997) found that 21% of patients with idiopathic pain presented with separation anxiety, 16% with overanxious disorder, 5% with social phobia, and 5% with OCD. Another study estimated the prevalence of anxiety disorders among children with idiopathic chronic pain according to parent report of symptoms to be 18%, with specific phobias being the most common. Notably, when examining the children's report of symptoms, the researchers found the estimate to be lower, at 12% (Knook et al., 2011).

Depression has also been identified among children with idiopathic chronic pain, though estimates are somewhat variable depending on the tools used to assess for depression and the current diagnostic criteria. Among children seen at a tertiary chronic pain clinic, 70.7%

displayed at least mildly elevated scores on the Children's Depression Inventory (CDI) (Kovacs, 1992), with 8% falling in the severely elevated range (Christopher Eccleston et al., 2004). Similarly, Aasland et al. (1997) found that 26% of children with idiopathic musculoskeletal pain met criteria for dysthymic disorder upon admission to the rheumatology department at a hospital. Knook and fellow researchers (2011) found a lower estimate when examining parent report, with 4.5% of children with idiopathic chronic pain in their study presenting with an affective disorder. However, child report of symptoms indicated a higher estimate of 19% presenting with an affective disorder.

Other psychological traits have also been noted among children with idiopathic chronic pain. Merlijn et al. (2003) found that adolescents with idiopathic chronic pain endorsed more feelings of insufficiency and negative fear of failure, and that they display greater characteristics of neuroticism. Among children referred to a tertiary specialist chronic pain clinic, children who exhibit depressive symptoms along with chronic pain displayed poorer functioning across social functioning, adaptive functioning, functional disability, and school attendance. Conversely, anxiety did not account for the variance in any domain after depression was accounted for (Gauntlett-Gilbert & Eccleston, 2007).

It has long been debated whether depression may be an antecedent or consequence of chronic pain. A review of 83 studies of chronic pain and depression, primarily in adults, indicated greater support for the consequence and scar hypotheses than for the antecedent hypothesis. The consequence hypothesis indicates that depression occurs as a result of chronic pain, while the scar hypothesis suggests that a genetic predisposition to depression may lead to increased incidence of depression in chronic pain patients (Fishbain, Cutler, Rosomoff, & Rosomoff, 1997).

Coping. Particular coping strategies tend to be more common in the pediatric chronic pain population. Carver, Scheier, and Weintraub (1989) discuss the traditional distinction between problem-focused coping, in which an individual actively attempts to change the stressor, and emotion-focused coping, in which an individual attempts to reduce or manage the emotional consequences of the stressor. The authors note that this distinction is overly simplified, and argue that coping can be discussed in terms of 13 different domains, which are listed below:

- 1. Active coping: actively attempting to change the stressor or its consequences.
- 2. *Planning:* considering how to address a stressor, including developing a plan to address the problem.
- 3. Suppression of competing activities: prioritizing problem solving related to the stressor over other activities.
- 4. Restraint coping: waiting to act on the stressor until the appropriate time.
- 5. Seeking social support for instrumental reasons: seeking input or information from others.
- 6. Seeking social support for emotional reasons: obtaining social support in the form of empathy and sympathy.
- 7. Focusing on and venting of emotions: focusing on and expressing the emotions associated with stress.
- 8. Behavioral disengagement: giving up on goals with which a stressor interferes.
- 9. *Mental disengagement*: distraction from the problem.
- 10. Positive reinterpretation and growth: also known as positive reappraisal, which is focused on managing the emotions associated with a stressor.

- 11. *Denial:* may include minimization of distress and/or the creation of further problems if the primary stressor cannot be ignored.
- 12. Acceptance: accepting of a stressor or stressful situation.
- 13. Turning to religion: finding support in religious sources.

Rosenstiel and Keefe (1983) distinguished among three broader categories of coping strategies among patients with chronic back pain. These factors included (1) cognitive coping and suppression, (2) helplessness, and (3) diverting attention or praying. Reid, Gilbert, and McGrath (1998) developed the *Pain Coping Questionnaire* specifically assessing coping with pain. Among children and adolescents, the researchers identified three primary domains of coping with pain, including approach, problem-focused avoidance, and emotion-focused avoidance. Eight sub-domains were also indicated, including information seeking, problem solving, seeking social support, positive self-statements, behavioral distraction, cognitive distraction, externalizing, and internalizing/catastrophizing (Reid et al., 1998).

Given that coping can happen in a positive manner as well as a maladaptive one, as in helplessness, it is important to consider the effects of particular coping approaches in individuals with chronic pain. Among children and adolescents with chronic headaches and with arthritis, Reid, Gilbert, and McGrath (1998) found that increased levels of emotion-focused avoidance were associated with increased emotional distress. Additionally, in participants with headaches, emotion-focused avoidance was related to less coping effectiveness, and in those with arthritis, it was associated with increased pain. Furthermore, an inverse relationship was found in those with headaches between approach coping and level of disability (Reid et al., 1998). Children and adolescents with chronic pain have been found to be significantly more likely to engage in emotion-focused avoidance coping strategies than peers without chronic pain are (Merlijn et al.,

2003). However, notable relationships have been found among psychological factors and coping methods. An internalizing/ catastrophizing approach to coping has been found to be predictive of depression and anxiety in children with chronic pain. However, seeking social support as a coping mechanism was associated with decreased levels of anxiety and depression. Additionally, behavioral distraction was inversely related to depression (Christopher Eccleston et al., 2004). Furthermore, Vervoort, Goubert, Eccleston, Bijttebier, and Crombez (2005) conducted two cross-sectional studies in which they examined somatic complaints, pain-related disability, catastrophic thinking about pain, pain severity, and negative affect in a sample of school children and in a sample of children with chronic pain. Among the community sample of school children, 54.9% reported at least one significant somatic complaints (e.g., headaches, stomachache pain, limb pain, sore muscles, and/or nausea/upset stomach) within the last two weeks, and 3.8% reported constant pain. Pain catastrophizing and negative affect were found to be significant predictors of somatic complaints and functional disability, and pain catastrophizing further predicted pain severity. Notably, pain catastrophizing was found to account for approximately 93% of the relationship between negative affect and somatic complaints, and approximately 82% of the relationship between negative affect and functional disability. Upon examining the second sample of children, who had a history of chronic pain and at the time of data collection were admitted to a pediatric hospital, the researchers found 90.7% reported at least one severe somatic symptom. As with the community sample, pain catastrophizing emerged as significant predictor variable for somatic complaints, pain severity, and functional disability. Negative affect also predicted somatic complaints, though pain catastrophizing accounted for approximately 85% of the relationship between negative affect and somatic complaints. Thus, in both a community sample of children and in a sample of children

hospitalized for pain-related causes, the tendency to catastrophize pain concerns has a significant effect on a child's level of functional disability as well as self-report of pain severity (Vervoort, Goubert, Eccleston, Bijttebier, & Crombez, 2005). Furthermore, feelings of helplessness that may be found in individuals who tend to catastrophize pain concerns can also play a role, as holding the belief that one has minimal control over his or her pain is associated with increased pain intensity (Miró, Huguet, & Jensen, 2014).

Social Factors

Parent Response to Pain

A parent's response to their child's pain can influence the degree of disability the child displays, as well as the symptoms that he or she endorses. Parent responses are often characterized as protective, minimizing, or distracting/monitoring. *Protective* responses include behaviors such as paying more attention to the child than usual, completing tasks for the child, taking the child to the doctor or administering medicine, and providing the child with special privileges. Minimizing responses include behaviors such as telling the child that nothing can be done, avoiding paying attention to the child, and telling the child to not worry so much about their pain. Conversely, distracting and monitoring responses include behaviors such as reassuring the child that he or she will be okay, encouraging the child to engage in activities, and checking on how the child feels (Van Slyke & Walker, 2006). Claar, Simons, and Logan (2008) found that the way a parent responds can be related to the child's level of functional disability and somatic symptoms, particularly when the child has heightened levels of depression or anxiety. Specifically, increased parent protective behavior was related to increases in the child's level of functional disability as well as reported non-specific somatic symptoms (e.g., "weakness" without an organic etiology). Parent minimization was positively correlated with

somatic symptoms, but not functional disability, whereas encouragement and monitoring behaviors were associated with increased functional disability, but not an increase in somatic symptoms. When considering psychological characteristics of the child, it was found that for children with higher levels of anxiety, parental protective behavior was associated with increased functional disability. However, for children with lower levels of anxiety, parent protective behavior was not predictive of functional disability. Similarly, high anxiety or depression along with high levels of parent minimization was associated with an increase in somatic symptoms, while in children with low levels of anxiety or depression, parent minimization did not predict somatic symptoms. Moreover, while parent protective behavior and child's symptoms of depression predicted degree of functional disability, the interaction between the two was not significant. Parent encouragement/monitoring did not significantly interact with psychological variables in impact on somatic symptoms nor functional disability. Furthermore, Kaczynski, Claar, and Logan (2009) found that protective parental responding was associated with increased functional disability. Similarly, Connelly and fellow researchers (2010) found that when parents of children with Juvenile Idiopathic Arthritis engaged in protective responding to pain, children displayed decreased activity levels and mood. However, distracting/monitoring responses were associated with increased activity levels, but only in children with severe forms of the condition. Furthermore, dysfunctional parent-child interaction style has been found to predict poorer social and adaptive functioning (i.e., school performance, peer relationships, family relationships, and home duties/self-care) in children with chronic pain (Gauntlett-Gilbert & Eccleston, 2007).

Family Functioning

Elevated parenting stress is common in chronic pain samples (Christopher Eccleston et al., 2004; Gauntlett-Gilbert & Eccleston, 2007). (Christopher Eccleston et al., 2004) found 39%

of parents to be in the high range of distress, with adolescent age and duration of pain significantly predicting parental distress. Moreover, adolescent age, pain duration, and adolescent symptoms of depression predicted parent-child dysfunctional interaction as well as difficult child scores on the PSI/SF. 46.8% were in the high range on the difficult child subscale, and 36.4% were in the high range for parent-child dysfunctional interaction. Levels of parent-child dysfunctional interaction and difficult child were also found to impact child coping and psychological variables, with parent-child dysfunctional interaction being associated with decreased positive self-statements and behavioral distraction and increased depression.

Similarly, the difficult child subscale was negatively correlated with positive self-statements and seeking social support, and positively correlated with adolescent depression, anxiety, and externalizing coping mechanisms.

