THE RELATIONSHIP OF CAREGIVER DEMAND AND PARENT DISTRESS IN JUVENILE RHEUMATIC DISEASE: THE MEDIATING EFFECT OF PARENT ATTITUDE TOWARD ILLNESS

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CHAPTER I

INTRODUCTION

Juvenile Rheumatic Diseases (JRDs) are a series of pediatric autoimmune disorders characterized by joint pain and swelling, connective tissue inflammation, chronic pain or tenderness in the joint(s), and possible limited movement in the affected joint(s) (Cassidy, Petty, Laxer, & Lindsley, 2010). A noteworthy feature of JRDs is the recurrent, unpredictable course and flare-ups, during which youth may experience significant pain, limited mobility, and fatigue (Sandstrom & Schanberg, 2004). In fact, Gerhardt and colleagues (2003) noted that as many as 75% of youth with JRDs may experience significant physical limitations as well as medical problems, including growth retardation and impaired vision.

Consistent with a family systems perspective, a stressor such as a chronic illness in any member of the family potentially affects all other members. Although caregiving is a normal part of parenting, providing high levels of care and illness management required by a child with a chronic illness can become burdensome and may impact both the physical and psychological health of the caregiver (Raina et al., 2004; Wolfe-Christensen, Mullins, Fedele, Rambo, Eddington, & Carpentier, 2010). Following the diagnosis of a JRD, for instance, parents often serve as primary medical caregivers, a role accompanied by new, complex responsibilities and considerable stress. Depending on the age and functional ability of a child with a JRD, caregivers are responsible for various aspects of

illness management, including administration of medication and injections, refilling prescriptions as needed, taking the child to doctor appointments, encouraging behaviors that help to prevent flare-ups (e.g., exercise, use of splints, massage), managing chronic pain (e.g., comfort, analgesics), and helping the child cope with their illness, among others (Cassidy et al., 2010). For parents of children with a JRD, the role of caregiver is a long-term endeavor, especially as the risk for functional limitations increases with severity and disease activity.

The extant literature has demonstrated the relationship between the demands of caring for a child with a chronic illness and negative physical and psychosocial outcomes among caregivers (Bauer, Vedhara, Perks Wilcock, Lightman, & Shanks, 2000; Brouwer, van Exel, van de Berg, Dinant, Koopmanschap, & van den Bos, 2004; Moskowitz et al., 2007; Scanlan, Vitaliano, Ochs, Savage, & Soo, 1998). For instance, the burden or stress associated with caring for a child with a chronic illness has been shown to influence immune dysfunction and disease susceptibility among caregivers, especially when the illness is chronic in nature (Kuster & Merkle, 2004), as prolonged stress associated with chronic illnesses may result in habituation to stress-response hormones (e.g., Herbert & Cohen, 1993) or immune suppression (e.g., Olff, 1999). Moreover, when financial constraints are present and relationships with the medical staff are strained, physical symptoms may be experienced by caregivers (Patterson, Leonard, & Titus, 1992).

In the adult rheumatoid arthritis (RA) literature, studies have examined the impact of caregiver demand on physical health and found that while caregivers are healthy, on average, these caregivers of patients with severe RA are relatively unhealthy, evidencing decreased mobility and self-care along with increased pain. This study underscores the

chronic nature of rheumatic diseases, as caregivers had been caring for RA patients for an average of 11 years (Brouwer et al., 2004).

Overall, a fairly consistent association exists between caregiving and poorer physical health. Strong relations between caregiver physical and psychological health have also been well documented (Kuster & Merkle, 2004), and given the aforementioned caregiving tasks associated with caring for a child with JRD, it is not surprising that these caregivers are at risk for poor psychological outcomes (e.g., increased distress). In general, research has shown that in comparison to healthy community samples of children, distress of parents caring for children with chronic conditions is higher (e.g., Canning, Harris, & Kelleher, 1996; Jessop, Reissman, & Stein, 1988). There are several determinants of caregiver well-being, including illness severity, the amount of time that is required for managing illness-related responsibilities, functional status, and the complexity of the treatment regimen (Bruns, Hilario, Jennings, Silva, & Natour, 2008; Moskowitz et al., 2007; Raina et al., 2004; Stephenson, 1999). For instance, Raina and colleagues (2005) examined the psychological and physical health of primarily female caregivers of children with cerebral palsy and found that parental adjustment was related to caregiving demands and child behavior, as well as a mediating role of perceived support and family functioning.

Although there is general support for the impact of child chronic illness on the physical and psychosocial adjustment of caregivers, results are mixed. Several have demonstrated that JRDs negatively impact family functioning (e.g., conflict), financial stability, and abrupt changes in family routines (Reisine, 1995; Vandvik, Hoyeraal, & Fagertun, 1989). On the other hand, some have found no significant adjustment

difficulties among caregivers of children with a JRD (Bruns et al., 2008; Daltroy et al., 1992; Harris, Newcomb, & Gewanter, 1991). Caregivers of children with JRDs have been shown to demonstrate adaptive skills similar to caregivers of healthy children (Harris et al., 1991), as well as comparable levels of anxiety and functional and social support (Gerhardt et al., 2003). Notably, Bruns and colleagues (2008) examined the quality of life and perceptions of burden on caregivers of children with Juvenile Idiopathic Arthritis (JIA) and found that burden was associated with caregiver emotional functioning rather than the child's illness variables (e.g., severity or functional status). In other words, the type or degree of physical limitation did not influence the caregiver's quality of life, suggesting that the main impact of JRDs on caregiver distress is related to the emotional aspects or perceptions of the disease (Bruns et al., 2008). Similarly, Vandvik and Eckblad (1991) found no significant association between disease severity and psychosocial outcomes (e.g., anxiety and depression) among female caregivers of children with JIA.

Therefore, it stands to reason that not all caregivers will experience distress, in part because of variability in stress appraisals (e.g., perceived support or parenting stress), which can either function to protect or exacerbate the likelihood of increased distress (e.g., Bruns et al., 2008; Manuel, Naughton, Balkrishnan, Paterson-Smith, & Koman, 2003). One such stress-processing factor that has been shown to be associated with adjustment outcomes is attitude toward illness, defined as an individual's evaluation of the medical condition, either favorable or unfavorable (e.g., Eagly & Chaiken, 1993; Olson, Goffin, & Haynes, 2007; Zanna & Rempel, 1988).

Children's attitudes toward their illness have been found to predict various adjustment-related outcomes, including depression and anxiety, self-esteem, behavioral problems, and functional abilities, across illness groups (e.g., Heimlich, Westbrook, Austin, Cramer, & Devinsky, 2000; Iobst, Nabors, Burnner, & Precht, 2007; LeBovidge, Lavigne, & Miller, 2005). Children who focus on the positive aspects of the illness experience and on what they can do rather than what they can't do are less likely to internalize their problems and more likely to display resilience than children with a negative attitude toward the illness (e.g., Austin & Huberty, 1993; Iobst et al., 2007; LeBovidge et al., 2005; Murphy, 1974). On the other hand, children who view their illness as debilitating to themselves and their family evidence maladaptive coping skills and social isolation (Lefebvre, 1983), lower self-efficacy and self-acceptance (Heimlich et al., 2000), and poorer family functioning (Iobst et al., 2007). Interestingly, however, research on caregivers' cognitive appraisals, including attitudes, of their child's JRD is relatively scarce.

Yet, based on findings that child attitudes influence their adjustment outcomes, it stands to reason that caregiver perceptions regarding their child's illness can influence parental distress, and ultimately child adjustment outcomes (Thompson, 1985). Indeed, parental adjustment to the child's JRD has demonstrated temporal precedence and a downstream effect on child depressive symptoms over time (Ryan, Ramsey, Fedele, Mullins, Chaney, & Jarvis, 2010), providing additional support for the need to examine the relation between parental attitudes and distress. Thus, the purpose of the current study is to address gaps in the literature by examining the relations between caregiver demands, parental attitudes toward the child's JRD, and parental distress. Specifically, the present

study aims to examine the relationship between caregiver demand and parental distress through the mediation of parent attitude toward illness. If parent attitudes are found to mediate this relation, it will provide a foundation for the development of interventions aimed at altering negative attitudes, which in turn has the potential to improve adjustment outcomes for both the parent and ultimately the child.

The subsequent chapter is a review of the extant literature relevant to the proposed project. This review is divided into six major sections. The first three sections will focus on issues related to diagnosing JRDs, subtypes and classification, prevalence, and prognosis. The fourth section includes an overview of treatments for JRDs, including medical, psychological, and alternative considerations. The fifth section discusses the responsibilities associated with caring for a child with a JRD and the physical and psychological difficulties that may be encountered by parents. Finally, the chapter will conclude with a discussion of attitudes toward illness and how this cognitive appraisal variable may be associated with parent distress.

CHAPTER II

REVIEW OF THE LITERATURE

Diagnostic Considerations

Juvenile Rheumatic Diseases (JRD) are a series of autoimmune disorders characterized by a similar presentation of symptoms, including joint pain and swelling, connective tissue inflammation, chronic pain or tenderness in the joint(s), and possible limited movement in the affected joint(s) (Cassidy et al., 2010). Some rheumatic conditions also involve internal organs and abnormalities of the immune system. As the term indicates, the cause of juvenile idiopathic arthritis (JIA) is unknown. Correctly diagnosing JIA and other rheumatic diseases is often a challenge, as many symptoms are similar among the different diseases yet vary across individuals and in terms of disease severity. Further complicating matters, no specific laboratory test can establish the diagnosis of JIA, and thus, it is a diagnosis of exclusion. An accurate diagnosis requires the following array of medical information: data from history, physical examination, laboratory testing (e.g., antinuclear antibody, complement, hematocrit), x-rays and other imaging tests (e.g., MRI), and subjective pain measurement (Lovell, 2008). However, there exist children that defy the classification system and present with features of several concurrent rheumatic diseases, or with a subset of symptoms. It may take years before enough disease manifestations develop before a definitive diagnosis can be made (if ever). Additionally, some children's illness will evolve from one diagnostic

category into another once specific disease markers are present (Cassidy et al., 2010). Consequently, a diagnostic process that is typically designed to provide clarity and reassurance to the family is often times marked by the unpredictable and unstable nature of the disease, further exacerbating stress and the potential for adjustment difficulties.

Disease Subtypes

As previously stated, JRDs consist of a series of autoimmune disorders including juvenile idiopathic arthritis (JIA), systemic lupus erthematosus(SLE), juvenile dermatomyositis (JDMA), and juvenile spondylarthropathies. Although they share several features, there are some distinctive characteristics, such as the number of affected joints and the degree of restricted movement.

Juvenile idiopathic arthritis (JIA), often referred to as juvenile rheumatoid arthritis (JRA), is the most commonly diagnosed JRD, with prevalence rates estimated to be between 57 and 220 per 100,000 children younger than 16 years (Borchers, Seemi, Chema, Keen, Sheonfeld, & Gershwin, 2006; Cassidy et al., 2010; Oen & Cheang, 1996). Although JIA affects a much smaller number of individuals compared to adult-onset arthritis, it affects approximately the same number of children as juvenile or Type 1 diabetes, at least four times as many children as sickle cell disease and cystic fibrosis, and 10 times as many as acute lymphocytic leukemia, hemophilia, or muscular dystrophy (Gortmaker & Sappenfield, 1984). In addition, JIA affects girls twice as often as boys and is more common in Caucasian children than in either African American or Asian American children (Lovell, 1997).

Key clinical features include swollen, stiff, and painful joints, inflammation, warmth and redness in the joint(s), pain, and fatigue (Woo, 2008). According to the

criteria of the American College of Rheumatology, the criteria for JIA require disease onset before 16 years of age, persistent and objective arthritis in one or more joints for at least six weeks, and exclusion of other causes of childhood arthritis (e.g., illness or injury; Cassidy et al., 2010; Hofer & Southwood, 2002).

The disease can be classified into seven categories depending on the number of/and which joints are involved, the symptoms present and their duration, and the presence or absence of specific antibodies. JIA is categorized into one the following subtypes: systemic, polyarthritis rheumatoid factor positive, polyarthritis rheumatoid factor negative, oligoarthritis (persistent or extended), psoriatic, enthesitis-related, and undifferentiated.

The most serious subtype, systemic JIA (sJIA), occurs independent of both age and gender and affects approximately 2-7% of children with JIA (Cassidy et al., 2010). This type of JIA is associated with high fevers, an evanescent and nonfixed rash, generalized lymph node enlargement, arthritis, and in some children, inflammation of internal organs (e.g., heart, liver, spleen). The following criteria need mention for a proper diagnosis: the fever must fall back to normal at least once each day, the rash should have a salmon pink appearance (not like a bruise), and the rheumatoid factor should not be present (Lovell, 2008). For some children, the fever and rash may disappear after the first few months of the illness, while the joint-related pain may persist for a longer period of time. The long-term prognosis for sJIA is determined by the severity of the arthritis, with inflammatory eye problems occurring less frequently compared to other types of JIA (Kotaniemi, Kaipiainen-Seppänen, Savolainen, & Karma, 1999).

To be classified as polyarthritis rheumatoid factor positive (poJIA), the child must have at least two positive results for rheumatoid factor (RF) at least three months apart during the first six months of the disease (Lovell, 2008). Approximately 2-17% of all children with JIA have RF+ poJIA, and 10-28% are diagnosed with RF- poJIA. Although onset is not necessarily age-specific, girls are three times more likely than boys to be diagnosed with this subtype (Lehman, 2004). Involving five or more joints, poJIA is serious and tends to affect the small joints (e.g., hands and feet) on both sides of the body (i.e., symmetrical arthritis). Common symptoms include a positive blood test for rheumatoid factor (RF+), bumps on parts of the body (e.g., elbow) that receive a lot of pressure from chairs, shoes, or other objects, and low red blood cell count (i.e., anemia; Cassidy et al., 2010). This is a heterogeneous group of diseases. For instance, there are two major peaks in the age of disease onset; between eighteen months and eight years of age and after eleven years of age. Furthermore, some children begin with arthritis in one or two joints, slowly spreading to other joints, while other children rapidly develop arthritis in multiple joints (Lehman, 2004).

Children with this subtype are at risk for developing chronic eye problems (e.g., uveitis) and should be evaluated by an ophthalmologist on a regular basis. Of similar importance, children with poJIA may develop permanent damage to some of their joints. For example, arthritis in the jaw may cause pain and discomfort in chewing, affecting dental care and eating habits, and ultimately impinging the growth process (Cassidy et al., 2010).

Oligoarthritis JIA (oJIA) is the most common form of JIA involving four or less joints and typically affects larger joints (e.g., knee, shoulder, hip). This type is further

divided into two subtypes: persistent and extended. Persistent oJIA never has more than four cumulative arthritic joints during the disease course, whereas extended oJIA affects five or more joints after the first six months of the disease. Occurring in approximately 24-58% of children with JIA, this type of disease most often affects girls younger than eight years (Lovell, 2008). Although arthritis is typically the only disease manifestation, children with this subtype have a 30-50% chance of developing inflammatory eye problems, and children younger than eight years are at greater risk of developing an adult form of arthritis (Kotaniemi et al., 1999).

Children presenting with chronic arthritis in association with psoriasis with an onset before 16 years are often diagnosed with psoriatic JIA (pJIA). Of note, the psoriatic rash may not appear until years after the onset of the arthritis. Other common manifestations include swelling of one or more fingers, nail pitting, and partial loosening or complete detachment of the nail from the nail bed (Hofer & Southwood, 2002). This form of JIA affects 2-10% of children with JIA, and the majority of cases involve peripheral, asymmetric arthritis in the knees, ankles, and small joints of the hands and feet (Lovell, 2008).

Enthesitis-related JIA (eJIA) is reserved for children who present with arthritis and inflammation at the insertion of the ligament, tendon, or joint capsule into the bone (i.e., enthesitis). In eJIA, tests for RF are negative, and arthritis will typically involve four or more joints (Lovell, 2008). Approximately 10% of all children with JIA are classified as eJIA.

Lastly, children who fit into more than one category or who do not satisfy all the inclusion criteria for any one category, the undifferentiated (uJIA) category is to be used.

Of those diagnosed with JIA, 2-23% are classified as uJIA, with 60% failing to meet the full inclusion criteria for one of the other JIA categories and 40% demonstrating criteria from more than one JIA category (Hofer & Southwood, 2002).

Although the following rheumatic diseases share the arthritic feature of the aforementioned JIA subtypes, they have distinct characteristics. Systemic lupus erythematosus (SLE), for example, is a disease characterized by periodic episodes of inflammation of the joints, tendons, and other connective tissue. Other systemic manifestations frequently include fever, loss of appetite, lethargy, weight loss, and fatigue. In severe cases, complications may include blood clots, strokes, and kidney and/or heart failure (Lovell, 2008). As opposed to the arthritis presentation in JIA, the affected joints are usually less swollen but more painful, but do not generally lead to joint damage or deformity (Lehman, 1997). SLE symptoms often emerge during early adolescence (10 years and older), with girls accounting for more diagnoses than boys. African Americans, Asian Americans, Latinos, and Native Americans are most often affected by SLE as compared to Caucasian Americans (Woo, Laxer, & Sherry, 2007). Similar to other rheumatic diseases, SLE is known to have flare-ups and periods of remission, although the severity of kidney involvement in SLE can vastly alter the outcome and survival rate of children with the disease.

Juvenile spondylarthropathies compromise a class of rheumatic diseases distinctly different from other rheumatic illnesses, with juvenile ankylosing spondylitis (JAS) as the most common subtype. Occurring two to three times more often in boys than girls, the onset of JAS typically takes place during late childhood or pre-adolescence (Lehman, 2004). JAS commonly causes pain and inflammation in the joints in the lower part of the

body, especially at the site of attachment of muscles, ligaments, and/or tendons to bone. Although prognosis is usually good if detected early (Lovell, 2008), severe cases can involve erosion at the joint between the spine and the hip bone and the formation of bony bridges between vertebrae in the spine, fusing the bones and permanently limiting mobility (Cassidy et al., 2010).

Lastly, juvenile dermatomyositis (JDM) is a disease characterized by muscle damage due to diffuse vasculitis (Woo et al., 2007). Symptoms often appear gradually and include fever, a rash around the eyelids and/or knuckles, joint pain and tenderness, and mouth ulcers. JDM is rare and affects girls two to five times more often than boys (Cassidy et al., 2010). Similar to other rheumatic diseases, the cause is unknown although JDM is usually triggered by a condition (e.g., infection, immunization, injury) that causes immune system activity that does not respond as it should (Feldman, Rider, Reed, & Pachman, 2008).

Prognosis

JRDs are unpredictable and unstable, often fluctuating between flare-ups and periods of remission (Cassidy et al., 2010). Approximately 80% of adolescents with JIA eventually enter remission with minimal functional loss or deformity (Lehman, 2004). However, because of further complications and the impact on other body systems, children and adolescents diagnosed with a JRD are more likely to have arthritis as adults and show greater mortality rates than the general population. Fortunately, JRDs are seldom life-threatening, with fatalities mostly occurring among children with SLE secondary to organ failure (Cassidy et al., 2010).

The key to a positive prognosis is the prevention and correction of potentially damaging affects with proper therapy. As with many other childhood diseases, the majority of children and families who keep their appointments and comply with medications and other interventions will do well as compared to those who regularly miss appointments for extended periods of time and refuse advice for more aggressive therapy (Lehman, 2004). Although the outlook is hopeful, there is no way to ultimately predict the outcome for children diagnosed with a JRD, further highlighting the importance of examining other factors associated with positive adjustment outcomes.

Treatment Considerations

Medical Treatments

For all children diagnosed with a JRD, the primary goals of any drug therapy are to reduce pain and inflammation of the joint(s) and maximize the individual's ability to perform activities of daily living. In the long-term, goals include the prevention of disease progression and destruction of bone, cartilage, and joints. The medical treatment for JRDs, JIA in particular, has changed dramatically over the past 15 years following research that concludes that most children will never reach long-term remission. For instance, studies have shown that 50-70% of children with poJIA or sJIA and 40-50% of those with oJIA will continue to have active joint inflammation and arthritis in adulthood (Hashkes & Laxer, 2005). Several predictors of a potentially poor outcome can help determine the need for early aggressive therapy, including poJIA, RF+, the presence of human leukocyte, nodules, and early-onset small joint arthritis.

The first line of therapy includes nonsteroidal anti-inflammatory drugs (NSAIDs). All NSAIDs interfere to varying degrees with the cyclooxygenase pathway, which is

responsible for the production of important inflammatory mediators (e.g., prostaglandins) (Cassidy et al., 2010). At low doses, NSAIDs help with a variety of problems, ranging from muscle aches to pain and fever, whereas higher prescribed doses help reduce joint inflammation. NSAIDs fall into three general categories: traditional NSAIDs (i.e., COX-1), COX-2 inhibitors, and salicylates.

Traditional NSAIDs compromise the largest subset, with three of them in lower-strength doses and available without a prescription. Because of the side effects of many traditional NSAIDs, including bleeding and liver and stomach problems, COX-2 inhibitors (e.g., Celebrex, Vioxx) have been developed. COX-2 inhibitors are a class of NSAIDs found to be gentler on the stomach (Lehman, 2004). As with any medication, however, there is a risk of side effects such as cough, cold, upper respiratory tract infection, headache, fever, and nausea. COX-2 inhibitors have been found to increase the risk of heart problems in adults, and studies are currently examining similar risks in children (Ilowite, 2002). Once the most common and single most effective anti-inflammatory medications used in treating JIA, the use of salicylates (aspirin) has greatly declined due to the risk of Reye's syndrome, a rare but potentially lethal disease that attacks the liver and brain when a person is recovering from a viral illness (Lehman, 2004).

Although only a handful are approved by the U.S. Food and Drug Administration (FDA) for use in JIA (e.g., ibuprofen, naproxen, tolmetin, and choline magnesium trisalicylate), many other NSAIDs are commonly prescribed, including indomethacin and diclofenac (Ilowite, 2002). Because of a lack of consensus on the best NSAID for patients with JIA, many medications are chosen on the basis of considerations such as cost,

dosing schedule, patient preference, or medication taste. Approximately 25-30% of patients with JIA, mainly those with oJIA, respond well to NSAIDs; however, because NSAIDs do not alter the disease course or prevent joint damage, they are primarily used to treat joint pain and stiffness (Hashkes & Laxer, 2005)

When NSAIDs alone fail to reduce pain and inflammation, physicians may prescribe additional disease-modifying anti-rheumatic drugs (DMARDs) to slow the progression of JIA. Commonly used DMARDs in the treatment of JIA include methotrexate (Rheumatrex) and sulfasalazine (Azulfidine). Methotrexate is currently the cornerstone managing JIA and poJIA (Hashkes & Laxer, 2005). The efficacy of Methotrexate differs by subtype, but the greatest efficacy has been shown in patients with extended oJIA, while less effective in eJIA (Woo et al., 2000). DMARDs are typically started early because it may take up to three to four months before effects are noticed (Lehman, 2004). Common side effects include nausea, minor changes in the white blood cell or platelet count, and liver irritation. In some cases, children develop an aversion to Methotrexate that can be improved by teaching relaxation or meditation techniques (Hashkes & Laxer, 2005).