Peer Response to Pain

Minimal research has been done on the role of peer response to pain. However, there is some evidence that the role of peer response differs from that of parental response. Merlijn and others (2003) found that adolescents receive more reinforcement from parents when in pain, while they receive greater reinforcement from peers when in pain-free situations. Notably, these researchers also found that parents of children without chronic pain provided greater reinforcement in pain situations than parents of children with chronic pain did. However, adolescents with chronic pain report lower perceived social acceptance as compared to peers without chronic pain. Additionally, adolescents with chronic pain were somewhat more likely to report having significant others who were also chronic pain sufferers than peers without chronic pain were (Merlijn et al., 2003).

Chronic pain can lead to a variety of social challenges associated with factors such as decreased participation in activities, psychological consequences such as anxiety or depression, and embarrassment or fear of feeling different from peers. Hoftun et al. (2011) found that adolescents with chronic pain report difficulties engaging in leisure activities, sitting during class, sleeping, and participating in physical exercise, with impact on leisure time being the most common complaint. Increased number of pain locations was associated with an increase in the level of subjective disability. Among children with chronic pain, Konijnenberg et al. (2005) found that those with musculoskeletal pain display the greatest impairment in physical functioning, while those with headache pain experienced the greatest social disruption. In the sample overall, 40% of children displayed impaired social functioning, attributed primarily to decreased contact with peers. Miró et al. (2014) found that children with chronic pain displayed decreased physical and psychological functioning, increased school absence, and increased reliance on medication and medical care for pain relief as compared to typical peers.

Consequences of Idiopathic Chronic Pain

Idiopathic chronic pain commonly results in functional disability across multiple domains. In some cases, there is a "the chicken or the egg" scenario in which it is difficult to determine whether a phenomenon is an outcome of chronic pain, or if it was a pre-existing condition or predisposition. In addition to the aforementioned factors that act as antecedents or potentially have a reciprocal relationship with pain, chronic pain is further associated with impacts on daily functioning, school, and healthcare utilization along with financial burden.

Functional Disability

Functional disability is a term used to describe the impact of a medical condition on an individual's participation and functioning in daily activities and roles. Physical, psychological,

and social functioning can be considered as part of functional disability (Walker & Greene, 1991). In cases of chronic pain, functional disability can be evident across school, social relationships, and mental health. Measures of functional disability commonly assess domains such as school absences and performance, symptoms of internalizing and externalizing behavior problems, medication use, participation in activities of daily living, participation in leisure activities, and somatic symptoms (Walker & Green, 1991). Hoftun et al. (2011) characterized functional disability through a series of 5 forced choice self-report questions, which included: (1) I have difficulties falling asleep because of pain and/or pain disturbs my sleep; (2) Because of pain I have difficulties sitting during a lesson; (3) Pain disturbs me if I walk more than 1 km; (4) Pain disturbs me during physical exercise class; and (5) All things considered, has pain made it difficult to do daily activities in leisure time?. They found that 67% of children who reported pain in at least five locations displayed "maximal disability," or agreement with all 5 statements. Furthermore, 58.4% of those reporting diffuse musculoskeletal pain indicated maximal disability. Frequency of pain was also found to be a contributing factor, as 66.9% of those who reported idiopathic pain almost daily in 2 or more locations reported disability across all five domains assessed (Hoftun et al., 2011).

School

Academic Performance. Some research as indicated that unrealistic worries about school performance are significantly more common in children with idiopathic chronic pain than in those with juvenile arthritis. Additionally, those with idiopathic pain exhibit significantly more learning difficulties than those with arthritis (Aasland et al., 1997). Children with idiopathic chronic pain are at increased risk for changes in academic functioning. In one study, parent report indicated that 44.3% of the adolescents with idiopathic pain had displayed a decline

in grades since the onset of their pain (Logan, Simons, Stein, & Chastain, 2008). Conversely, while examining academic data collected from 220 adolescents with a three-month history of idiopathic pain who were evaluated at a tertiary care chronic pain clinic, Logan et al. (2008) noted that the self-perceived academic competence among youth with chronic pain was similar to that in the normative population. Teacher perceived academic competence was found to be high, and almost 80% of the sample were rated one standard deviation or more above the mean in terms of school adjustment. Additionally, teacher perceptions of academic competence were found to be unrelated to pain severity or duration. However, teacher perceptions of academic competence were found to be related to students' dominant type of pain complain, with increased academic competence perceived in children displaying neuropathic pain than in children with migraine. The authors note that self and teacher perceived academic competence is consistent with existing characterizations of children with chronic pain as being motivated and high achieving students (Logan et al., 2008).

Attendance. Estimates of the impact of chronic pain on school attendance vary significantly. For example, in a sample of 110 patients referred to a tertiary specialist chronic pain clinic in the UK, 39% were not attending school, and 37% were attending only partial school days (Gauntlett-Gilbert & Eccleston, 2007). Conversely, patients seen at an outpatient multidisciplinary pediatric pain clinic in the United States had missed an average of 7.85 days of school prior over the 3 months prior to their intake appointment (Ho et al., 2008). Other estimates obtained from a tertiary care clinic indicated that over 1/3 of children with chronic pain had missed more than 25% of the school days in the preceding month (Logan et al., 2008). Notably, pain intensity has been found to be predictive of overall functional disability, but not social/adaptive functioning nor school attendance (Gauntlett-Gilbert & Eccleston, 2007).

Healthcare Utilization and Financial Burden

Chronic pain results in increased healthcare utilization as well as financial burden for patients and their families. Patients seen at an outpatient multidisciplinary pediatric pain clinic had spent an average of 28.54 hours at medical appointments over the past three months, resulting in multiple missed school days for the patients and missed workdays for their parents. Additionally, the average financial cost of outpatient visits (e.g., physical therapy, psychotherapy, medical appointments) to the family over that time was \$1,761.06 (Ho et al., 2008). Notably, management of idiopathic chronic pain may have greater financial implications than management of medically explained pain. In a study of adolescents with idiopathic chronic pain in the United Kingdom, average costs including direct and indirect (e.g., missed work) costs were approximately £14160 for adolescents with idiopathic pain conditions, while costs averaged £4495 for children with rheumatic diseases (e.g., Juvenile Idiopathic Arthritis) (Sleed et al., 2005).

Treatment

As discussed previously, the process leading up to diagnosis of idiopathic chronic pain is often characterized by visits with multiple specialists to evaluate potential organic causes of the pain (Sherry, 2011). Once the pain has been characterized as idiopathic, the child is typically referred for some combination of outpatient physical therapy, occupational therapy, and psychotherapy with a focus on pain management. As Sherry (2011) notes,

Treatment should have two goals: restoration of function and relief of pain. Anything less is not ideal, although, because pain is subjective and not directly amenable to specific treatment, there are patients in whom restoration to full function without total pain relief has to be accepted. Helping the child develop skills to cope with the pain is often

effective in relieving distress and dysfunction, even if the pain persists (Sherry, 2011: p. 726).

If the child participates in outpatient therapies for a period of time without benefit, inpatient treatment may be indicated. Inpatient treatment involves admission to a rehabilitation hospital for two or more weeks, with full days of intensive therapies. Physical therapy and occupational therapy address regaining physical strength and the ability to complete daily activities. For example, goals are created related to completing developmentally appropriate aerobic exercise, engaging in a developmentally appropriate level of independence in activities of daily living, and making functional gains in areas that are relevant to the patient, such as working toward returning to a sport or activity. Additionally, for patients with allodynia, desensitization and reactivation treatments may be completed. Biomechanical issues related to things such as flexibility and posture may also be addressed (Landry et al., 2015). Psychotherapy is another key component of treatment of idiopathic chronic pain, and typically integrates cognitive behavioral therapy (CBT) techniques with pain management strategies and includes psychoeducation for the parent and patient, identification of stressors and distortions, development of appropriately coping and pain management strategies. CBT is a psychotherapeutic approach that considers the interaction of cognition, behavior, and emotion, and focuses on supporting clients in identifying and changing maladaptive thought processes and behaviors to improve their emotional functioning. CBT has been found to be successful for treatment of anxiety in pediatric populations, with evidence for positive long-term effects. A study of 52 individuals who were diagnosed with anxiety disorders as children and received CBT indicated that gains were made through treatment, at that an average of six years post-treatment, 85.7% no longer met criteria for an anxiety disorder as adolescents and young adults (Barrett,

Duffy, Dadds, & Rapee, 2001). CBT has also been identified as an efficacious treatment for depression in pediatric patients (Reinecke, Ryan, & DuBois, 1998). However, some authors note that much of the research on effectiveness of CBT for treatment of depression has focused on adult populations, and that different benefits may be seen in children. For example, Yang et al. (2017) found in a meta-analysis of randomized controlled trials that, while children treatment with CBT display greater benefit than children in non-treatment groups, they do not differ significantly from children in placebo or wait list groups. However, the authors note that few studies have been completed that address this topic with a high level of experimental control, and only nine studies were included within their meta-analysis (Yang et al., 2017).

In addition to the treatment of anxiety and depression, CBT is becoming an increasingly common approach to treatment of chronic pain. Biofeedback is also sometimes used alongside CBT as part of treatment, potentially in conjunction with techniques such as relaxation training, to increase the patient's awareness of their physiological responses to stress as well as to pain, and to better regulate their body's responses through the use of appropriate coping strategies (Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010; Sherry, 2011). A meta-analysis examining data from 25 research studies found that psychological treatment has positive effects for children and adolescents with types of chronic pain, including headache, abdominal pain, and fibromyalgia. The treatments examined included CBT, relaxation therapy, and biofeedback. Treatment with any of the three modalities resulted in a statistically significant effect on pain reduction. Few studies in the sample included assessment of effect on emotional functioning and level of functional disability. among those that did, effect sizes were found to be small and not statistically significant. The authors note that further work is needed to examine the effect of

psychotherapeutic treatment on these domains among the pediatric chronic pain population (Palermo, Eccleston, Lewandowski, de C. Williams, & Morley, 2010).

Summary

In this chapter, the reader was provided with a review of existing literature regarding the biological, psychological, and social characteristics of pediatric idiopathic chronic pain. The biopsychosocial model was discussed, along with the ways in which it can specifically be applied to consideration of chronic pain. Finally, consequences of idiopathic pain along with typical treatment modalities were addressed.

Chapter III: METHOD

Participants

Participants included children and adolescents admitted to a chronic pain inpatient treatment program in Pittsburgh, Pennsylvania from July 2011 to December 2016. Admissions to the inpatient treatment program included patients with idiopathic chronic pain that did not resolve with outpatient occupational therapy, physical therapy, and/or psychotherapy treatments. Typically, patients were referred to the program by a Physical Medicine and Rehabilitation (PM&R) physician. To be included in the present research, the patient must be a patient who completed the inpatient chronic pain program. Participants with an age below 8 years or above 17 years at time of admission or those with significant missing data for multiple key variables were excluded. Those with a significantly elevated inconsistency index on the MASC were also excluded from relevant analyses. Additionally, only the first admission was included for patients with multiple admissions. After exclusionary criteria were applied, 78 participants were included in the sample. Of the 78 participants included in analysis, 82.1% were female. Average age at admission was 14.66 (SD = 9.47).

Upon admission to the inpatient program, patients began a structured treatment program that included physical therapy, occupational therapy, and psychotherapy. Patients participated in a structured treatment protocol that included 12 hours of physical therapy per week, 12 hours of occupational therapy per week, 5 hours of individual psychotherapy peer week, and 3 hours of group psychotherapy per week. Patients participated in full days of therapy on weekdays and partial days of therapy on Saturdays, with no therapies on Sundays. Additionally, parents participated in individual or family sessions with staff as needed to provide education on the

child's condition and appropriate strategies for home. Length of admission varied depending on each patient's initial level of deconditioning and his or her response to treatment.

Measures

As part of the inpatient treatment program protocol, patients and their parent or guardian completed assessment packets at time of admission. Additionally, patients completed an additional assessment packet at time of discharge. For the present research, the following scales were used from admission and discharge packets completed by each patient: Pain Self Efficacy Questionnaire (PSEQ) (Nicholas, 1989), Children's Depression Inventory (CDI) (Kovacs, 1992), and Multidimensional Anxiety Scale for Children (MASC) Self Report (J. S. March, 2012). From the parent completed admission packet, the following measure was used: Adult Responses to Children's Symptoms (ARCS) (Van Slyke & Walker, 2006).

Pain Self Efficacy Questionnaire

The Pain Self Efficacy Questionnaire (PSEQ) is a 10-item questionnaire in which patients rate how confident they are that they can do particular activities despite the pain. Items include statements such as, "I can enjoy things, despite the pain.", "I can still do many of the things I enjoy doing, such as hobbies or leisure activity, despite pain.", "I can cope with my pain without mediation." Each item is rated on a scale of 0 to 6, with 0 indicating "not at all confident" and 6 indicating "completely confident." Summed scores then result in a total overall score. With regard to the psychometric properties of the PSEQ, exploratory and confirmatory analysis has been found to support a single factor structure. Internal consistency has been found to be high (Cronbach's alpha = .90 to .92). Additionally, test-retest reliability has been found to be adequate, with an intraclass correlation coefficient of 0.76. Moreover, convergent validity with other measures has also been established, with statistically significant correlations in the

expected directions with the Pain Disability Index (r = -.518), Roland Disability Questionnaire (r = -.649), Tampa Scale of Kinesiophobia 11 Items (r = .472), McGill Pain Questionnaire – Affective Scale (r = -.409), Rand-36 Item Health Survey – Physical Functioning (r = .315), Rand-36 Item Health Survey – Social Functioning (r = .489), and verbal report of pain intensity on an 11 point scale (r = -.310) (van der Maas, de Vet, Koke, Bosscher, & Peters, 2011).

Children's Depression Inventory

The Children's Depression Inventory (CDI) is a rating scale that assesses for depressive symptomology in children aged 7 through 17 years. The self-report form of the CDI was used for the present research. This measure yields T scores in the domains of negative mood, interpersonal problems, ineffectiveness, anhedonia, and negative self-esteem, as well as a CDI Total T score. Reliability and validity of the CDI has been well-established. In terms of reliability, internal consistency estimates using Cronbach's alpha have ranged from 0.71 to 0.89 (Kovacs, 1992). Additionally, test-retest reliability has been found to be .81 (Masip, Amador-Campos, Gomez=Benito, & del Barrio, 2010). Regarding validity, high concurrent validity has been identified, with strong correlations with the Reynolds Adolescent Depression Scale (RADS) (r = .81) and the Reynolds Child Depression Scale (RCDS) (r = .76), as well as with the anxiety/depression (r = .64) and internalization scales (r = .63) of the Youth Self Report (Masip, Amador-Campos, Gomez-Benito, & del Barrio, 2010).

Multidimensional Anxiety Scale for Children

The Multidimensional Anxiety Scale for Children (MASC) is a rating scale that examines anxiety-related symptoms across the subscales of Social Anxiety, Humiliation/Rejection,

Performance Fears, Obsessions and Compulsions, Physical Symptoms, Panic, Tense/Restless, and Harm Avoidance. The MASC is designed for use in children and adolescents aged 8 to 19

years. Self-report forms were used in the present study. The scale yields T Scores for each domain as well as a MASC Total T Score. The MASC has been found to have adequate reliability and validity. In terms of reliability, the internal consistency has been found to be strong for the self-report form Total Score (coefficient alpha= .90). Furthermore, estimates of test-retest reliability for the self-report form are favorable, with average intraclass correlation coefficients of .785 at a three-week interval and of .933 at a three-month interval. Moreover, convergent validity has been established using the RCMAS, with moderate correlations between the Revised Children's Manifest Anxiety Scale (RCMAS) and MASC Self Report (*r*=.633) (John S. March, Parker, Sullivan, Stallings, & Conners, 1997a).

Adult Responses to Children's Symptoms

The Adult Responses to Children's Symptoms (ARCS) is a parent-completed scale that examines parent responses to children's symptoms. Parents provide responses on a 5-point scale that ranges from 0 (never) to 4 (always). Raw scores are obtained for the following domains: Protect, Minimize, and Encourage/Monitor. Confirmatory factor analysis has established the validity of the three factor ARCS model (Claar, Guite, Kaczynski, & Logan, 2010). For the present study, it was originally proposed that each subscale would be computed as a mean score, and whichever is highest would be considered the dominant parent response style. However, as discussed in Chapter 4, an alternative approach, using the total raw scores, was implemented due to insufficient group membership using the method originally proposed.

Research Design

Dependent Variables

Benefit from Program. A patient's benefit from the inpatient program was characterized by the Pain Self Efficacy Questionnaire (PSEQ) at time of discharge.

Independent Variables

Parent Response to Pain. The Adult Responses to Children's Symptoms (ARCS) was used to examine parent response to pain. Specifically, the raw scores for the Protect, Distract, and Minimize scales were used.

Depression. Depression was characterized using the Total T-Score from the CDI Self Report form, collected at time of admission.

Anxiety. The Total T-Score from the MASC Self Report, collected at time of admission, was used as a measure of anxiety.

Covariates

In addition to the variables noted above, the PSEQ total score at time of admission was analyzed as a covariate.

Procedures

Following approval from Duquesne University Internal Review Board (IRB), an Information Systems request was completed for the data to be mined from the electronic records, and the dataset was then sent to an honest broker employed by the hospital for deidentification. Data to be accessed included results of assessment packets completed by patients and their parents or guardians at time of admission and assessment packets completed by patients at time of discharge. Additionally, select data recorded by the treating psychology clinician during the intake interview and discharge conference was accessed in order to identify pain ratings, antecedents of the pain condition, family history of chronic pain, and reason for discharge. In addition, demographic characteristics (i.e., age, gender) were accessed from the patients' electronic medical record. Once data was accessed and deidentified, it was provided

electronically to the principal investigator for the present study and was then entered into SPSS for analysis.

Data Analysis

Five research questions and corresponding hypotheses were explored, as discussed below.

Research Question 1

What relationships exist among patient levels of depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels? This exploratory analysis examined the relationships among variables. It was hypothesized that significant correlations would exist among levels of depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels. Research Question 1 was examined by calculating the Pearson correlation coefficients among the variables.

Research Question 2

While controlling for initial levels of coping, do levels of post-treatment coping differ between children with elevated levels of depression and children with normal levels of depression? Research has found that children with chronic pain who display elevated depressive symptoms tend to exhibit greater deficits across domains, including social and adaptive functioning, along with greater overall functional disability (Gauntlett-Gilbert & Eccleston, 2007), which indicates difficulties coping with pain in order to participate in daily activities. Therefore, it was hypothesized that while controlling for initial coping levels, children with normal levels of depression would display significantly higher levels of coping after treatment than children with elevated levels of depression would. Research Question 2 was examined through a one-way ANCOVA, which included a single categorical independent

variable with two levels (i.e., elevated depression; normal depression), a covariate (i.e., PSEQ score at admission), and a single dependent variable (i.e., PSEQ score at discharge).

Research Ouestion 3

While controlling for initial levels of coping and depression, do levels of post-treatment coping differ between children with elevated levels of anxiety and children with normal levels of anxiety? Previous research has found that, after controlling for the effects of depression, anxiety alone does not account for variability in social functioning, adaptive skills, or functional disability (Gauntlett-Gilbert & Eccleston, 2007). Therefore, it was hypothesized that while controlling for initial coping levels and levels of depression, there would not be a significant difference in levels of coping after treatment between children with normal levels of anxiety and children with elevated anxiety. Research Question 3 was examined through a one-way ANCOVA, which included a single categorical independent variable with two levels (i.e., elevated anxiety; normal anxiety), two covariates (i.e., CDI total T-Score; PSEQ score at admission), and a single dependent variable (i.e., PSEQ score at discharge).

Research Question 4

While controlling for initial levels of coping, do parent response behaviors predict levels of post-treatment coping? Previous research has suggested that a parent's tendency to distract, minimize, or protect in response to a child's chronic pain is associated with variability in the degree of functional disability displayed by the child. Specifically, high levels of protective behaviors have been associated with greater functional disability and somatic symptoms (e.g., Claar, Simons, & Logan, 2008; Connelly et al., 2010). Thus, it was hypothesized that after accounting for the effects of initial coping levels, parent response behaviors, including distraction, minimization, and protection, would predict post-treatment coping. It was originally

proposed that Research Question 4 would be examined through a one-way ANCOVA, which would include a single categorical independent variable with three levels (i.e., dominant distract, minimize, or protect), one covariate (i.e., PSEQ score at admission), and a single dependent variable (i.e., PSEQ score at discharge). However, as noted in Chapter 4, due to insufficient group membership, a hierarchical regression analysis was completed. Independent variables included ARCS-Minimize, ARCS-Distract, and ARCS-Protect scores as well as Admission PSEQ score as the covariate. The dependent variable was the Discharge PSEQ score.