Biologic-modifiers are a new class of medications that have been shown to provide significant relief to children with JIA, including those who failed Methotrexate. Bilogic-modifiers work by targeting a specific molecule that plays an important role in the inflammatory process (Hashkes & Laxer, 2005). Enbrel, the first widely available biologic, has been found to be effective for most children with poJIA, spondyloarthropathies, and some children with sJIA. Enbrel works by interfering with the function of a molecule called tumor necrosis factor alpha (TNF-α), a messenger molecule

that often causes people to feel ill when released in large amounts (Lehman, 2004). Early studies suggest that Enbrel works within hours of the first dose and not only prevents symptoms of the disease, but also allows healing of the bone and joint damage to begin. Adverse effects are generally mild and include injection site inflammation, runny nose, skin reactions, and headaches. Further considerations prior to using Enbrel include its high cost and the subcutaneous injections required twice weekly (Lehman, 2004).

Psychological Treatments

While medical treatment of JIA serves its purpose in the management of the disease symptoms, psychological interventions have also been shown to be important adjunctive therapies that help patients manage pain, facilitate psychosocial adjustment, and enhance adherence to medication regimens. For example, cognitive-behavioral therapy (CBT) for pain management has demonstrated that by improving an individual's sense of self-efficacy, patients were more likely to report a reduction in pain and depressive symptoms and take their medication as prescribed (Buenaver, McGuire, & Haythornthwaite, 1996). Furthermore, CBT for children with JIA has been shown to reduce pain after the introduction of various self-regulatory techniques (Walco, Varni, & Ilowite, 1992), as well as improve coping and emotional stability (Leibing, Pfingsten, Bartmann, Ruger, & Schuessler, 1999).

In general, traditional CBT for pain management/coping skills training consists of three phases: 1) psychoeducation about the biofeedback model of pain; 2) skills-training (e.g., relaxation training, pleasant activity scheduling, imagery, cognitive restructuring, problem solving, goal setting); and 3) practice and application of skills in real-life situations (Turk, 2002). Specifically, children between the ages of 4.5 and 16.9 years with

JIA were seen for eight individual sessions and taught a variety of techniques (e.g., muscle relaxation, meditative breathing). Results indicated a reduction in self-reported pain and increased adaptive functioning that continued at 6- and 12-month follow-up (Turk, 2002).

Complementary to CBT for pain management, biofeedback training helps patients increase control over their physiological processes that may contribute to the experience of pain. During training, a clinician educates the patient on physiological responses to stress and pain, often with the use of an electronic device that records physiological signals (e.g., heart rate, blood pressure). The goal is to help the patient gain control over the responses (e.g., muscle tension, anxiety) that can contribute to pain (Dixon, Keefe, Scipio, Perri, & Abernethy, 2007). Indeed, Achterberg and colleagues (1981) examined the utility of relaxation and biofeedback strategies in a sample of 24 patients diagnosed with rheumatoid arthritis (RA) and found that while measures of psychological adjustment tended to remain constant pre- and post-treatment, biofeedback resulted in improved physical functioning (e.g., pain, sleep, and heart rate).

Unlike CBT and biofeedback for pain management, stress management training primarily focuses on managing stress, not pain per se. Similarly, however, stress management training includes skill building, such as deep breathing, meditation, relaxation training, and visual imagery. Although the major goal is the reduction of stress, it is expected that this will translate into better symptom control and pain management (Dixon et al., 2007). For instance, Rhee and colleagues (2000) examined the relation between stress management training in addition to rheumatologic care versus rheumatologic care alone in a sample of 47 adults with RA. They found that significant

decreases in pain and depressive symptoms in the adjunctive group were associated with increased self-efficacy, coping strategies, and perceptions of control regarding arthritis.

Another more recent treatment option that has received increased attention is operant interventions, focused on modifying the child's overt manifestations of pain (e.g., crying, groaning, and grimacing). The use of these procedures arose from speculation that pain behaviors are maintained by positive or negative reinforcement, such as extra time with a parent, visits from friends, or gifts from friends or family. Allen and Shriver (1998) examined the frequency in which parents engaged in pain response behaviors, such as attending to the pain, assisting with treatment, and suggesting or allowing a reduction in activity level, comparing biofeedback alone and in combination with an operant intervention. Children between the ages of 7- and 18-years with migraine headaches received positive reinforcement for adaptive coping strategies and maintaining daily activities and no reinforcement for inappropriate pain behaviors. Although both groups reported significantly less migraines, children in the operant intervention condition made greater gains and were more likely to be free of migraines at the end of treatment and at three-month follow-up. However, the groups were not significantly different at one-year follow-up (Allen & Shriver, 1998).

Lastly, because children with JIA face numerous stressful situations associated with the acute exacerbations of their disease and the long-term functional disability that may follow, the family has been identified as an important source of support in the coping process (Varni, Wilcox, & Hanson, 1988). For instance, Wallander and Varni (1989) demonstrated that chronically ill or handicapped children with high social support showed significantly better psychological adjustment when compared to those with low

social support. Several earlier studies have further supported the positive relationship between social support and functioning in adults with RA (Kaplan & Delongis, 1983; Nicassio, Brown, Wallston, & Szydlo, 1985; Weinberger, Hiner, & Tierney, 1986).

Currently, youth with a JRD are the primary focus of assessment and psychological interventions. However, recent findings by Ryan and colleagues (2010) suggest that the family system can be an important target of empirical investigation and clinical intervention in the JRD population. Specifically, Ryan and colleagues (2010) found that child distress may reflect the downstream effects of parent distress, and as such, early interventions aimed at parent distress and coping may be important when addressing the child's adjustment concerns. Unfortunately, to our knowledge, no such interventions have been developed at this time.

In sum, a number of psychosocial treatments for JRDs have been developed, with each offering a unique aspect of treatment depending on various disease and environmental factors, such as disease severity or subtype, and the cost, preference, and feasibility of the treatment options.

Alternative Treatments

The medical treatment of JRDs is only one facet of therapy. A multidisciplinary team of pediatric rheumatologists, opthalmologists, dentists, physical and occupational therapists, dietitians, psychologists, and educational counselors are involved in treating patients with JRDs.

Because potential side effects and long-term damage are a concern to most parents of children with JIA, many inquire about alternatives, including vitamins and supplements. Lehman (2004) does not suggest replacing prescription medication, but

rather to assess the child's diet and individual needs when considering adding the aforementioned alternatives. Children, especially those with a chronic condition, should be on a daily vitamin that contains the appropriate amounts of vitamins A, B, C, D, E, and K, folic acid, iron, and calcium. Again, it is important to consult with a physician because large amounts of some vitamins (e.g., A and D) can cause severe illness and even death.

Dietary consultation may also be needed because some children with JIA have a poor appetite and inadequate growth from several factors including active disease, arthritis of the TMJ, and various medications (Hashkes & Laxer, 2005). Consulting with a dietitian is also important for children prescribed corticosteroids in preventing excessive weight gain, bone loss, and hypertension.

When considering supplements, it is important to remember that "all-natural" does not necessarily mean safe. One supplement that has been shown to be of benefit to people with arthritis is glucosamine, an ingredient used to manufacture cartilage. Omega-3 fatty acids are another supplement that has been recommended for people with arthritis. However, early studies in the 1980s showed initial improvement in six to eight weeks with a slow return to impairment in the long run (Lehman, 2004). Additionally, patients were consuming 10 or more capsules a day in order to reach the claimed effect.

In addition to the claimed effects of medication and supplements, therapeutic exercise has been shown to be beneficial to children with JIA. Both physical and occupational therapy can make it easier to move difficult joints but should be tailored by the degree of arthritis and the joints involved. While medication helps reduce pain and inflammation, only therapeutic exercise can restore lost motion in a joint (Lehman,

2008). Examples include range-of-motion exercises used to keep joints flexible and make it easier for children to perform daily living skills, such as eating, writing, and dressing. In general, activities that are less weight bearing, such as swimming or cycling, are preferred but most activities that do not involve contact (e.g., football, hockey, wrestling, etc.) are acceptable.

Other daily activities include hot and cold treatments and massages. Heat treatment is used for decreasing the rigidity of the joints while increasing the flexibility of the fibrous tissue in the joint capsules and tendons. Applying heat treatment before therapeutic exercises will increase the efficiency of the treatment. Cold treatment, on the other hand, is used for analgesic and vasoconstriction purposes in inflamed joints. Daily massages can help with pain and prevent adhesions in the subcutaneous tissues (Lehman, 2008). Field and colleagues (1997) found that children with mild to moderate JIA who were massaged 15 minutes a day for 30 days reported a decrease in pain, congruent with parent- and physician-reports. Furthermore, there was also a reduction in anxiety and cortisol levels as compared to the control group.

Another option that a physical or occupational therapist may recommend is a splint or orthotic, often used to help keep joints in the correct position and relieve pain (Lehman, 2008). If a joint is at risk of becoming permanently deformed, a splint may help position and stretch it back to its normal position. Some commonly used splints include knee extension splints, wrist extension splints, and ring splints for the fingers.

In sum, although development of new therapies has markedly increased the ability to effectively treat children with JRDs, the best or optimal treatment remains elusive in many regards and will continue to pose a challenge to clinicians. There is also a lack of

evidence-based medicine in the treatment of some JIA subtypes. Our inability to predict individual outcomes with 100% accuracy remains a barrier to optimal disease management. Thus, additional studies on factors associated with favorable and unfavorable prognoses will be invaluable in guiding the most suitable and appropriate treatment.

What is clear is that treatments for JRDs, whether they be medical or psychosocial, have the potential to place great demands on parents. In the section to follow, the literature on caregiver demands in the context of JRDs will be reviewed.

Caregiver Demand in JRDs

Consistent with a family systems perspective, a stressor such as a chronic illness in any member of the family potentially affects all other members. Although caregiving is a normal part of parenting, providing high levels of care and illness management required by a child with a chronic illness can become burdensome and may impact both the physical and psychological health of the caregiver (Raina et al., 2004; Wolfe-Christensen et al., 2010). Following the diagnosis of a JRD, parents often serve as primary caregivers, a role accompanied by new responsibilities and considerable stress. For instance, depending on the age and functional ability of a child with JRD, caregivers are responsible for various aspects of illness management, including administration of medication and injections, refilling prescriptions as needed, taking the child to doctor appointments, encouraging behaviors that help to prevent flare-ups (e.g., exercise, use of splints, massage), managing chronic pain (e.g., comfort, analgesics), and helping the child cope with their illness, among others. For parents of children with a JRD, the role of

caregiver is a long-term endeavor, especially as the risk for functional limitations increases with severity and disease activity.

Caregiver Demand and Physical Health Outcomes

The extant literature has demonstrated the relationship between the demands of caring for a child with a chronic illness and physical and psychosocial outcomes among caregivers (Bauer et al., 2000; Brouwer et al., 2004; Moskowitz et al., 2007; Scanlan et al., 1998). For instance, the burden or stress associated with caring for a child with a chronic illness has been shown to influence immune dysfunction and disease susceptibility, especially when the illness is chronic in nature (Kuster & Merkle, 2004). Unlike caring for a child with an acute illness, prolonged stress associated with chronic illnesses may result in habituation to stress-response hormones (e.g., Herbert & Cohen, 1993) or immune suppression (e.g., Olff, 1999). Moreover, when financial constraints are present and relationships with the medical staff are strained, physical symptoms may be experienced by caregivers (Patterson et al., 1992). The implications are concerning considering that effective immune functioning is necessary for cell growth and the fight against micro-organisms.

In the adult rheumatoid arthritis literature, studies have examined the impact of perceived caregiver burden on physical health and found that while caregivers are healthy, on average, those caring for an adult with severe RA are relatively unhealthy with decreased mobility and self-care along with increased pain. This study underscores the chronic nature of rheumatic diseases, as caregivers had been caring for RA patients for an average of 11 years (Brouwer et al., 2004).

To summarize, studies have examined the relationship between caregiving and adverse physical health outcomes using a range of health indicators. Overall, a fairly consistent association exists between caregiving and poorer physical health. Strong relations between caregiver physical and psychological health have been well documented (Kuster & Merkle, 2004), and the following section will further describe the association between the demands of caring for a child with a chronic illness and its impact on caregivers' psychological health.

Caregiver Demand and Psychosocial Outcomes

Given the aforementioned caregiving tasks, it is not surprising that caregivers of children with a chronic illness are at risk for increased distress. In general, research has shown that in comparison to healthy community samples of children, distress of parents caring for children with chronic conditions is higher (e.g., Canning et al., 1996; Jessop et al., 1988). Several studies of caregivers of chronically ill children indicate that there are several determinants of caregiver well-being, including illness severity, the amount of time that is required, functional status, and the complexity of the treatment regimen (Bruns et al., 2008; Moskowitz et al., 2007; Raina et al., 2004; Stephenson, 1999). For instance, in a sample of 14 maternal caregivers of children with sickle cell disease, perceived burden was associated with the unpredictable nature of the pain crises as well as illness stigma (Moskowitz et al., 2007). Likewise, Raina and colleagues (2005) examined the psychological and physical health of primarily female caregivers of children with cerebral palsy and found that parental adjustment was related to caregiving demands and child behavior, in addition to the mediating role of perceived support and family functioning.

Notably, social support has been shown to protect parents from the potentially detrimental effects of caregiving demands (Wang, Wu, & Liu, 2003; Weiss, 2002). Emotional and physical support from a spouse and close friends are significant predictors of stress in mothers of children with a chronic illness (Holaday, 1997). Perceived social support has also been shown to moderate the relationship between the impact of child functioning and maternal depressive symptoms in a sample of children with cerebral palsy (Manuel et al., 2003).

Although there is general support for the impact of child chronic illness on the physical and psychosocial adjustment of caregivers, results are mixed. Several have demonstrated that JRDs negatively impact family functioning (e.g., conflict), financial stability, and abrupt changes in family routines (Reisine, 1995; Vankvik et al., 1989). On the other hand, some have found no significant adjustment difficulties among caregivers of children with a JRD (Bruns et al., 2008; Daltroy et al., 1992; Harris et al., 1991). Caregivers of children with JRDs have been shown to demonstrate adaptive skills similar to caregivers of healthy children (Harris et al., 1991), as well as comparable levels of anxiety and functional and social support (Gerhardt et al., 2003). Bruns and colleagues (2008) examined the quality of life and perceptions of burden on caregivers of children with JIA and found that burden was associated with caregiver emotional functioning rather than the child's illness variables (e.g., severity or functional status). In other words, the type or degree of physical limitation did not influence the caregiver's quality of life, suggesting that the main impact of JRDs on caregiver distress is related to the emotional aspects or perceptions of the disease (Bruns et al., 2008). This is consistent with a study by Vandvik and Eckblad (1991), in which there was no significant association between

disease severity and psychosocial outcomes (e.g., anxiety and depression) among female caregivers of children with JIA.

In sum, although caregivers of children with a JRD are generally at greater risk for a variety of difficulties (e.g., Barlow, Harrison, & Shaw, 1998; Raina et al., 2004; Reisine, 1995), not all caregivers will experience distress, in part because of variability in stress appraisals, such as attitudes towards the child's illness. In fact, a number of evaluative variables, including perceived support, perceived stress, and other cognitive factors function to protect or exacerbate the likelihood of adjustment problems (e.g., Bruns et al., 2008; Manuel et al., 2003). Interestingly, however, research on caregivers' cognitive appraisals, including attitudes, of their child's JRD is relatively scarce.

Attitude Toward Illness

Research has indicated that caregivers of children with a chronic illness face a host of potential difficulties as a result of the unpredictable nature of the disease state and demands placed upon the caregiver (Canning et al., 1996; Jessop et al., 1988; Moskowitz, 2007; Raina et al., 2005). However, poor caregiver adjustment outcomes are not universal, and they are often influenced by individual differences in stress-processing factors, such as attitudes.

In psychology research, an attitude is typically defined as an individual's evaluation of a target, either favorable or unfavorable (e.g., Eagly & Chaiken, 1993; Olson et al., 2007; Zanna & Rempel, 1988). Direct self-report measures have been the most common form of attitude assessment because respondents presumably are aware of their attitudes and willing to report their attitudes honestly as long as the demands of social desirability are low. Indeed, literature supports the validity of self-report measures

of attitudes with their ability to predict a variety of other variables, including behavior, knowledge, and past events (Albarracin, Johnson, & Zanna, 2005). Attitude toward illness is no exception; self-report measures have been found to predict various adjustment-related outcomes among children, including depression and anxiety, self-esteem, behavioral problems, and functional abilities, across illness groups (e.g., Heimlich et al., 2000; Iobst et al., 2007; LeBovidge et al., 2005).

Several studies have found that children who focus on the positive aspects of the illness experience and what they can do rather than what they can't do, are less likely to internalize their problems and more likely to display resilience than children with a negative attitude toward the illness (e.g., Austin & Huberty, 1993; Iobst et al., 2007; LeBovidge et al., 2005; Murphy, 1974). If, on the other hand, children have a negative attitude toward their illness (e.g., the illness is viewed as debilitating to the child and family), these negative feelings can contribute to the demands and stress already placed on the family by the illness. For instance, Lefebvre (1983) examined the influence of attitude toward illness on coping and adjustment and found that children with a handicap who viewed their illness negatively were more likely to engage in maladaptive coping skills and become socially isolated compared to children with a handicap who reported a positive attitude toward their illness.

Similarly, Heimlich and colleagues (2000) found that among a sample of adolescents diagnosed with epilepsy, those with negative attitudes reported lower self-efficacy and self-acceptance as compared to adolescents with a more positive outlook. Of note, the study found that girls, older adolescents, and those with more severe epilepsy

reported more negative attitudes toward their illness than boys, younger adolescents, and those with mild to moderate epilepsy (Heimlich et al., 2000).

Specific to illness-related factors, Iobst and colleagues (2007) examined the relationship between disease severity (e.g., experience of pain and fatigue) and attitude toward illness among youth diagnosed with a JRD, finding that those experiencing more pain and fatigue were more likely to report negative attitudes and experience poorer family functioning than those with lower levels of pain and fatigue. In addition, children and adolescents who reported more positive attitudes had parents who tended to report higher, more positive family functioning.

Together, these studies illustrate the relationship between attitude toward illness and adjustment outcomes among a variety of chronic illnesses. Most research on adjustment to pediatric chronic illness has focused on the child's attitudes and feelings about their illness and has failed to include attitudes toward the child's illness from the perspective of the parent or primary caregiver. Yet, based on the transactional stress and coping model (Thompson, 1985), parental perceptions and stress related to the child's illness hypothetically can either increase the risk for or protect the child and parent from further psychological distress. Indeed, parental adjustment to the child's JRD has demonstrated temporal precedence and a downstream effect on child depressive symptoms over time (Ryan et al., 2010).

As such, it is important to continue research in this area, as positive attitudes toward chronic illnesses may foster resiliency in terms of adaptive coping skills and positive psychosocial and physical outcomes among caregivers. Perhaps the most important clinical reason to explore the relationship between attitude toward illness and

adjustment outcomes is because attitudes, when defined as evaluative judgments and their associated feelings, are amenable to change. Therefore, while illness-related variables (e.g., experience of pain and fatigue) and caregiver demands (e.g., time or effort put into caretaking responsibilities) are difficult to alter, the potential to effect attitude change has far-reaching implications for both the parents and the family as a whole.

Thus, the purpose of the current study was to address gaps in the literature by examining the relations between caregiver demands, parental attitudes toward the child's JRD, and parental distress. Specifically, the present study examined the relationship between caregiver demand and parental distress through the mediation of parent attitude toward illness. If parent attitudes are found to mediate this relation, it will provide a foundation for the development of interventions aimed at altering negative attitudes, which in turn has the potential to improve adjustment outcomes for both the parent and ultimately the child.

CHAPTER III

PRESENT STUDY

Based on the aforementioned review of the literature, it is apparent that parents of children diagnosed with a JRD are at risk for psychosocial difficulties, including depression and anxiety, social withdrawal, marital conflict, and financial hardship. From the time of diagnosis, parents often take on caregiving responsibilities above what is typical for healthy children. Specific to JRDs, this includes administering medication and injections, refilling prescriptions as needed, taking the child to doctor appointments, encouraging behaviors that help to prevent flare-ups (e.g., exercise, use of splints, massage), managing chronic pain (e.g., comfort, analgesics), and helping the child cope with their illness, among others. For parents of children with a JRD, the role of caregiver is a long-term endeavor, especially as the risk for functional limitations increases with severity and disease activity.

Although caregiving is a normal part of parenting, providing high levels of care and illness management required by a child with a chronic illness can become burdensome, with resulting impact on both the physical and psychological health of caregivers (e.g., Barlow et al., 1998; Herbert & Cohen, 1993; Kuster & Merkle, 2004; Raina et al., 2004; Reisine, 1995). However, not all caregivers will experience distress or poor adjustment outcomes related to caregiver demands as a result of variability in perceptions and attitudes towards the child's illness.

It has been documented that a child's attitude toward illness is associated with adjustment outcomes. Therefore, it stands to reason that parent attitudes towards their child's illness will be associated with caregiver demand and parental distress. Because parental distress influences child adjustment to a chronic illness, such information could be useful in the development of appropriate interventions aimed to change negative attitudes and ultimately, parent and child psychosocial health.

The current investigation was designed to extend existing pediatric literature by 1) examining the relation between caregiver demand and parental distress; 2) examining the direct effects of parent attitude toward the illness on parental distress; and 3) examining the potential mediating role of parent attitude toward the illness in the relation between caregiver demand and parental distress. Accordingly, the hypotheses of the current study were as follows:

Hypothesis 1: Consistent with previous studies, it was hypothesized that caregiver demand, as measured by the adapted Caring for My Child with a Rheumatic Disease Scale (Caregiver Demand), would be positively related to parental distress, as measured by the Global Severity Index (GSI) of the Brief Symptom Inventory (BSI).

Hypothesis 2: Consistent with previous studies examining the association between child attitudes and adjustment outcomes, it was hypothesized that parent attitudes toward their child's illness would be negatively related to parental distress (BSI_GSI).

Hypothesis 3: Because caregiver demand may lead to increased negative attitudes toward the illness for the parent, resulting in higher self-reported distress, parent attitude

toward their child's illness was examined as a potential mediator. It was hypothesized that the absolute size of the direct effect between caregiver demand and parental distress would be significantly reduced after controlling for parent attitudes, thus revealing mediation.