Research Question 5

While controlling for initial levels of coping and depression, do parent response behaviors predict levels of post-treatment coping? Previous research has suggested that a significant interaction exists between parent response style and anxiety symptoms on functional disability. Specifically, an interaction between parent protective behavior and anxiety has been found to be associated with higher levels of disability. However, a significant interaction has not been identified between parent protective behavior and depressive symptoms (Claar, Simons, and Logan, 2008). Therefore, it was hypothesized that while controlling for initial coping levels and depression, parent response behaviors would continue to predict post-treatment coping levels. The proposed analysis for Research Question 5 was a one-way ANCOVA, with a single categorical independent variable with three levels (i.e., dominant distract, minimize, or protect), two covariates (i.e., CDI total T-Score; PSEQ score at admission), and a single dependent variable (i.e., PSEQ score at discharge). However, due to the issue noted in Research Question 4 with establishing a dominant parent response style, a hierarchical regression analysis was completed instead. Independent variables included ARCS-Minimize, ARCS-Distract, and

ARCS-Protect scores as well as Admission PSEQ and CDI Total Scores as the covariates. The dependent variable was the Discharge PSEQ score.

Power Analysis

The most robust analyses originally planned were for Research Question 5. Sample size for an ANCOVA with three independent variable levels and one covariate was determined using G*Power. The power analysis was conducted using an alpha of 0.05, a power of 0.80, and a medium effect size (f = 0.25) (Faul et al., 2013). The desired sample size given this information was 158 participants.

Summary

In this chapter, the reader was provided with an overview of the methodology and analyses for the present study. Five research questions and relevant analyses were described, focusing on the relationships among depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels, the impact of depression and anxiety on treatment outcomes, and the effect of parent response styles on treatment outcomes.

Chapter IV: RESULTS

Descriptive Statistics

The initial data set obtained included 165 cases. Participants with an age below 8 years or above 17 years at time of admission or those who were missing data for multiple key variables were excluded. Additionally, the initial dataset included data from second admissions for three participants; the data from the second admissions for these participants were also excluded. MASC data for cases with significant inconsistency index scores were also excluded from relevant analyses. After exclusionary criteria were applied, participants included 80 children and adolescents. Preliminary analyses indicated two cases that exhibited outlier values on PSEQ discharge score, CDI Total Score, and MASC Total Score; these cases were removed from analyses. Thus, 78 participants were included in analyses for Research Questions 1, 2, and 3. Of the 78 participants, 82.1% were female. Average age at admission was 14.66 (SD = 9.47). Due to missing parent data, a subset of participants was used for Research Questions 4 and 5, including 51 participants (80.4% female). Average age of participants included in analyses for Research Questions 4 and 5 was 14.79 (SD = 2.17).

Statistical Analyses

Research Question 1: What relationships exist among patient levels of depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels? Pearson correlation coefficients were computed to assess the relationships among depression (CDI Total T Score), anxiety (MASC Total T Score), parent protect response (ARCS Protect raw score), parent distract/monitor response (ARCS Distract/Monitor raw score), parent minimize response (ARCS Minimize raw score), admission coping levels (Admission PSEQ total score), and discharge coping levels (Discharge PSEQ total score). Significant correlations (p < .01) were

identified between admission and discharge coping (r = .42), admission coping and CDI (r = .46), admission coping and MASC (r = -.34), discharge coping and CDI (r = -.30), CDI and MASC (r = .53), and distract/monitor response and protect response (r = .44). Results of the correlation analysis are presented along with further descriptive statistics in Table 2. Notably, the PSEQ scores improved from time of admission to time of discharge. A paired samples t-test found that PSEQ scores at admission (M=30.27, SD=11.81) were significantly different than those at discharge (M=46.94, SD=9.47), t (77) = -12.62, p < .001.

Table 2

Correlations Among and Descriptive Statistics for Key Study Variables

| | N | M(SD) | Disc. PSEQ | CDI | MASC | Minimize | Distract | Protect |
|------------|----|---------------|------------|------|-------|----------|----------|---------|
| Adm. PSEQ | 78 | 30.27 (11.81) | .42** | 46** | 34** | .125 | 115 | 211 |
| Disc. PSEQ | 78 | 46.94 (9.47) | | 30** | 19 | .21 | 14 | 07 |
| CDI Total | 75 | 55.72 (13.66) | | | .53** | 08 | .13 | .18 |
| MASC Total | 65 | 58.28 (12.41) | | | | 10 | .08 | .112 |
| Minimize | 59 | 9.83 (3.21) | | | | | .22 | .01 |
| Distract | 60 | 23.13 (4.42) | | | | | | .44** |
| Protect | 55 | 16.58 (7.41) | | | | | | |
| | | | | | | | | |

^{**}p < .01.

Research Question 2: While controlling for initial levels of coping, do levels of posttreatment coping differ between children with elevated levels of depression and children with normal levels of depression? It was hypothesized that while controlling for initial coping levels, children with normal levels of depression would display significantly higher levels of

coping after treatment than children with elevated levels of depression would. Research Ouestion 2 was examined through a one-way ANCOVA. The independent variable, depression, included two levels: elevated (CDI Total T Score => 65) and normal (CDI Total T Score < 65). The dependent variable was coping at discharge (i.e., PSEQ score at discharge) and the covariate was coping at admission (i.e., PSEQ score at admission). Additionally, the following assumptions, necessary for conducting an ANCOVA, were examined: assumption of normality, assumption of homogeneity of variance, and the assumption of homogeneity of regression slopes (Mertler & Vannatta, 2005; Green & Salkind, 2008). The assumption of normality was confirmed by examining the skewness and kurtosis of continuous variables included in the analysis, which were the admission PSEQ score and discharge PSEQ score. Admission PSEQ was normally distributed, with skewness of .15 (SE = .27) and kurtosis of -.55 (SE = .54). Discharge PSEO was also normally distributed, with skewness of -.69 (SE = .27) and kurtosis of -.22 (SE = .54). Levene's Test of Equality of Error Variances confirmed homogeneity of variance, F(3, 59) = .91, p = .44. Finally, a univariate ANCOVA was used to assess homogeneity of regression slopes. The effect of the interaction between CDI Elevation and Admission PSEQ was not significant, F(1, 71) = 3.55, MSE = 255.75, p = .06, thus confirming the assumption of homogeneity of regression slopes.

Following preliminary analyses, a one-way ANCOVA was conducted. Results indicated that, after controlling for initial coping levels, depression elevation did not have a significant effect on discharge coping levels, F(1, 72) = 1.34, MSE = 99.60, p = .25. There was a significant effect of the covariate, initial coping levels, on discharge levels, F(1, 72) = 11.63, MSE = 867.09, p = .001.

Research Question 3: While controlling for initial levels of coping and depression, do levels of post-treatment coping differ between children with elevated levels of anxiety and children with normal levels of anxiety? It was hypothesized that while controlling for initial coping levels and levels of depression, there would not be a significant difference in levels of coping after treatment between children with normal levels of anxiety and children with elevated anxiety. Research Question 3 was examined through a one-way ANCOVA. The independent variable, anxiety, included two levels: elevated (MASC Total T Score => 65) and normal (MASC Total T Score < 65). The dependent variable was coping at discharge (i.e., PSEQ score at discharge) and the covariates were coping at admission (i.e., PSEQ score at admission) and depression (CDI Total T Score).

As noted in the analysis for Research Question 2, the assumption of normality was confirmed for Admission PSEQ and Discharge PSEQ scores. Admission PSEQ was normally distributed, with skewness of .15 (SE = .27) and kurtosis of -.55 (SE = .54). Discharge PSEQ was also normally distributed, with skewness of -.69 (SE = .27) and kurtosis of -.22 (SE = .54). The CDI Total T Score was also normally distributed, with skewness of .53 (SE = .28) and kurtosis of -.21 (SE = .55). Levene's Test of Equality of Error Variances confirmed homogeneity of variance, F(1, 61) = .14, p = .72. Finally, a univariate ANCOVA was used to assess homogeneity of regression slopes. The effect of the interaction between CDI Total T Score, MASC Elevation, and Admission PSEQ was not significant, F(2, 55) = .46, MSE = 29.95, p = .63, thus confirming the assumption of homogeneity of regression slopes.

Following preliminary analyses, a one-way ANCOVA was conducted. Results indicated that, after controlling for initial coping levels and depression levels, anxiety elevation did not have a significant effect on discharge coping levels, F(1, 59) = .03, MSE = 1.67, p = .87. There

was a significant effect of initial coping levels on discharge coping levels, F(1, 59) = 12.32, MSE = 766.56, p = .001, while there was not a significant effect of depression levels on discharge coping levels, F(1, 59) = 1.45, MSE = 90.27, p = .23.

Research Question 4: After controlling for initial levels of coping, do parent response behaviors significantly predict levels of post-treatment coping? Previous research has suggested that a parent's tendency to distract, minimize, or protect in response to a child's chronic pain is associated with variability in the degree of functional disability displayed by the child. Specifically, high levels of protective behaviors have been associated with greater functional disability and somatic symptoms (e.g., Claar, Simons, & Logan, 2008; Connelly et al., 2010). Thus, it was hypothesized that while controlling for initial coping levels, distraction, minimization, and protection parent response behaviors would significantly predict post-treatment coping levels.

It was proposed that Research Question 4 would be examined through a one-way ANCOVA, including a single categorical independent variable with three levels (i.e., dominant distract, minimize, or protect), one covariate (i.e., PSEQ score at admission), and a single dependent variable (i.e., PSEQ score at discharge). However, upon examination of the data, it was found that a "dominant" parent response style could not be identified, as the highest average score per scale was for the distract scale across participants. Given this, it was more appropriate to examine how well each type of parent response behavior predicted treatment outcomes rather than to categorize into separate parent response styles. Thus, a hierarchical regression analysis was completed. Independent variables included ARCS-Minimize, ARCS-Distract, and ARCS-Protect scores as well as Admission PSEQ score as the covariate. The dependent variable was the Discharge PSEQ score.