CHAPTER IV

METHOD

Participants and Procedure

Participants for the current study included 69 children and adolescents between the ages of 7 and 18, who were diagnosed with either juvenile idiopathic arthritis (JIA), systemic lupus erthematosus (SLE), juvenile dermatomyositis (JDMA), juvenile spondylarthropathies (JAS), or other rheumatic diseases (e.g., mixed connective tissue) and their parents. Please see Table 1 for participant demographic information.

Participants were be recruited from the pediatric rheumatology clinic at Children's Hospital of Oklahoma. Inclusion criteria for participation were as follows: 1) a diagnosis of one of the above-mentioned JRDs and 2) between the ages of 7 and 18. Exclusion criteria were as follows: 1) parent or child demonstrates comorbid cognitive deficit (e.g., intellectual disability), 2) child demonstrates comorbid chronic illnesses, and 3) parent or child are non-fluent English speakers.

Once the pediatric rheumatologist determined that a patient was eligible for participation, a research assistant trained in informed consent and HIPAA recruited each participant. The current study was approved by the Oklahoma University Health Sciences Center and Oklahoma State University Institutional Review Board (IRB), and all aspects of the project were conducted in compliance with the APA ethical guidelines for research. Written informed assent and consent were obtained from each participant,

parent, or legal guardian. Participants completed packets in the clinic or at home and then returned them via postage-paid mail. The total time commitment for each family to complete the questionnaires was approximately 60 minutes. Upon completion, participants were compensated with a $20 check. One-hundred and nineteen parents of children with a rheumatic disease were approached for recruitment into the current study. Of the 118 parents consented to participate (consent rate = 99.16%), 66.95% (n = 79) of them completed the study. The remaining 39 participants took the measures home and did not return them, even after receiving in-clinic reminders and /or phone calls. Of those that provided reasons for not completing the study, not having time (n = 1) and feeling items asking about suicidal ideation were inappropriate for their child (n = 1) were noted.

Measures

Background Information Questionnaire

A background information questionnaire ascertained the following information: child's age and grade, parent's age, child and parent's ethnicity, child's living arrangement, the highest grade completed and occupations of the child's parents, parent marital status, annual household income, and child history of psychoactive medication and/or psychological counseling/therapy (see Appendix A).

Physician-Report Measure

Physician-Rated Functional Disability (PRFD; Hochberg, Chang, Dwosh, Lindsey, Pincus, & Wolfe, 1992). The pediatric rheumatologist completed a provider questionnaire to obtain information regarding diagnosis, date of diagnosis, and functional disability. PRFD was determined by rheumatologist classification of children into one of four functional classes ranging from class I (*limited or no disability in vocational and*

self-care activities) to class IV (*severe disability*) (e.g., Hochberg et al., 1992). This classification system has been shown to be a valid index of functional disability in children with JRD (Baildam, Holt, Conway, & Morton, 1995). The rheumatologist provided disability classifications following a routine physical examination (see Appendix B).

Parent-Report Measures

Caring for My Child with a Rheumatic Disease Scale (adapted from the Caring for My Child with Cancer Scale; CMCCS, Wells et al., 2002). The original CMCCS is a 28-item caregiver-report measure designed to assess caregiver demand. Respondents rated the degree of difficulty/effort and the amount of time it takes to complete a given task across five dimensions of caregiving: physical care of the child, emotional care of all family members, financial management, maintenance of family roles and functions, and communication with healthcare professionals and other related agencies. For purposes of the current study, the original CMCCS was adapted to be specific to JRDs. The measure was scored by multiplying the "difficulty/effort" and "time" scores for each item and then taking the square root of the product, resulting in a "demand" score ranging from 1 to 5. These scores were then summed for a total score (*possible range*: 24-120), with higher scores indicative of greater demand. In its use with pediatric cancer populations, the CMCCS has demonstrated high internal consistency (Cronbach's α = .93) and high test-retest reliability (r = .90 over a 3- to 7-day period; Wells et al., 2002; see Appendix C). Cronbach's alpha for the current sample was .94.

Parent Attitude Toward Child's Illness Scale (adapted from the Child Attitude Toward Illness Scale; Austin & Huberty, 1993). The Parent Attitude Toward Child's

Illness Scale is a 13-item self-report measure that assesses how positively or negatively parents of children diagnosed with a chronic illness feel about their child's chronic condition. For the present study, following the guidelines of the original authors of the CATIS, the measure was adapted to be specific to JRDs. Furthermore, the measure was adapted by the investigator to assess parental attitudes toward their child's illness. The content of the items on the adult version were based on the original child version but contained modified wording directed towards parent attitudes (e.g., "How good or bad do you feel it is that your child has a rheumatic illness?"), rather than child attitudes. Similar to the child version, the parent-report measure yielded a single composite score, whereby items were summed and then divided by 13 (*possible range*: 1-5). Higher scores indicated more favorable attitudes toward the child's chronic medical condition (see Appendix D). The parent-version of the CATIS had good internal reliability ($\alpha = .84$).

Brief Symptom Inventory (BSI; Derogatis, 1993). The BSI is a 53-item self-report measure that assesses adult global psychological adjustment. Respondents rated the degree to which psychological symptoms (e.g., poor appetite, difficulty making decisions, feelings of guilt) caused distress during the past seven days. Items were rated from 1 (*not at all*) to 4 (*extremely*), and then scores were summed and divided by the total number of items to obtain a Global Severity Index (GSI) which was used as the measure of parental distress (*possible range*: 0-4). The BSI has been found to have satisfactory internal consistency, ranging from .71 to .85 (Derogatis & Melisaratos, 1983). Additionally, the BSI has been used extensively as a measure of parental distress in studies examining parent contributions to child adaptation and outcome to chronic illness

(Mullins et al., 1995; Wagner et al., 2003; White et al., 2005; see Appendix E). Cronbach's alpha for the current sample was .97.

CHAPTER V

RESULTS

Overview of Analyses

First, the data set was examined for missing values, and if 5% or less of items were missing from any single measure, participant-specific mean values were imputed; however, if more than 5% of items were missing from any single measure, pairwise deletion was applied. In addition, the sample was examined for outliers (i.e., ≥ 3SDs from the mean; Wainer, 1976), and Global Severity Index (i.e., parental distress) scores that were more than 3SDs were examined for data entry errors, intentional misreporting, and legitimate outliers that were sampled from the correct population. Legitimate outliers (n = 2, < 1%) who represent the sample of the population at risk for increased adjustment difficulties were truncated to be within 3SDs of the mean. Through truncation, the relative ordering of the data were maintained, and the parents with greater distress had the highest GSI scores, yet the distributional problems were reduced (Orr, Sackett, & DuBois, 1991). Of note, approximately 20% (n = 14) of caregivers in the current study reported distress scores greater than the cut off score (GSI ≥ .67). This indicates that although the majority of parents fall within the normal range, a small subset appear to experience significant distress. Refer to Table 2 for a correlation matrix.

Theoretically-driven covariates were selected given their association with parental distress across pediatric chronic illness populations (Thompson & Gustafson, 1996). As

such, physician-rated functional disability (PRFD), household annual income, child-reported distress (Children's Depression Inventory total scores), and child age were entered as covariates in all subsequent analyses. Following the guidelines of Baron and Kenny (1986) and Holmbeck (1997), the conditions of mediation were tested using a series of hierarchical multiple regression analyses to determine whether parent attitudes toward their child's rheumatic disease mediated the relation between caregiver demand and parental distress. First, the direct relation between caregiver demand and parental distress was examined. Then, the relation between caregiver demand and parent attitudes toward their child's illness was analyzed. Third, the association between parent illness attitudes and parental distress was examined. Lastly, the relation between caregiver demand and parental distress was investigated while controlling for parent attitudes toward their child's illness.

An additional assumption of mediation is that no predictor x mediator interaction exists (MacKinnon, Fairchild, & Fritz, 2007), such that parent attitudes toward their child's illness moderates the relation between caregiver demand and parental distress. To test for a predictor x mediator interaction, caregiver demand and parent illness attitude scores were first centered by subtracting the sample mean from each score (Aiken & West, 1991). Next, the same four covariates were entered on the first step of the regression equation. Centered caregiver demand and parent illness attitude scores were entered on the second step, and the caregiver demand x parent illness attitudes score was entered on the third step of the equation.

If mediation was present, post-hoc probes of the indirect effect of parent attitudes toward their child's illness on the caregiver demand to parental distress association was

planned using a bootstrapping approach (Preacher & Hayes, 2004, 2008). Bootstrapping was selected because it allows for testing the indirect effect of a variable when conducting research with small to modest sample sizes, and it accounts for non-normality in the sampling distribution (Preacher & Hayes, 2004). The indirect effect was created using re-sampling with replacement to estimate 5,000 samples that were derived from the original sample (n = 69; Preacher & Hayes, 2004). Significance of the indirect effect was determined by examining the 95% confidence interval of the sampling distribution of the mean. The indirect effect was deemed significant at the .05 level if the confidence interval did not include zero.

More recent research has suggested that global or multidimensional measures of parental distress, such as the BSI (Derogatis, 1993), may be problematic as a result of inconsistent factor structures (Greenblatt & Landsberger, 2002) and the notion that high levels on general measures may be a result of other stressors related to caring for a child with a chronic illness rather than depression per se (Fisher et al., 2007). Although the BSI may contribute to the assessment of some distinct aspects of symptomotology in caregivers of children with a chronic illness, it may not adequately or solely measure general distress. Therefore, domain-specific indices of parental adjustment (i.e., depression) may be helpful in delineating specific features of parent adjustment that are most influential in determining child adjustment outcomes. As such, exploratory analyses included only the depression-specific items from the BSI to more closely examine the relation between caregiver demand and domain-specific distress versus general parental distress.

Primary Analyses

Regression analyses revealed a significant direct relation between caregiver demand and parental distress, after controlling for covariates (β = .32, $t(67) = 2.69$, p = .009), such that higher levels of caregiver demand were related to higher levels of parental distress. Next, a significant relation between caregiver demand and parent illness attitudes was found (β = -.55, $t(67) = -5.18$, $p < .001$), such that higher levels of caregiver demand were related to more negative parent attitudes toward their child's rheumatic disease (i.e., lower illness attitude scores). Additionally, regression analyses revealed a significant relation between parent attitudes toward their child's rheumatic disease and their levels of distress (β = -.41, $t(67) = -3.67$, $p < .001$), such that parents who reported more negative attitudes toward their child's illness endorsed higher levels of distress. Finally, after controlling for parent illness attitudes, caregiver demand was no longer a significant predictor of parental distress (β = .11, $t(67) = .81$, p = .42). Thus, parent attitudes toward their child's rheumatic disease mediated the relation between caregiver demand and parental distress. Post-hoc bootstrapping analyses revealed a significant indirect effect indicating that parent illness attitudes did, indeed, mediate the relation of caregiver demand and parental distress (95% CI = .003 to .014; see Figure 1).

Results of the hierarchical regression analyses revealed that the interaction of caregiver demand x parent illness attitudes did not contribute significant variance to the prediction of parental distress ($p > .05$). Thus, parent attitudes toward their child's rheumatic disease did not moderate the relation between caregiver demand and parental distress.

Exploratory Analyses

Using only the depression-specific items on the BSI, regression analyses revealed a significant relation between caregiver demand and domain-specific parental distress (β = .34, $t(67) = 2.72$, $p = .01$). Furthermore, there was a significant relation between parent illness attitudes and domain-specific distress (β = -.45, $t(67) = -3.91$, $p < .001$), such that more negative attitudes were associated with higher levels of depressive symptoms among parents. Lastly, after controlling for parent illness attitudes, caregiver demand was no longer significantly related to domain-specific (i.e., depression) parental distress (β = .10, $t(67) = .73$, $p = .47$), indicating that parent attitudes toward their child's illness mediated the relation of caregiver demand to depression-specific distress.