The data were examined for univariate outliers by calculating standardized values for each variable. All values were within three standard deviations of the mean (Mertler & Vannatta, 2005) and thus no outliers were identified. The data were also examined for multivariate outliers. The Mahalanobis distance across all 5 variables was less than the critical value at p=.001 with df=5 of 20.515, indicating no evidence of multivariate outliers (Mertler & Vannatta 2005). Standardized values for residuals were then assessed. Of the 51 cases, 4 were identified (i.e., 7.8% of the dataset) that had residual z-scores with absolute values greater than 1.96. Given that this exceeds the recommended 5% guideline provided by Field (2013), there is possible cause for concern. As will later be described, the regression analysis was thus run once with these 4 cases included, and then again with them excluded.

The assumption of normality was then assessed for the Admission PSEQ, Discharge PSEQ, ARCS-Minimize, ARCS-Distract, and ARCS-Protect variables. Admission PSEQ was normally distributed, with skewness of .26 (SE = .33) and kurtosis of .49 (SE = .66). Discharge PSEQ was also normally distributed, with skewness of .91 (SE = .33) and kurtosis of .26 (SE = .66). Values for ARCS-Distract (skewness = -.29; SE = .33; kurtosis = -.31; SE = .66), ARCS-Minimize (skewness = 1.02; SE = .33; kurtosis = 1.50; SE = .66), and ARCS-Protect (skewness = .36; SE = .33; kurtosis = -.75; SE = .66) were also within acceptable values (Mertler & Vannatta 2005). Scatterplots with standardized residuals plotted against standardized predicted values were examined to evaluate for problems with linearity or homoscedasticity. No systematic relationship between error values and predicted values was identified, and assumptions for homoscedasticity and linearity were thus met. Finally, multicollinearity was assessed through examination of VIF and tolerance statistics. VIF values were well under 10 and tolerance statistics were above .2; therefore, it was concluded that there was no collinearity present within

the data (Field, 2013). After evaluating the assumptions, preliminary analyses were run to examine the descriptive statistics of each variable as well as relationships among the variables; these analyses are summarized in Table 3. Significant correlations were identified between Admission PSEQ and Discharge PSEQ, r = .40, p < .01, between ARCS-Distract and ARCS-Protect scores, r = .42, p < .01, between Admission PSEQ and CDI score, r = -.39, p < .01, and Discharge PSEQ and CDI score, r = -.32, p < .05.

Table 3

Correlations Among and Descriptive Statistics for Key Variables in Research Questions 4 and 5

| | M(SD) | Disc. PSEQ | Minimize | Distract | Protect | CDI^\dagger |
|------------------------|---------------|------------|----------|----------|---------|------------------------|
| Adm. PSEQ | 31.35 (11.96) | .40** | .12 | 12 | 19 | 39** |
| Disc. PSEQ | 48.14 (9.35) | | .23 | 15 | 08 | 32* |
| Minimize | 9.88 (3.23) | | | .19 | .01 | 05 |
| Distract | 23.16 (4.24) | | | | .42** | .16 |
| Protect | 16.80 (7.53) | | | | | .15 |
| CDI^\dagger | 54.55 (13.75) | | | | | |

^{**}p < .01; *p < .05

A multiple regression analysis was then conducted to evaluate how well the parent responses predicted level of coping at discharge, after accounting for coping level at admission. Admission PSEQ was entered as a predictor in the first block of the analysis, and then ARCS-Minimize, ARCS-Distract, and ARCS-Protect were entered as predictors in the second block. This analysis was completed with the ARCS scores included together using the enter selection method, and once entering them separately using the stepwise method. Additionally, the

[†]Used in Research Question 5 only.

analyses were completed twice; once with the four cases with outlier standardized values for residuals included, and once with those four cases excluded. Similar results were obtained in terms of significance, and the results from the full data set are reported here.

Results indicated that coping at admission accounted for a significant amount of variability in coping at discharge, $R^2 = .16$, F(1, 49) = 9.10, p < .01. The data were then examined to determine whether ARCS scores predicted Discharge PSEQ scores over and above Admission PSEQ scores. The three ARCS scores when placed in the model together using the enter selection method did not account for a significant proportion of the discharge coping variance after controlling for the effects of admission coping, R^2 change= .06, F(4, 46) = 3.17, p = .34. Furthermore, when the ARCS scores were entered separately using a stepwise method, none of the scores were found to be significant predictors. These results suggest that parent response does not account for variability in patient coping at discharge after accounting for patient coping at admission.

Research Question 5: After controlling for initial levels of coping and depression, do parent response behaviors significantly predict levels of post-treatment coping? Previous research has suggested that a significant interaction exists between parent response style and anxiety symptoms on functional disability. Specifically, an interaction between parent protective behavior and anxiety has been found to be associated with higher levels of disability. However, a significant interaction has not been identified between parent protective behavior and depressive symptoms (Claar, Simons, and Logan, 2008). Therefore, it was hypothesized that while controlling for initial coping levels and depression, distraction, minimization, and protection behaviors would predict post-treatment coping. It was proposed that Research Question 5 would be examined through a one-way ANCOVA, including a single categorical

independent variable with three levels (i.e., dominant distract, minimize, or protect), two covariates (i.e., CDI total T Score; PSEQ score at admission), and a single dependent variable (i.e., PSEQ score at discharge). However, as noted previously, upon examination of the data, it was found that a "dominant" parent response style could not be identified, as the highest average score per scale was for the distract scale across participants. Given this, it was more appropriate to examine how well each type of parent response behavior predicted treatment outcomes rather than to categorize into separate parent response styles. Thus, a hierarchical regression analysis was completed. Independent variables included ARCS-Minimize, ARCS-Distract, and ARCS-Protect scores as well as Admission PSEQ and CDI Total Scores as the covariates. The dependent variable was the Discharge PSEQ score.

The data were examined for univariate outliers by calculating standardized values for each variable. All values were within three standard deviations of the mean (Mertler & Vannatta, 2005) and thus no outliers were identified. The data were also examined for multivariate outliers. The Mahalanobis distance across all 6 variables was less than the critical value at p=.001 with df=6 of 22.457, indicating no evidence of multivariate outliers (Mertler & Vannatta 2005). Standardized values for residuals were then assessed. Of the 51 cases, 1 was identified that had a residual z-score with absolute values greater than 1.96, which is acceptable (Field, 2013).

The assumption of normality was then assessed for the Admission PSEQ, Discharge PSEQ, ARCS-Minimize, ARCS-Distract, ARCS-Protect, and CDI Total variables. As noted previously, Admission PSEQ was normally distributed, with skewness of .26 (SE = .33) and kurtosis of -.49 (SE = .66). Discharge PSEQ was also normally distributed, with skewness of -.91 (SE = .33) and kurtosis of .26 (SE = .66). Values for ARCS-Distract (skewness = -.29; SE = .91) and kurtosis of .26 (SE = .66).

.33; kurtosis = -.31; SE = .66), ARCS-Minimize (skewness = 1.02; SE = .33; kurtosis = 1.50; SE= .66), and ARCS-Protect (skewness = .36; SE = .33; kurtosis = -.75; SE = .66) were also within acceptable values. Finally, CDI Total values were normally distributed, with skewness of .54 (SE = .33) and kurtosis of -.28 (SE = .66) (Mertler & Vannatta 2005). Scatterplots with standardized residuals plotted against standardized predicted values were examined to evaluate for problems with linearity or homoscedasticity. No systematic relationship between error values and predicted values was identified, and assumptions for homoscedasticity and linearity were thus met. Finally, multicollinearity was assessed through examination of VIF and tolerance statistics. VIF values were well under 10 and tolerance statistics were above .2; therefore, it was concluded that there was no collinearity present within the data (Field, 2013). After evaluating the assumptions, preliminary analyses were run to examine the descriptive statistics of each variable as well as relationships among the variables; these analyses are summarized previously in Table 3. As noted previously, significant correlations were identified between Admission PSEQ and Discharge PSEQ, r = .40, p < .01, between ARCS-Distract and ARCS-Protect scores, r= .42, p < .01, between Admission PSEQ and CDI score, r = -.39, p < .01, and Discharge PSEQ and CDI score, r = -.32, p < .05.

A multiple regression analysis was then conducted to evaluate how well the parent responses predicted level of coping at discharge, after accounting for level of depression and coping level at admission. Admission PSEQ and CDI Total were entered as predictors in the first block of the analysis, and then ARCS-Minimize, ARCS-Distract, and ARCS-Protect were entered as predictors in the second block. As with Research Question 4, the ARCS scores were entered together for one analysis, and also entered separately in a stepwise analysis. Results of the initial analysis in which the ARCS scores were entered together indicated that coping at

admission and level of depression accounted for a significant amount of variability in coping at discharge, $R^2 = .19$, F(2, 48) = 5.53, p < .01. The data were then examined to determine whether ARCS scores predicted Discharge PSEQ scores over and above Admission PSEQ and CDI scores. The three ARCS scores did not account for a significant proportion of the discharge coping variance after controlling for the effects of admission coping, R^2 change= .05, F(5, 45) = 2.87, p = .37. Moreover, the stepwise analysis indicated that the ARCS scores were not separately significant predictors. These results suggest that parent response does not account for variability in patient coping at discharge after accounting for patient coping at admission and level of depression.

Chapter V: DISCUSSION

Idiopathic chronic pain is a common problem among children that is costly both in terms of functional disability that can manifest in affected children and the financial consequences for families. Multiple biological, psychological, and social factors have been found to be associated with pediatric idiopathic chronic pain. Of relevance to the present study, research has suggested that children with idiopathic chronic pain tend to display elevated levels of depression and anxiety as compared to typical children (e.g., Wasan, Sullivan, & Clark, 2010; Eccleston et al., 2004; Campo et al., 2002). Moreover, specific parent responses to pain have significant relationships with a variety of consequences, particularly related to functional disability. Among parent response behaviors, protective responding has been identified as an area of particular concern, as protective responses tend to be related to increased functional impairments and somatic symptoms ((Kaczynski et al., 2009); Claar, Simons, & Logan, 2008).

Existing research has focused primarily on the relationships among chronic pain and various biological, psychological, and social factors. However, few studies have examined the potential impact of these factors on an individual's benefit from treatment. Anecdotal observations reveal that while many patients benefit from intensive multidisciplinary inpatient treatment program, some report minimal improvement in their own ability to manage symptoms. The purpose of this study was to provide a better understanding of the variables that impact a patient's potential to benefit from treatment and thus lead to more informed decisions regarding treatment, including referrals and admissions to inpatient rehabilitation programs. Additionally, the present research aimed to offer insight to aspects of treatment that may need to be given greater emphasis for certain patients.