To better understand what component of parent illness attitudes may be directly related to increased parental distress, specific items of the Parent Attitude Toward Child Illness Scale were selected to examine the potential mediating effect of the caregiver demand-parent distress relation. Based on previous research indicating that parents of children with a chronic medical condition are often responsible for providing illness-related physical and emotional care (Raina et al., 2004; Wolfe-Christensen et al., 2010) and are therefore less likely to participate in positive and personally rewarding experiences (i.e., illness intrusiveness; Devins, Seland, Klein, Edworthy, & Saary, 1993), it stands to reason that items specific to feeling different from parents of healthy children might help explain general distress. As such, items 8 ("How often do you feel that your child's rheumatic illness keeps you from starting new things?"), 9 ("How often do you feel different from others because of your child's rheumatic illness?), and 13 ("How often

do you feel just as good as other parents even though your child has a rheumatic illness?) were selected for exploratory analyses.

Overall, analyses revealed that each of the aforementioned items individually mediated the relation between caregiver demand and parental distress in parents of youth with rheumatic disease. Specifically, regression analyses controlling for the same four covariates (i.e., child age, child distress, PRFD, and annual income) revealed a significant association between caregiver demand and "feeling that the child's rheumatic disease kept them from starting new things" (β = -.37, t(67) = -3.23, p = .002). Moreover, "feeling limited by the child's illness" was related to parental distress (β = -.36, t(67) = -3.17, p = .002). Finally, caregiver demand was no longer associated with levels of distress after controlling for the degree to which parents "felt their child's rheumatic disease kept them from doing things" (β = .19, t(67) = 1.56, p = .12).

With regard to item 9, parents who reported high levels of caregiver demand "felt different from other parents as a result of their child's illness" (β = -.45, t(67) = -4.26, p < .001). Further, "feeling different from other parents" was associated with increased parental distress (β = -.39, t(67) = -3.37, p = .001). Lastly, after controlling for parents who "felt different compared to other parents of healthy children," caregiver demand was not significantly associated with parental distress (β = .20, t(67) = 1.63, p = .11).

For the last item, regression analyses revealed a significant direct effect of caregiver demand on "feeling worse than other parents because of their child's rheumatic disease" (β = -.38, t(67) = -3.08, p = .003), in addition to an association between "not feeling as good as other parents" and levels of distress (β = -.36, t(67) = -3.44, p = .001). Caregiver demand was not significantly related to parental distress, however, once the

frequency of “feeling worse than other parents because of the child’s illness” was controlled for in the regression analysis (β = .21, $t(67) = 1.72$, p = .09). Together, these results indicate that compared to feelings of guilt or overall sadness regarding their child’s illness, parents who feel different from other parents or held back by their child’s rheumatic disease are more likely to experience increased parental distress.

CHAPTER VI

DISCUSSION

The present study was designed to examine the mediating effect of parent attitudes toward their child's JRD on the relationship between caregiver demand and parental distress. Three specific hypotheses were proposed: 1) caregiver demand would be positively related to parental distress; 2) parent illness attitudes and distress would be negatively related; and 3) parent attitudes toward their child's rheumatic disease would mediate the direct effect between caregiver demand and parental distress. In addition, exploratory analyses examined the mediating effect of parent illness attitudes on the relation between caregiver demand and domain-specific (i.e., depression) distress in parents, although no specific directional hypotheses were made. Moreover, illness attitudes specifically related to feeling different than parents of healthy children or hindered by their child's rheumatic disease (i.e., Items 8, 9, and 13) were examined separately as potential mediators of the caregiver demand-parental distress relation.

Consistent with hypotheses and existing literature (e.g., Barlow et al., 1998; Herbert & Cohen, 1993; Kuster & Merkle, 2004; Raina et al., 2004; Reisine, 1995), regression analyses revealed a significant direct effect of caregiver demand on parental distress, such that higher levels of demand were associated with increased distress. As hypothesized, results also revealed that parent attitudes toward their child's rheumatic disease were negatively associated with parental distress, such that more negative

evaluations of the illness (e.g., feeling the child's illness was unfair or that it kept the parent from doing things he/she wanted to do) were related to higher levels of distress. This result is consistent with the pediatric literature that has demonstrated the impact of negative illness attitudes on child adjustment difficulties, including increased depression and anxiety, decreased self-esteem, and academic problems, across illness groups (e.g., Heimlich et al., 2000; Iobst et al., 2007; LeBovidge et al., 2005). Lastly, parent illness attitudes mediated the caregiver demand-distress relation, after controlling for a large number of theoretically-driven demographic and disease variables. Thus, parents with negative perceptions or attitudes toward their child's rheumatic disease were at increased risk for experiencing general distress. This was further supported by the examination of depression-specific distress, which demonstrated that parent illness attitudes were also associated with depressive symptoms among caregivers. More importantly, parent attitudes, rather than the physical and emotional demands of caring for their child, explained higher levels of parental distress. Specific to parent illness attitudes, exploratory analyses revealed that feeling different than other parents or limited by their child's rheumatic disease were more likely to experience increased parental distress, compared to illness attitudes related to feelings of guilt or sadness.

The caregiver demand-parent cognitive appraisal relation seen in the present study is well documented in both adults and children with a chronic medical condition (e.g., Raina et al., 2004; Wolfe-Christensen et al., 2010). Specific to JRDs, parents often serve as primary caregivers and must quickly learn how to manage their child's illness following the diagnosis, a role accompanied by new responsibilities and considerable stress. Although parents can get into a routine and adapt to the new demands of managing

their child's illness, there is reason to suspect that the observed caregiver demand-illness attitude association is more likely to occur in parents of youth with JRDs, due in part to the chronic and unpredictable nature of the disease course (Cassidy et al., 2010). For parents of youth with a JRD, strict adherence to prescribed medical regimens may not protect the child from experiencing symptom flare-ups (Hommel, Chaney, Wagner, & Jarvis, 2006). Although speculative, these findings suggest that persistent behavior-outcome noncontingencies may lead caregivers to become increasingly frustrated, feel a loss of control about their ability to help their child cope with their rheumatic disease, and develop more negative attitudes toward their child's illness, ultimately increasing parental distress.

The current results help to explain the role of parent cognitive appraisals in the caregiver demand-distress relation in parents of youth with JRDs and the mixed results in the current literature. Although several studies have supported the notion that caring for a child with a chronic medical condition and being responsible for managing their illness on a daily basis puts parents at increased risk for both physical and psychosocial adjustment difficulties (e.g., Bauer et al., 2000; Brouwer et al., 2004; Moskowitz et al., 2007; Scanlan et al., 1998), others have found caregivers of children with a JRD to be quite resilient (Bruns et al., 2008; Daltroy et al., 1992; Harris et al., 1991). In fact, caregivers of children with JRDs have been shown to demonstrate adaptive skills similar to caregivers of healthy children (Harris et al., 1991), as well as comparable levels of anxiety and functional and social support (Gerhardt et al., 2003). Bruns and colleagues (2008) found that the type or degree of physical limitation associated with the child's idiopathic arthritis was not associated with the caregiver's quality of life; rather, they

suggested that the main impact of JRDs on caregiver distress is related to the emotional aspects or perceptions of the disease. Indeed, results of the current study suggest that it is the variability in stress appraisals, specifically how favorably or unfavorably a parent views their child's rheumatic disease, that is strongly related to levels of distress, and thus helps to explain why a subsample of caregivers are at risk for adjustment difficulties.

Limitations

The findings in the present study must be qualified by a couple of limitations. First, comorbid cognitive deficits and/or medical conditions resulted in a modest sample size. Additionally, generalization of these results are somewhat limited by a relatively homogenous sample of participants from similar sociocultural backgrounds. However, this concern was attenuated somewhat by the inclusion of a culturally heterogeneous sample. Nevertheless, caution should be exercised in generalizing the findings to other pediatric illness groups and more diverse groups of caregivers of children with JRDs.

Another limitation involves the use of self-report inventories (i.e., Caregiver Demand, Parent Attitude Toward Child Illness, and BSI) that may have resulted in spurious correlations due to shared method variance rather than actual associations between the target variables (e.g., Podsakoff, MacKenzie, Lee, & Podsakoff, 2003). However, it is likely that if the same source of variance was responsible for the observed relation between all three variables, regression results would have most likely shown that caregiver demand and parent illness attitudes would have exerted significant effects on parent distress. Because parent attitudes toward their child's rheumatic disease made a unique contribution to parent distress, independent of caregiver demand, it is unlikely that the observed associations were due to shared method variance.

Lastly, despite controlling for child age in regression analyses, the inclusion of youth in both pre-adolescent and adolescent phases of development should be noted as a limitation of the current study. Given that developmental issues are fairly different for these groups, such as increasing responsibility for managing one's rheumatic disease with age, it is possible that caregiver responsibilities, illness attitudes, and subsequent distress may be different at various stages of child development.

Implications and Recommendations for Future Research

The aforementioned limitations notwithstanding, the finding that parent illness attitudes mediates the relation between caregiver demand and parental distress has a number of treatment and research implications. Currently, there are no known studies in the extant literature that have addressed parent attitudes toward their child's illness and how it may impact parental distress. The current study addresses a gap in the pediatric chronic illness literature, and the findings suggest the importance of parent-focused interventions aimed at altering negative perceptions held by caregivers (e.g., Kazak et al. 2005; Sahler et al., 2005; Streisand, Rodrigue, Houck, Graham-Pole, & Berlant, 2000). Given recent findings that parent distress temporally precedes child distress in JRDs (Ryan et al., 2010), parent-focused and family systems interventions have the potential to prevent further parent distress and subsequent child adjustment difficulties. Moreover, parent illness attitudes, when defined as evaluative judgments and their associated feelings, are amenable to change. Therefore, while other illness-related variables (e.g., pain and fatigue) and caregiver demands may be quite difficult to alter, the ability to target attitude change has far-reaching implications for both the child and family. Notably, results from exploratory analyses support the need for addressing specific

aspects of illness attitudes, particularly feelings of isolation and detachment from other parents, in order to help parent's problem-solve ways they can be involved in personally gratifying activities and simultaneously care for their ill child.

Furthermore, youth with a rheumatic disease are typically the primary focus of assessment and intervention. Yet, results of the present study underscore the importance of acknowledging and screening for parent adjustment concerns when addressing child adjustment. This can be done several ways, such having parents complete a brief measure of psychological adjustment (e.g., Beck Depression Inventory or Brief Symptom Inventory) in the waiting room when they bring their child for routine medical appointments or encouraging physicians to check-in with parents and ask how they are adjusting to the diagnosis and newly acquired caretaking responsibilities. Open lines of communication between the medical team and family and validation of the stress that accompanies caring for a child with a chronic medical condition may help foster a supportive relationship and result in positive adjustment outcomes (e.g., increased adherence, less parental distress, and decreased parent-child conflict) for both the child and parent.

Future research should examine the role of parent illness attitudes in the caregiver demand-distress association over time in order to investigate the temporal precedence of negative illness attitudes relative to parental distress and other adjustment difficulties (e.g., marital conflict, isolation, parent-child conflict). This would also allow examination of the association between caregiver demands and distress over the course of an unpredictable illness. Moreover, future studies need to continue examining the relation between parent and child illness attitudes, including age and sex differences, to better

elucidate the cognitive appraisal mechanisms underlying the transactional parent-child distress association in this population.