Research Question 1

The first research question examined the extent to which patient levels of depression, anxiety, admission coping levels, discharge coping levels, and parent pain response behaviors are related. It was hypothesized that *significant correlations would exist among levels of depression, anxiety, parent pain response style, admission coping levels, and discharge coping levels.*Results indicate partial support for the hypothesis that significant relationships would exist among study variables.

Admission coping and depression were inversely related, as were discharge coping and depression. That is, participants who reported higher levels of depressive symptoms possess lower levels of coping at admission and at discharge, and those with lower levels of depression had greater capacity for coping at admission and at discharge. This finding is consistent with the relationship identified between depression symptoms and pain self-efficacy (i.e., confidence in coping) among two samples of adults with chronic pain in Brazil and Australia (Sardá, Nicholas, Asghari, & Pimenta, 2009). Similarly, in a meta-analysis of studies focused on adults with chronic pain, Jackson, Wang, Wang, and Fan (2014) found pain self-efficacy to be consistently negatively correlated with affective distress. Given that previous research regarding pain self-efficacy and symptoms of depression was conducted with adults, the present study expands on existing findings as a similar relationship was also found in children and adolescents.

An inverse relationship was also identified between admission coping and anxiety.

Despite the relationship between anxiety and admission coping, there was not a significant relationship between anxiety and coping at time of discharge. Kalapurakkel, Carpino, Lebel, and Simons (2015) found that among children with chronic headache, pain self-efficacy had a significant inverse relationship with depressive symptoms and anxiety symptoms. However,

pain intensity and depression were significantly related while pain intensity and anxiety were not. It is possible that anxiety was not significantly related to pain self-efficacy at the time of discharge because the aspects of pain self-efficacy that related to anxiety at admission were more effectively treated, thus reducing the relationship between anxiety and coping, while aspects of pain self-efficacy that were related to depression were not as effectively treated. However, further research is necessary to better understand this finding.

Relationships among parent response behaviors were also analyzed, including protective, minimizing, and distracting/monitoring behaviors. *Protective* responses include parent behaviors such as paying more attention to the child than usual, completing tasks for the child, taking the child to the doctor or administering medicine, and providing the child with special privileges. Minimizing responses include parent behaviors such as telling the child that nothing can be done, avoiding paying attention to the child, and telling the child to not worry so much about their pain. Conversely, distracting and monitoring responses include parent behaviors such as reassuring the child that he or she will be okay, encouraging the child to engage in activities, and checking on how the child feels (Van Slyke & Walker, 2006). In the initial factor analysis completed by Van Slyke and Walker (2006) to assess the psychometric properties of the ARCS, it was found that there was a significant positive correlation between the Protect scale and the Distract/Monitor scale. A positive relationship of similar magnitude between distracting/ monitoring and protecting behaviors was identified in the present research. However, Van Slyke and Walker found that the Minimize scale was negatively correlated with the Protect scale, although these scales were not found to be significantly related in the present research. Moreover, counter to the hypothesized results, other than the relationship between distract and protecting parent responses, parent response behaviors were not otherwise significantly related to any other variable, including admission or discharge coping, depression, or anxiety. A possible factor that may have contributed to this pattern of results is that the measures of coping, depression, and anxiety were self-reported, while parent response behaviors were parent-reported.

Coping at admission, as expected, was strongly and positively related to coping at discharge. Improvements in coping secondary to the treatment received were anticipated and evidenced in results. However, participants who had lower coping levels at admission had lower coping levels at discharge relative to the overall sample, and those with higher admission coping levels also had higher discharge coping levels. Depression and anxiety scores on the CDI and MASC, respectively, were also positively correlated in the present study. Previous research on the divergent validity of the MASC did not indicate a significant relationship between the MASC and CDI total scores in the context of school-based population studies (March, Parker, Sullivan, Stallings, & Conners, 1997). However, among children with chronic headaches, a relationship of similar magnitude has been identified (Kalapurakkel et al., 2015).

Research Question 2

The second research question examined whether levels of coping after treatment differed between participants with elevated levels of depression and those with normal levels of depression, after controlling for initial coping levels. It was hypothesized that while controlling for initial coping levels, children with normal levels of depression would display significantly higher levels of coping after treatment than children with elevated levels of depression. Results indicated insufficient evidence to support the hypothesis.

After controlling for initial coping, patients with elevated levels of depression and patients with normal levels of depression did not differ in coping levels at discharge. This

finding is somewhat in contrast to existing research. Previous studies have generally suggested a positive relationship between elevated depressive symptoms and greater functional disability (e.g., Gauntlett-Gilbert & Eccleston, 2007), which indicates difficulties coping with pain sufficiently to participate in daily activities. However, in the present research, after accounting for a child's initial level of coping, patient level of coping after treatment was not significantly impacted by depressive symptoms. This result may in part be attributed to the strong effect of initial coping on levels of discharge coping. Further, while there is an association between coping and depression (as noted in the findings from Research Question 1), this relationship does not change as the result of the inpatient treatment. However, one other study produced a somewhat similar outcome. In research with adults with chronic low back pain, Koenig, Kupper, Skidmore, and Murphy (2014) found that psychological functioning was not a significant predictor of pain severity or pain self-efficacy (i.e., confidence in coping with pain). Conversely, poorer physical health and poorer social functioning did predict greater pain severity. Social functioning in that study also predicted pain self-efficacy, while physical functioning did not predict pain self-efficacy. Thus, it appears that the construct of confidence in coping with pain (i.e., pain self-efficacy) may be separate from the construct of functional disability, particularly in terms of the relationship of each with psychological factors.

Research Question 3

The third research question examined whether levels of coping after treatment differed between participants with elevated levels of anxiety and those with normal levels of anxiety, after controlling for initial coping levels and anxiety. Given previous research indicating that anxiety does not account for variation in functional disability and somatic symptoms beyond the effects of depression, it was hypothesized that *while controlling for initial coping levels and*

levels of depression, there would not be a significant difference in levels of coping after treatment between children with normal levels of anxiety and children with elevated anxiety. Results did not provide evidence to support this hypothesis.

After controlling for initial coping and depression, coping at discharge did not significantly differ between participants with high anxiety and those with normal anxiety. There was a significant effect of initial coping on discharge coping, but no significant effect of depression or anxiety on discharge coping. Previous research has indicated that anxiety does not account for variability in social functioning, adaptive skills, or functional disability once depressive symptoms are accounted for (Gauntlett-Gilbert & Eccleston, 2007), so it was anticipated that after controlling for depression, anxiety would not influence discharge coping. Notably, the lack of group differences is generally consistent with the hypothesized result, but the finding that depression did not significantly affect discharge coping is not consistent. As discussed in relationship to Research Question 2, it appears that the construct of pain self-efficacy may be separate from the construct of functional disability, particularly in terms of the relationship of each with psychological factors, resulting in this inconsistency. In general, results of analyses for Research Question 3 indicate that initial coping levels offer the most information regarding discharge coping levels.

Research Question 4

The fourth research question examined whether levels of coping after treatment differed based on parent response behaviors, after controlling for initial levels of coping. It was hypothesized that after accounting for the effects of initial coping levels, parent response behaviors, including distraction, minimization, and protection, would predict post-treatment coping. Results indicated insufficient evidence to support the hypothesis.

After controlling for the effects of initial coping, parent response behaviors did not significantly predict post-treatment coping. This finding is somewhat incongruent with previous literature (e.g., Claar, Simons, & Logan, 2008; Connelly et al., 2010), which has indicated that high levels of protective behaviors have been associated with greater functional disability and somatic symptoms.

It is possible that the lack of significant findings regarding the effect of parent response is due to the small sample size of the present study. The available parent data was far more limited than anticipated, and possible is that the small sample resulted in insufficient statistical power to produce significant findings. In addition to the potential role of sample size, there is an important distinction between the present research and previous findings that may have also impacted the results. The present study focused on the patient's confidence in coping with pain rather than assessing functional disability or somatic symptoms. Moreover, the present study focused on parent responses and treatment outcomes among the chronic pain population rather than examining how outcomes and parent responses may differ within this population as compared to the general population. Therefore, the contrast in the variables that were evaluated and the context within which they were assessed (i.e., within an inpatient treatment protocol) as compared to previous studies may also account for some the inconsistent results. Further research with more adequate sample size focusing on these dynamics is warranted.

Research Question 5

The fifth research question examined whether levels of coping after treatment differed based on parent response behaviors, after controlling for initial levels of coping as well as level of depression. It was hypothesized that while controlling for initial coping levels and

depression, parent response behaviors would continue to predict post-treatment coping levels.

Results indicated insufficient evidence to support the hypothesis.

As found in the results for Research Question 4, coping at admission predicted coping at discharge, and neither depression nor parent response behaviors accounted for any further variance past that. As with the finding for Research Question 4, this finding is inconsistent with previous literature (e.g., Claar, Simons, & Logan, 2008; Connelly et al., 2010), which has indicated a relationship between parent protective behaviors and level of functional disability and somatic symptoms. Possible factors affecting this result may include the small sample size, as well as the distinction between the construct of functional disability and that of confidence in coping with pain. The inconsistency of the present results with previous findings suggests that clarification regarding the relationship between pain self-efficacy and level of functional impairment among children and adolescents is needed.

Biopsychosocial Impacts

The biopsychosocial model can be used to understand the outcomes of the present research more generally, particularly as they relate to characteristics of the sample as well as the patterns of outcome data obtained. From a biological perspective, the age of the participants may play an important role in manifestations of symptoms as well as parent responding. However, the average age of participants in the present sample was approximately 14 years, which is commensurate with that of the primary research studies used to inform the hypotheses and research questions. For example, the sample used by Gauntlett-Gilbert & Eccleston (2007) had a mean age of 14.6 years, and results of their work indicated that children with greater depressive symptoms displayed greater functional disability. A similar relationship between elevated depressive symptoms and post-treatment coping was not identified in the current research.

Furthermore, in the work of Claar and fellow researchers (2008), relationships between emotional distress, parent response behaviors, and functional disability were identified within a similarly aged sample (*M*=13.94 years). A combination of biological/developmental and psychological factors may play an important role in symptom manifestation around that age. Specifically, the psychopathology examined in the present research, including anxiety and depression, often emerges in early adolescence, which may influence the pattern of symptoms along with related coping mechanisms being employed by the children and adolescents in the sample. Furthermore, developmental and social factors also often intersect at this age, particularly related to parenting behaviors.

The timing of data collection in the present study may be one of the most salient factors contributing to conflicting results, particularly related to psychological and social dynamics. Data regarding parent response behaviors as well as anxiety and depression symptoms were collected upon admission to an inpatient treatment program. Given the criteria for admission to such a program, it is likely that patients and parents were in a significantly elevated state of stress, and that the patients' idiopathic pain symptoms were at a peak at that time. Therefore, it is possible that data collected regarding parent behaviors and patient psychopathology may pertain to a period that, for many families, is a time of crisis, and may not be generalizable to their typical behaviors and psychological symptoms. Some previous research has included data collected at time of evaluation for inpatient/residential treatment (e.g., (Gauntlett-Gilbert & Eccleston, 2007), while other studies have been focused on tertiary pain clinics (e.g., Claar, Simons, & Logan, 2008) and outpatient clinics (Connelly et al., 2010). These varying levels of care and circumstances may also contribute to the differences in the present research as compared to existing work.

Implications

Several implications for practice are evident in the contrast of present results with prior research. Of particular interest is that levels of anxiety, depression, or specific parent response behaviors did not affect levels of coping after treatment beyond the influence of coping prior to treatment. Thus, future coping is best predicted by past coping. Notably, there was a significant relationship between depressive symptoms and coping at admission and discharge, and between anxiety and coping at admission, suggesting that these factors are related However, if the question is *How well will patients in inpatient treatment cope at the time of discharge?* their level of coping at admission offers more information than their symptoms of depression or anxiety do.

In the present research, patients' confidence in their ability to cope with pain was assumed to be closely related to functional disability (i.e., functional consequences of pain such as limited independence in self-care, decreased participation in activities or school) given past research suggesting such a relationship (Costal, Maherl, McAuleyl, Hancockl, & Smeetsl, 2011; Jackson, Wang, Wang, & Fan, 2014; Turner, Ersek, & Kemp, 2005). Therefore, existing research on functional disability in the chronic pain population was extrapolated to inform the research questions and hypotheses related to coping. However, it appears that, at least for the population examined, pain self-efficacy/coping and functional disability may be separate constructs. Notably, in contrast to the present research, much of the existing literature regarding pain self-efficacy and functional disability focuses on adults, and it may also be possible that the relationship between these constructs among children and adolescents is different than the relationship found for adults. If such a distinction was evidenced, possible contributing factors should be explored. A specific topic area that may merit further investigation is whether

assessments of pain self-efficacy result in a greater response bias among children (e.g., indicating greater confidence in coping due to the phrasing or presentation of the questions) than measures of functional disability do.

Limitations

Inherent in the use of an existing data set were multiple factors that limited the present research. The sample size was smaller than anticipated, and the amount of missing data, particularly for parent responses, was significantly greater than expected. Moreover, the use of the PSEQ as the outcome measure of benefit from treatment is another factor that offers somewhat limited information about other important treatment outcomes, such as the participants' functional disability. Additional treatment outcomes beyond pain self-efficacy would have enabled a more comprehensive understanding of patients' benefit from treatment, and ways in which the psychological and parent variables may have related to different outcomes. This is particularly salient given the differences in the results of the present research and those of previous studies that focused on functional disability.

Several threats to internal validity were present in the current research. The data were collected over a period of 5 ½ years, and given the retroactive review of the data, it was not possible to monitor treatment integrity across that time. In particular, the integrity of the data collection procedures could not be assessed, nor could the treatment integrity of the inpatient rehabilitation program. Moreover, history effects may have been present, not necessarily in terms of effects on individual participant scores, but rather on the overall sample. Furthermore, a variety of other factors that could not be adequately controlled for were threats both to internal and external validity. Most notable is changes in healthcare over that 5 ½ year data collection period. Variability in healthcare availability and coverage may have influenced which patients

were admitted to the inpatient program and therefore included in the present study, and which patients were excluded secondary to insurance coverage or other factors. There are additional threats to external validity in the current study, and generalizability of results is limited. The population and setting studied is highly specialized, and the results cannot validly be applied beyond that population and treatment setting. Moreover, there is a potential bias related to which participants and parents completed and returned the packets from which the data were gathered.

Recommendations for Future Research

Results of the present study raise several areas of inquiry in need of further examination. Though a sizable body of research exists regarding the prevalence of various biological, psychological, and social factors among children and adolescents with chronic pain, little is known regarding the effects of these variables on treatment outcomes (Eccleston, 2003). Further work should be done to establish which factors may be most effective in predicting patients' benefit from treatment. Moreover, the topic of treatment outcomes should be further examined, and in particular, the relationship between pain self-efficacy and functional disability in children. Such an evaluation should include self-report measures of pain self-efficacy and functional disability alongside informant measures (e.g., completed by parents and/or clinicians) to consider the relationship among input from multiple reporters. An evaluation of treatment outcomes that includes measures of coping along with measures of functional disability could help to clarify some of the inconsistent results obtained in the present study. Additional research regarding parent response behaviors and the relationship that these behaviors may have with pain selfefficacy as well as other treatment outcomes would also assist in clarifying some of the questions raised in the present research. It would also be helpful to examine parent response behaviors prior to receiving psychoeducation (i.e., such as that which would typically be provided to

parents during an inpatient admission) and after receiving psychoeducation, to examine the effects of psychoeducation on changing parent response behaviors.

Further evaluation of treatment outcomes across multiple levels of care would also be prudent. For example, exploring differences in outcomes between inpatient programs and intensive outpatient programs could provide valuable information about whether inpatient admissions are warranted, or if similar gains could be made with an intensive outpatient program or day program that does not require the child to stay at a hospital. An improved understanding of outcomes according to various combinations of therapies (e.g., psychotherapy, biofeedback, occupational therapy, physical therapy, and medication management) could also lead to improved treatment decisions. Establishing whether treatment outcomes differ according to antecedents of the pain (e.g., injury, stress, illness, etc.) and the location of the pain (e.g., diffuse, back, headaches, etc.) could also be helpful. Moreover, future research could also incorporate longitudinal studies to better understand characteristics of patients who require more treatment and/or repeat inpatient admissions.

Conclusion

Although the results of the present study were limited in terms of their statistical significance, they offer a foundation and direction for future research. While relationships between variables such as coping and depression were confirmed, the relationships that were not significant provide perhaps even more information about what is yet to be determined regarding the characteristics of children and adolescents with idiopathic chronic pain and their treatment. Through an improved understanding of these characteristics and their effect on treatment, there is immense potential for improved treatment outcomes for those who suffer from chronic pain.

References

- Aasland, A., Flatö, B., & Vandvik, I. H. (1997). Psychosocial factors in children with idiopathic musculoskeletal pain: a prospective, longitudinal study. *Acta Paediatrica* (Oslo, Norway: 1992), 86(7), 740–746.
- Barrett, P. M., Duffy, A. L., Dadds, M. R., & Rapee, R. M. (2001). Cognitive-behavioral treatment of anxiety disorders in children: long-term (6-year) follow-up. *Journal of Consulting and Clinical Psychology*, 69(1), 135–141.
- Campo, J. V., Comer, D. M., Jansen-McWilliams, L., Gardner, W., & Kelleher, K. J. (2002).

 Recurrent pain, emotional distress, and health service use in childhood. *The Journal of Pediatrics*, *141*(1), 76–83. https://doi.org/10.1067/mpd.2002.125491
- Carter, B. D., & Threlkeld, B. M. (2012). Psychosocial perspectives in the treatment of pediatric chronic pain. *Pediatric Rheumatology*, 10(1), 15. https://doi.org/10.1186/1546-0096-10-15
- Claar, R. L., Guite, J. W., Kaczynski, K. J., & Logan, D. E. (2010). Factor Structure of the Adult Responses to Children's Symptoms: Validation in Children and Adolescents With Diverse Chronic Pain Conditions: *The Clinical Journal of Pain*, 26(5), 410–417. https://doi.org/10.1097/AJP.0b013e3181cf5706
- Claar, R. L., Simons, L. E., & Logan, D. E. (2008). Parental response to children's pain: The moderating impact of children's emotional distress on symptoms and disability. *Pain*, 138(1), 172–179. https://doi.org/10.1016/j.pain.2007.12.005

- Connelly, M., Anthony, K. K., Sarniak, R., Bromberg, M. H., Gil, K. M., & Schanberg, L. E. (2010). Parent pain responses as predictors of daily activities and mood in children with juvenile idiopathic arthritis: the utility of electronic diaries. *Journal of Pain and Symptom Management*, 39(3), 579–590. https://doi.org/10.1016/j.jpainsymman.2009.07.013
- Costal, L. da C. M., Maherl, C. G., McAuleyl, J. H., Hancockl, M. J., & Smeetsl, R. J. E. M. (2011). Self-efficacy is more important than fear of movement in mediating the relationship between pain and disability in chronic low back pain. *European Journal of Pain*, 15(2), 213–219. https://doi.org/10.1016/j.ejpain.2010.06.014
- Eccleston, C. (2003). Managing chronic pain in children and adolescents. *BMJ*, 326(7404), 1408–1409. https://doi.org/10.1136/bmj.326.7404.1408
- Eccleston, Christopher, Crombez, G., Scotford, A., Clinch, J., & Connell, H. (2004).

 Adolescent chronic pain: patterns and predictors of emotional distress in adolescents with chronic pain and their parents. *Pain*, *108*(3), 221–229.

 https://doi.org/10.1016/j.pain.2003.11.008
- Fishbain, D. A., Cutler, R., Rosomoff, H. L., & Rosomoff, R. S. (1997). Chronic pain-associated depression: antecedent or consequence of chronic pain? A review. *The Clinical Journal of Pain*, *13*(2), 116–137.
- Gatchel, R. J., Peng, Y. B., Peters, M. L., Fuchs, P. N., & Turk, D. C. (2007). The biopsychosocial approach to chronic pain: Scientific advances and future directions.
 Psychological Bulletin, 133(4), 581–624. https://doi.org/10.1037/0033-2909.133.4.581

- Gauntlett-Gilbert, J., & Eccleston, C. (2007). Disability in adolescents with chronic pain:

 Patterns and predictors across different domains of functioning. *Pain*, *131*(1–2), 132–141. https://doi.org/10.1016/j.pain.2006.12.021
- Ho, I. K., Goldschneider, K. R., Kashikar-Zuck, S., Kotagal, U., Tessman, C., & Jones, B. (2008). Healthcare Utilization and Indirect Burden among Families of Pediatric Patients with Chronic Pain. *Journal of Musculoskeletal Pain*, 16(3), 155–164. https://doi.org/10.1080/10582450802161853
- Hoftun, G. B., Romundstad, P. R., Zwart, J.-A., & Rygg, M. (2011). Chronic idiopathic pain in adolescence high prevalence and disability: The young HUNT study 2008: *Pain*, *152*(10), 2259–2266. https://doi.org/10.1016/j.pain.2011.05.007
- Jackson, T., Wang, Y., & Fan, H. (2014). Self-Efficacy and Chronic Pain Outcomes:

 A Meta-Analytic Review. *The Journal of Pain*, 15(8), 800–814.

 https://doi.org/10.1016/j.jpain.2014.05.002
- Johan, D., & Loeser, M. (2012, May 22). International Association for the Study of Pain.