In general, results of the present study suggest that parent attitudes toward their child's illness play an important role in the association between the demands and responsibilities of caring for a child with a rheumatic disease and parental distress. These findings underscore the importance of acknowledging and regularly addressing the concerns of parents caring for a child with a chronic medical condition in an attempt to prevent distress and adjustment difficulties for the entire family.

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Table 1

Participant Demographics

	Observed Range	*M (SD)*
Child Age (in years)	7 – 18	13.20 (2.88)
Child Gender (% female)	69.6%	
Maternal Age (in years)	29 – 55	39.45 (6.40)
Paternal Age (in years)	29 – 60	41.84 (7.45)
Mothers to complete packets	87.0%	
Married parents	50.7%	
Ethnicity (child)		
Caucasian	67.2%	
Native American	16.4%	
African American	4.5%	
Hispanic	4.5%	
Asian	3.0%	
Other	4.5%	
Annual Family Income		
< $20,000	17.4%	
$20,000 - $40,000	29.0%	
$40,000 - $60,000	15.9%	
> $60,000	37.5%	
Child Diagnosis		
Juvenile Idiopathic Arthritis	67.2%	
Juvenile Dermatomyositis	10.9%	
Lupus	6.2%	
Spondylarthropathy	4.7%	
Other rheumatic disease	10.9%	
Physician-Rated Functional Disability	1 – 4	1.33 (.63)
Parent Attitude Toward Child's Illness Scale	1 – 4	2.88 (.57)
Caregiver Demand	25 – 90	48.08 (13.27)
Brief Symptom Inventory (Global Severity Index)	0 – 1.8	.40 (.42)
Children's Depression Inventory	0 – 28	7.30 (6.66)

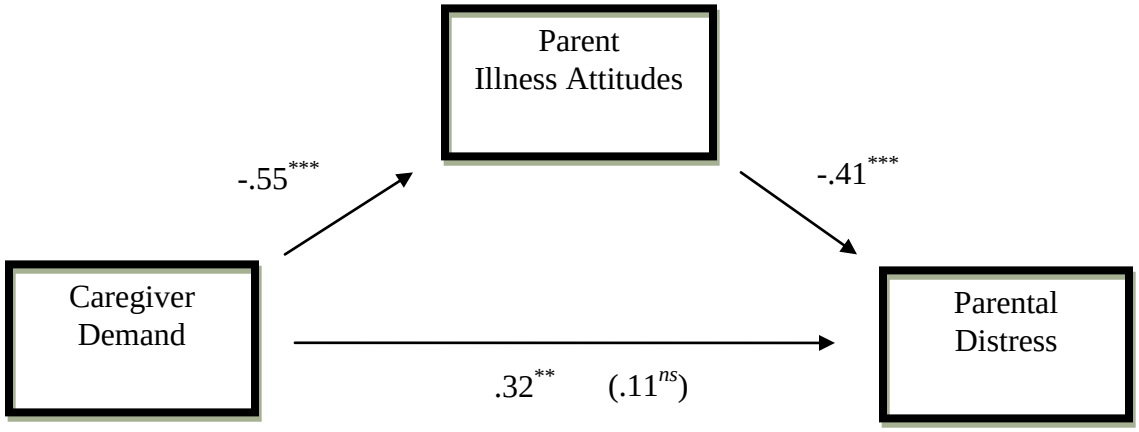
Table 2

Zero-order correlations for demographic variables, disease parameters, and outcome variables

Variable	*1*	*2*	*3*	*4*	*5*	*6*	*7*	*8*	*9*	*10*
1. Child gender		.15	.05	.01	.04	.03	.09	.11	-.09	.10
2. Child age			.19	.02	.12	.08	.01	-.03	-.02	-.03
3. Parent marital status				-.14	.10	-.02	.07	.02	-.03	.15
4. Annual income					.14	-.02	-.02	-.02	.07	-.21
5. Diagnosis						.16	-.03	.09	-.06	.18
6. Functional disability							.35**	.38**	-.32**	.37**
7. Child distress								.41***	-.44***	.36**
8. Caregiver demand									-.66***	.46***
9. Parent illness attitudes										-.52***
10. Parental distress										

*$p < .05$, **$p < .01$, ***$p < .001$.

Figure 1. Parent attitude toward their child's rheumatic disease as a mediator between caregiver demand and parental distress. Note. Values on paths are path coefficients (standardized betas). The path coefficient in parentheses is standardized partial regression coefficient controlling for parent illness attitudes. ** p < .01 *** *p* < .001.



APPENDIX A

Background Information Questionnaire

Background Information Questionnaire

Hometown: _____

1. Child's Age: _____ Parent's Age: Father: _____ Mother: _____

2. Child's Gender: M F Parent's Gender: M F

3. Child's Ethnicity:

Caucasian	African American	Hispanic	Native American	Asian	Other
1	2	3	4	5	6

4. Father's Ethnicity:

Caucasian	African American	Hispanic	Native American	Asian	Other
1	2	3	4	5	6

5. Mother's Ethnicity:

Caucasian	African American	Hispanic	Native American	Asian	Other
1	2	3	4	5	6

6. Child's current or highest grade completed:
   1 Elementary
   2 Middle School
   3 High School
   4 Some college: Years:___

7. Marital Status:
   1 Never married
   2 Married
   3 Divorced
   4 Cohabitation (living with partner)
   5 Widowed
   6 Other:_____

8. Parent's highest level of education:

   Father:
   1 Middle School
   2 High School
   3 Some College: Years:_____
   4 College Degree
   5 Post-Graduate Degree

Mother:	1	Middle School
	2	High School
	3	Some College: Years:_____
	4	College Degree
	5	Post-Graduate Degree

7. Parent's Occupation: Father:_____ Mother:_____

8. Living Arrangement:	1	Live alone
	2	Live with both parents
	3	Live with one parent; Specify parent:_____
	4	Other; Specify:_____

9. Income: What is the total yearly income of the primary wage earner in your house? (*This will be held strictly confidential*.)

_____ 0 – 9,999	_____ 50,000 – 59,999
_____ 10,000-19,999	_____ 60,000 – 69,999
_____ 20,000-29,999	_____ 70,000 – 79,999
_____ 30,000- 39,999	_____80,000 – 89,999
_____ 40,000 – 49,999	_____ 90,000 – 99,999
	_____ 100,000 or greater

10. Is your child currently taking any psychoactive medication (e.g., antidepressants, anti-anxiety)?

Yes No

11. Has your child ever received any type of psychological counseling/therapy?

Yes No

12. Has your child ever received counseling directly related to Juvenile Rheumatic Disease (JRD) (includes juvenile rheumatoid arthritis (JRA), systemic lupus erythematosus (SLE), juvenile spolndylarthropathies, juvenile dermatomyositis (JDMA)

Yes No

APPENDIX B

Provider Questionnaire

Provider Questionnaire

1. Patient's name:_____

2. Patient's diagnosis (if multiple diagnoses, please list the rheumatic illness first; please indicate if patient is seropositive or ANA-positive)

   _____
   _____
   _____

3. When was the patient diagnosed with the above rheumatic illness?

   Date of diagnosis: _____

4. What is the patient's current medication regimen?

   _____ _____
   _____ _____
   _____ _____

5. Currently, how active is the patient's illness?

1	2	3	4
Clinical Remission (off meds 12 mo)	Clinical Remission (on medication)	Inactive Disease	Active Disease

6. Currently, how severe is the patient's illness?

1	2	3	4	5
Inactive		Mild		Severe

7. Compared to other patients, how well does this patient adhere to his/her treatment regimen?

1	2	3	4	5	6	7
Adheres Very poorly		Worse than most patients		Better than most patients		Adheres Extremely Well

8. Compared to other patients, how well does this patient cope with his/her illness?

1	2	3	4	5	6	7
Copes Very poorly		Worse than most patients		Better than most patients		Copes Extremely Well

Based on the patient's physical exam, please classify him/her into one of the following four classes:

Class I	**Class II**	**Class III**	**Class IV**
Completely able to perform usual activities of daily living (self-care, vocational, and avocational)	Able to perform usual self-care and vocational activities, but limited in avocational activities	Able to perform usual self-care activities, but limited in vocational and avocational activities	Limited ability to perform usual self-care, vocational and avocational activities

APPENDIX C

Caring for My Child with a Rheumatic Disease

Caring for My Child with a Rheumatic Disease

Parents/Guardians put time and effort into taking care of their child with a Juvenile Rheumatic Disease. We want to better understand how much time and effort certain care-giving tasks require. Please indicate below the amount of time and the amount of effort during a typical week that these tasks have required of you.

I. Physical Care

1. Preparing and giving medication (This includes: Administering Methotrexate, Administering Corticosteroids, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

2. Managing physical side effects of the Juvenile Rheumatic Disease or its treatment (Examples include: Difficulty writing, Decreased mobility, Vision problems, Fatigue, Helping with writing skills).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

3. Coordinating, arranging, and managing medical services (Examples include: Scheduling appointments, Locating equipment, Negotiating services, Maintaining splints/braces, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

4. Time spent in transit or at the hospital and doctor's office for appointments (Examples include: Ophthalmologist, Other Tests).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

II. Emotional Care

5. Providing emotional support for your child with a Juvenile Rheumatic Disease.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

6. Providing emotional support for other children in the family.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

7. Providing emotional support for other family members (Examples include: Grandparents, Aunts, Uncles, Friends, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

8. Providing emotional support for your spouse/partner.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

9. Meeting your own emotional support needs.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

10. Comforting your child physically through the pain of his or her Juvenile Rheumatic Disease and its treatment (Examples include: Procedures, Arthritis flares, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

III. Family/Interpersonal Relationships

11. Planning activities for your child with a juvenile rheumatic disease around the treatment and illness (Examples include: Less physically demanding after school activities, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

12. Getting child care/babysitting help for your ill child.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

13. Obtaining child care/babysitting for the brother/sisters of your ill child.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

IV. Communication

14. Discussing each parent/caregiver's feelings about the strain of caring for a child with a Juvenile Rheumatic Disease (Examples include: Pressure to maintain medication/exercise regimen, Worry about injury, Amount of needed social support, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot

_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

15. Communicating information about the Juvenile Rheumatic Disease to schools, day care, babysitters, extended family and friends (Examples include: Limitation on activities, Absenteeism, Strategies for remembering medication, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

16. Getting information on your child's illness and the treatment (Examples include: Library, Medical Team, Community Agencies).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

In this section, we want to better understand certain tasks related to the Juvenile Rheumatic Disease and the relationship with your spouse/partner. Please indicate below the amount of time and the amount of effort during a typical week that these tasks have required of you both before and after the onset of the Juvenile Rheumatic Disease.

V. Changes since the onset of the Juvenile Rheumatic Disease

17. Discussing/taking care of finances and bills with your spouse/partner, including finances and bills related to your child's illness.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

18. Discussing/taking care of finances and bills with your spouse/partner **before** the onset of the Juvenile Rheumatic Disease.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

19. Doing activities with your spouse/partner independent of your ill child or the siblings of your ill child (Examples include: Time together out of the house, one-on-one time at home, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

20. Doing activities with your spouse/partner independent of your child **before** the onset of the Juvenile Rheumatic Disease.

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Did Not Do	_____None

21. Discussing parenting issues with your partner/spouse about your child with a juvenile rheumatic disease. (Examples include: Protectiveness regarding activity level, Managing treatment plan, Encouraging adherence to recommended exercises, other general parenting issues).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

22. Discussing parenting issues for this specific child with your spouse/partner **before** the onset of the Juvenile Rheumatic Disease (Examples include: Protectiveness regarding activity level, Managing treatment plan, Encouraging adherence to recommended exercises, other general parenting issues).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Did Not Do	_____None

23. Communicating about your relationship with your spouse/partner, including the demands of your child's illness (Examples include: Stress, Time spent together, Personal time, Financial strains, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

24. Communicating about your relationship with your spouse/partner **before** the onset of the Juvenile Rheumatic Disease (Examples include: Stress, Time spent together, Personal time, Financial strains, Other).

Time	**Effort/Difficulty**
_____ > 5 Hours/Week	_____ A Great Deal
_____ > 2 – 5 Hours/Week	_____ Quite A Lot
_____ 1 -2 Hours/Week	_____ Moderate
_____ < 1 Hour/Week	_____ A Small Amount
_____ Do Not Do	_____None

APPENDIX D

Parent Attitude Toward Illness Scale

Parent Attitude Toward Illness Scale
(adapted from CATIS, Austin & Huberty, 1993)

Instructions: Please read each statement below and indicate how the statement describes your attitude by circling one of the responses that follow.

1. How good or bad do you feel it is that your child has a rheumatic illness?

   Very good A little good Not sure A little bad Very bad

2. How fair is it that your child has a rheumatic illness?

   Very fair A little fair Not sure A little unfair Very unfair

3. How happy or sad is it for you that your child has a rheumatic illness?

   Very sad A little sad Not sure A little happy Very happy

4. How bad or good do you feel it is that your child has a rheumatic illness?

   Very good A little good Not sure A little bad Very bad

5. How often do you feel your child's rheumatic illness is your fault?

   Never Not often Sometimes Often Very often

6. How often do you feel that your child's rheumatic illness keeps you from doing things you like?

   Very often Often Sometimes Not often Never

7. How often do you feel that your child will always be sick?

   Never Not often Sometimes Often Very often

8. How often do you feel that your child's rheumatic illness keeps you from starting new things?

   Very often Often Sometimes Not often Never

9. How often do you feel different from others because of your child's rheumatic illness?

   Never Not often Sometimes Often Very often

10. How often do you feel bad for your child because he/she has a rheumatic illness?

Very often Often Sometimes Not often Never

11. How often do you feel sad about your child being sick?

    Never Not often Sometimes Often Very often

12. How often do you feel happy even though your child has a rheumatic illness?

    Never Not often Sometimes Often Very often

13. How often do you feel just as good as other parents even though your child has a rheumatic illness?

    Very often Often Sometimes Not often Never

APPENDIX E

Brief Symptom Inventory

Brief Symptom Inventory (BSI)

INSTRUCTIONS:

On the next page is a list of problems people sometimes have. Please read each one carefully, and blacken the circle that best describes HOW MUCH THAT PROBLEM HAS DISTRESSED OR BOTHERED YOU DURING THE PAST 7 DAYS INCLUDING TODAY. Blacken the circle for only one number for each problem and do not skip any items. If you change your mind, erase your first mark carefully. Read the example before beginning, and if you have any questions please ask them now.

Not at all	A little bit	Moderately	Quite a bit	Extremely	Example How much were you distressed by:
0	1	2	3	4	Bodyaches

Not at all	A little bit	Moderately	Quite a bit	Extremely	How much were you distressed by:
0	1	2	3	4	Nervousness or shakiness inside
0	1	2	3	4	Faintness or dizziness
0	1	2	3	4	The idea that someone else can control your thoughts
0	1	2	3	4	Feeling others are to blame for most of your troubles
0	1	2	3	4	Trouble remembering things
0	1	2	3	4	Feelings easily annoyed or irritated
0	1	2	3	4	Pains in heart or chest
0	1	2	3	4	Feeling afraid in open spaces or on the streets
0	1	2	3	4	Thoughts of ending your life
0	1	2	3	4	Feeling that most people cannot be trusted
0	1	2	3	4	Poor appetite
0	1	2	3	4	Suddenly scared for no reason
0	1	2	3	4	Temper outbursts that you could not control
0	1	2	3	4	Feeling lonely even when you are with people
0	1	2	3	4	Feeling blocked in getting things done
0	1	2	3	4	Feeling lonely
0	1	2	3	4	Feeling blue
0	1	2	3	4	Feeling no interest in things
0	1	2	3	4	Feeling fearful
0	1	2	3	4	Your feelings being easily hurt
0	1	2	3	4	Feeling that people are unfriendly or dislike you
0	1	2	3	4	Feeling inferior to others
0	1	2	3	4	Nausea or upset stomach
0	1	2	3	4	Feeling that you are watched or talked about by others
0	1	2	3	4	Trouble falling asleep
0	1	2	3	4	Having to check and double-check what you do
0	1	2	3	4	Difficult making decisions
0	1	2	3	4	Feeling afraid to travel on buses, subways, or trains
0	1	2	3	4	Trouble getting your breath
0	1	2	3	4	Hot or cold spells
0	1	2	3	4	Having to avoid certain things, places, or activities because they frighten you

Not at all	A little bit	Moderately	Quite a bit	Extremely	How much were you distressed by
0	1	2	3	4	Your mind going blank
0	1	2	3	4	Numbness or tingling in parts of your body
0	1	2	3	4	The idea that you should be punished for you sins
0	1	2	3	4	Feeling hopeless about the future
0	1	2	3	4	Trouble concentrating
0	1	2	3	4	Feeling weak in parts of your body
0	1	2	3	4	Feeling tense or keyed up
0	1	2	3	4	Thoughts of death or dying
0	1	2	3	4	Having urges to beat, injure, or harm someone
0	1	2	3	4	Having urges to break or smash things
0	1	2	3	4	Feeling very self-conscious with others
0	1	2	3	4	Feeling uneasy in crowds, such as shopping or at a movie
0	1	2	3	4	Never feeling close to another person
0	1	2	3	4	Spells of terror or panic
0	1	2	3	4	Getting into frequent arguments
0	1	2	3	4	Feeling nervous when you are left alone
0	1	2	3	4	Others not giving you proper credit for your achievements
0	1	2	3	4	Feeling so restless you couldn’t sit still
0	1	2	3	4	Feelings of worthlessness
0	1	2	3	4	Feeling that people will take advantage of you if you let them
0	1	2	3	4	Feelings of guilt
0	1	2	3	4	The idea that something is wrong with your mind

APPENDIX F

IRB Approval Form

Oklahoma State University
Institutional Review Board

Protocol Expires: 2/10/2005

Date: Thursday, February 12, 2004 IRB Application No AS00104

Proposal Title: PSYCHOLOGICAL COMORBIDITY IN JUVENILE RHEUMATOID DISEASES: A COMPARISON OF AMERICAN INDIANS AND CAUCASIANS.

Principal Investigator(s):

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Reviewed and Processed as: Expedited (Spec Pop)

Approval Status Recommended by Reviewer(s): Approved

Dear PI :

Your IRB application referenced above has been approved for one calendar year. Please make note of the expiration date indicated above. It is the judgment of the reviewers that the rights and welfare of individuals who may be asked to participate in this study will be respected, and that the research will be conducted in a manner consistent with the IRB requirements as outlined in section 45 CFR 46.

As Principal Investigator, it is your responsibility to do the following:

1. Conduct this study exactly as it has been approved. Any modifications to the research protocol must be submitted with the appropriate signatures for IRB approval.
2. Submit a request for continuation if the study extends beyond the approval period of one calendar year. This continuation must receive IRB review and approval before the research can continue.
3. Report any adverse events to the IRB Chair promptly. Adverse events are those which are unanticipated and impact the subjects during the course of this research; and
4. Notify the IRB office in writing when your research project is complete.

Please note that approved projects are subject to monitoring by the IRB. If you have questions about the IRB procedures or need any assistance from the Board, please contact me in 415 Whitehurst (phone: 405-744-5700, colson@okstate.edu).

Sincerely,

Carol Olson

Carol Olson, Chair
Institutional Review Board

VITA

Jamie Lynn Ryan

Candidate for the Degree of

Doctor of Philosophy

Dissertation: THE RELATIONSHIP OF CAREGIVER DEMAND AND PARENT DISTRESS IN JUVENILE RHEUMATIC DISEASE: THE MEDIATING EFFECT OF PARENT ATTITUDE TOWARD ILLNESS

Major Field: Clinical Psychology

Biographical:

Education:

Completed the requirements for the Master of Science in Clinical Psychology at Oklahoma State University, Stillwater, Oklahoma in July, 2010.

Completed the requirements for the Master of Arts in Clinical Psychology at the University of Northern Iowa, Cedar Falls, Iowa in May, 2008.

Completed the requirements for the Bachelor of Arts in Psychology and Criminal Justice at Mount Mercy College, Cedar Rapids, Iowa in May, 2005.

Experience:

Clinical Neuropsychological Clinic, Practicum Student
Solid Organ Transplant Clinic, Practicum Student
Psychological Services Center, Associate
American Indians Into Psychology, Student Representative
Graduate Psychology Admissions, Student Representative
Graduate Cognitive Assessment, Graduate Teaching Assistant
Abnormal Psychology, Graduate Teaching Assistant/Discussion Leader
Clinical Psychology Advisory Committee, 2nd Year Representative
Psychology Graduate Student Association, Treasurer
Psychology Diversified Students Program, Mentor
Camp Cavett, Voluntary Camp Counselor

Professional Memberships:

American Psychological Association, Division 54
American Psychological Association, Division 37
Oklahoma Psychological Association

Name: Jamie Lynn Ryan Date of Degree: July, 2013

Institution: Oklahoma State University Location: Stillwater, Oklahoma

Title of Study: THE RELATIONSHIP OF CAREGIVER DEMAND AND PARENT DISTRESS IN JUVENILE RHEUMATIC DISEASE: THE MEDIATING EFFECT OF PARENT ATTITUDE TOWARD ILLNESS

Pages in Study: 91 Candidate for the Degree of Doctor of Philosophy

Major Field: Clinical Psychology

Scope and Method of Study: The current study examined the association between parent illness attitudes, caregiver demand, and parental distress in a sample of parents of youth with Juvenile Rheumatic Diseases (JRDs, $n = 69$). Specifically, the study examined the potential mediating effect of parent illness attitudes on the caregiver demand to parental distress relation. Participants were recruited from the pediatric rheumatology clinic at a large teaching hospital in the Midwest. Parents completed the Caring for My Child with a Rheumatic Disease Scale, Parent Attitudes Toward Child's Illness Scale, and the Brief Symptom Inventory. The pediatric rheumatologist completed a measure of functional ability.

Findings and Conclusions: Consistent with hypotheses, higher caregiver demand and negative illness attitudes was associated with increased parental distress. Additionally, parent illness attitudes mediated this relation, such caregivers with more negative evaluations of their child's rheumatic disease are at increased risk for adjustment difficulties. Finally, exploratory analyses revealed that caregiver demand and parent illness attitudes were associated with both general and domain-specific indices of parent distress. Specific to illness attitudes, exploratory analyses revealed that parents who feel different compared to other parents of health children or feel limited in participating in activities as a result of their child's illness are more likely to experience increased parental distress, compared to feelings of guilt of sadness. The current study is limited by sample size and inclusion of youth in pre-adolescent and adolescent stages of development. However, results of the current study highlight the importance of addressing and screening for parent adjustment difficulties during their child's medical visits. Furthermore, family systems and parent-focused interventions aimed at altering negative illness attitudes may be beneficial in preventing adjustment difficulties for the entire family.

ADVISER'S APPROVAL: Larry L. Mullins, Ph.D.