 Retrieved from www.iasp-pain.org/Taxonomy
- Kaczynski, K. J., Claar, R. L., & Logan, D. E. (2009). Testing Gender as a Moderator of Associations Between Psychosocial Variables and Functional Disability in Children and Adolescents with Chronic Pain. *Journal of Pediatric Psychology*, 34(7), 738–748. https://doi.org/10.1093/jpepsy/jsn113
- Kalapurakkel, S., A. Carpino, E., Lebel, A., & E. Simons, L. (2015). "Pain Can't Stop Me": Examining Pain Self-Efficacy and Acceptance as Resilience Processes Among Youth

- With Chronic Headache. *Journal of Pediatric Psychology*, 40(9), 926–933. https://doi.org/10.1093/jpepsy/jsu091
- King, S., Chambers, C. T., Huguet, A., MacNevin, R. C., McGrath, P. J., Parker, L., & MacDonald, A. J. (2011). The epidemiology of chronic pain in children and adolescents revisited: A systematic review: *Pain*, 152(12), 2729–2738.
 https://doi.org/10.1016/j.pain.2011.07.016
- Knook, L. M. E., Konijnenberg, A. Y., van der Hoeven, J., Kimpen, J. L. L., Buitelaar, J. K., van Engeland, H., & de Graeff-Meeder, E. R. (2011). Psychiatric disorders in children and adolescents presenting with unexplained chronic pain: what is the prevalence and clinical relevancy? *European Child & Adolescent Psychiatry*, 20(1), 39–48. https://doi.org/10.1007/s00787-010-0146-0
- Konijnenberg, A. Y., Uiterwaal, C. S. P. M., Kimpen, J. L. L., van der Hoeven, J., Buitelaar, J. K., & de Graeff-Meeder, E. R. (2005). Children with unexplained chronic pain: substantial impairment in everyday life. *Archives of Disease in Childhood*, 90(7), 680–686. https://doi.org/10.1136/adc.2004.056820
- Kovacs, M. (1992). *Children's depression inventory*. North Tonawanda, NY: Multi-Health System.
- Kozlowska, K., Rose, D., Khan, R., Kram, S., Lane, L., & Collins, J. (2008). A Conceptual
 Model and Practice Framework for Managing Chronic Pain in Children and Adolescents:
 Harvard Review of Psychiatry, 16(2), 136–150.
 https://doi.org/10.1080/10673220802069723

- Landry, B. W., Fischer, P. R., Driscoll, S. W., Koch, K. M., Harbeck-Weber, C., Mack, K. J., ... Brandenburg, J. E. (2015). Managing Chronic Pain in Children and Adolescents: A Clinical Review. *PM&R*, 7(11), S295–S315. https://doi.org/10.1016/j.pmrj.2015.09.006
- Lester, N., Lefebvre, J. C., & Keefe, F. J. (1994). Pain in young adults: I. Relationship to gender and family pain history. *The Clinical Journal of Pain*, 10(4), 282–289.
- Logan, D. E., Simons, L. E., Stein, M. J., & Chastain, L. (2008). School Impairment in Adolescents With Chronic Pain. *The Journal of Pain*, 9(5), 407–416. https://doi.org/10.1016/j.jpain.2007.12.003
- March, J. S. (2012). *Multidimensional Anxiety Scale for Children 2nd Edition (MASC 2)*.

 MultiHealth Systems; Toronto, CA.
- March, John S., Parker, J. D. A., Sullivan, K., Stallings, P., & Conners, C. K. (1997a). The Multidimensional Anxiety Scale for Children (MASC): Factor Structure, Reliability, and Validity. *Journal of the American Academy of Child & Adolescent Psychiatry*, 36(4), 554–565. https://doi.org/10.1097/00004583-199704000-00019
- March, John S., Parker, J. D. A., Sullivan, K., Stallings, P., & Conners, C. K. (1997b). The Multidimensional Anxiety Scale for Children (MASC): Factor Structure, Reliability, and Validity. *Journal of the American Academy of Child & Adolescent Psychiatry*, 36(4), 554–565. https://doi.org/10.1097/00004583-199704000-00019
- Merlijn, V. P. B. M., Hunfeld, J. A. M., van der Wouden, J. C., Hazebroek-Kampschreur, A. A. J. M., Koes, B. W., & Passchier, J. (2003). Psychosocial factors associated with chronic pain in adolescents. *Pain*, *101*(1–2), 33–43.

- Miró, J., Huguet, A., & Jensen, M. P. (2014). Pain Beliefs Predict Pain Intensity and Pain Status in Children: Usefulness of the Pediatric Version of the Survey of Pain Attitudes.

 Pain Medicine, 15(6), 887–897. https://doi.org/10.1111/pme.12316
- Nicholas, M. K. (1989). *Self-Efficacy and Chronic Pain*. Presented at the Annual Conference of the British Psychological Society, St. Andrews.
- Palermo, T. M., Eccleston, C., Lewandowski, A. S., de C. Williams, A. C., & Morley, S. (2010). Randomized controlled trials of psychological therapies for management of chronic pain in children and adolescents: An updated meta-analytic review: *Pain*, 148(3), 387–397. https://doi.org/10.1016/j.pain.2009.10.004
- Puzino, K., & Mindell, J. A. (2015). Sleep disturbances in pediatric chronic pain patients: The role of cognitions. *Translational Issues in Psychological Science*, *1*(1), 6–15. https://doi.org/10.1037/tps0000013
- Reid, G. J., Gilbert, C. A., & McGrath, P. J. (1998). The Pain Coping Questionnaire: preliminary validation. *Pain*, 76(1–2), 83–96.
- Reinecke, M. A., Ryan, N. E., & DuBOIS, D. L. (1998). Cognitive-Behavioral Therapy of Depression and Depressive Symptoms During Adolescence: A Review and Meta-Analysis. *Journal of the American Academy of Child & Adolescent Psychiatry*, *37*(1), 26–34. https://doi.org/10.1097/00004583-199801000-00013
- Sardá, J., Nicholas, M. K., Asghari, A., & Pimenta, C. A. M. (2009). The contribution of self-efficacy and depression to disability and work status in chronic pain patients: A comparison between Australian and Brazilian samples. *European Journal of Pain*, 13(2), 189–195. https://doi.org/10.1016/j.ejpain.2008.03.008

- Schechter, N. L., Palermo, T. M., Walco, G. A., & Berde, C. B. (2010). Persistent Pain in Children. In S. Fishman, J. Ballantyne, J. P. Rathmell, & J. J. Bonica (Eds.), *Bonica's management of pain* (4th ed, pp. 767–782). Baltimore, MD: Lippincott, Williams & Wilkins.
- Sherry, D. D. (2011). Pain Amplification Syndromes. In J. T. Cassidy, R. E. Petty, R. M. Laxer, & C. B. Lindsley (Eds.), *Textbook of pediatric rheumatology* (6th ed, pp. 718–729). Philadelphia: Saunders.
- Sherry, David D., & Malleson, P. N. (2002). The idiopathic musculoskeletal pain syndromes in childhood. *Rheumatic Diseases Clinics of North America*, 28(3), 669–685.
- Sleed, M., Eccleston, C., Beecham, J., Knapp, M., & Jordan, A. (2005). The economic impact of chronic pain in adolescence: Methodological considerations and a preliminary costs-of-illness study: *Pain*, *119*(1–3), 183–190. https://doi.org/10.1016/j.pain.2005.09.028
- Turk, D. C., & Okifuji, A. (2010). Pain Terms and Taxonomies of Pain. In S. Fishman, J. Ballantyne, J. P. Rathmell, & J. J. Bonica (Eds.), *Bonica's management of pain* (4th ed, pp. 13–23). Baltimore, MD: Lippincott, Williams & Wilkins.
- Turner, J., Ersek, M., & Kemp, C. (2005). Self-Efficacy for Managing Pain Is Associated With Disability, Depression, and Pain Coping Among Retirement Community Residents With Chronic Pain. *The Journal of Pain*, *6*(7), 471–479. https://doi.org/10.1016/j.jpain.2005.02.011
- Van Slyke, D. A., & Walker, L. S. (2006). Mothers' Responses to Children's Pain. *The Clinical Journal of Pain*, 22(4), 387–391. https://doi.org/10.1097/01.ajp.0000205257.80044.01

- Vervoort, T. (2005). Catastrophic Thinking About Pain is Independently Associated with Pain Severity, Disability, and Somatic Complaints in School Children and Children with Chronic Pain. *Journal of Pediatric Psychology*, *31*(7), 674–683. https://doi.org/10.1093/jpepsy/jsj059
- Walker, L. S., & Greene, J. W. (1991). The functional disability inventory: measuring a neglected dimension of child health status. *Journal of Pediatric Psychology*, 16(1), 39–58.
- Wasan, A. D., Sullivan, M. D., & Clark, M. R. (2010). Psychiatric Illness, Depression,
 Anxiety, and Somatoform Pain Disorders. In S. Fishman, J. Ballantyne, J. P. Rathmell,
 & J. J. Bonica (Eds.), *Bonica's management of pain* (4th ed, pp. 393–417). Baltimore,
 MD: Lippincott, Williams & Wilkins.
- Yang, L., Zhou, X., Zhou, C., Zhang, Y., Pu, J., Liu, L., ... Xie, P. (2017). Efficacy and Acceptability of Cognitive Behavioral Therapy for Depression in Children: A Systematic Review and Meta-analysis. *Academic Pediatrics*, 17(1), 9–16.
 https://doi.org/10.1016/j.acap.2016.08.002