

CHILDREN'S AGE AS A POTENTIAL MODERATOR
OF THE ILLNESS INTRUSIVENESS-DISTRESS
RELATIONSHIP IN JUVENILE
RHEUMATIC DISEASE

By

NICOLE RENEE ANDREWS

Bachelor of Science in Psychology
University of Oklahoma
Norman, Oklahoma
2002

Master of Science
Oklahoma State University
Stillwater, Oklahoma
2005

Submitted to the Faculty of the
Graduate College of the
Oklahoma State University
in partial fulfillment of
the requirements for
the Degree of
Doctor of Philosophy

CHILDREN'S AGE AS A POTENTIAL MODERATOR
OF THE ILLNESS INTRUSIVENESS-DISTRESS
RELATIONSHIP IN JUVENILE
RHEUMATIC DISEASE

Dissertation Approved:

Dr. John M. Chaney

Dissertation Adviser

Dr. Larry Mullins

Dr. Melanie Page

Dr. Laura Hubbs-Tait

Dr. A. Gordon Emslie

Dean of the Graduate College

ACKNOWLEDGEMENTS

I would like to thank my major adviser, Dr. John Chaney, for his expertise and mentorship throughout my time at Oklahoma State University. He allowed me a considerable measure of independence on this project, which has been an invaluable professional learning experience.

I would also like to express my gratitude to my other committee members, Drs. Larry Mullins, Melanie Page, and Laura Hubbs-Tait. Their comments and suggestions have been extremely helpful, both with respect to this project, and to my graduate career in general. In addition, I would like to express my appreciation to Dr. James Jarvis, who provided our lab with access to his rheumatology clinic. Further, I would like to thank my fellow lab members, especially Dr. Janelle Wagner, for their suggestions regarding the formulation of this project and for assistance with data collection.

Finally, I would like to thank my family for their support. My husband, Tim, has been encouraging throughout this process, as have my parents, Wayne and Laura Felts. Their faith in me has truly been invaluable, and I will always be thankful for their patience and love.

TABLE OF CONTENTS

Chapter	Page
I. INTRODUCTION	1
II. REVIEW OF THE LITERATURE	8
Medical and Clinical Issues in JRD	8
Issues Associated with Diagnosis	8
Disease Subtypes	9
Long-term Prognosis	12
Issues Associated With Treatment	13
Socio-emotional Issues in Pediatric Chronic Illness	13
Parent Adjustment	13
Factors Affecting Parent Adjustment	17
Cognitive Appraisals	17
Illness Intrusiveness	18
Development and Age of the Child	21
Rolland's Family Systems Model	28
Summary	29
III. THE PRESENT STUDY	31
Primary Hypotheses	33
Hypothesis 1	33
Hypothesis 2	33
IV. METHODS	34
Participants and Procedures	34
Instruments	35
Parent and Child-Report	35
Physician-Report	37
V. RESULTS	38
Preliminary Analyses and Selection of Covariates	38
Primary Analyses	38

Chapter	Page
Hypothesis 1	38
Hypothesis 2	39
Post-hoc Probes	39
VI. DISCUSSION	41
Strengths and Limitations	45
Future Studies	48
Clinical Implications	49
REFERENCES	51
APPENDICES	63
APPENDIX A—BRIEF SYMPTOM INVENTORY	64
APPENDIX B—ILLNESS INTRUSIVENESS SCALE-PARENT.....	65
APPENDIX C—JUVENILE ARTHRITIS FUNCTIONAL ASSESSMENT REPORT-CHILD FORM	66
APPENDIX D—CHILD DEPRESSION INVENTORY	67
APPENDIX E—PROVIDER QUESTIONNAIRE	72
APPENDIX F—IRB APPROVAL FORM	73
APPENDIX G—INFORMED CONSENT FORM	74

LIST OF TABLES

Table

1. Disease, Demographic, and Psychosocial Variables: Means	75
2. Hierarchical Regression Analyses of Parent-Reported Illness Intrusiveness on GSI.....	76
3. Hierarchical Regression Analyses of Parent-Reported Illness Intrusiveness on GSI as Moderated by Child Age	77

CHAPTER 1

INTRODUCTION

Juvenile rheumatic diseases (JRD) are one of the more common chronic illnesses of childhood. Currently, 65,000-70,000 children are affected by juvenile rheumatoid arthritis (JRA; Lovell, 1997); that diagnosis alone affects as many children as does juvenile diabetes (Cassidy & Petty, 2001). Within this group, there are three subtypes reflecting the level of joint involvement—pauciarticular, polyarticular, and systemic. Among the general population, the pauciarticular subtype is most prevalent (Lovell, 1997). However, JRA comprises only around half of JRD cases; systemic lupus erythematosus (SLE), juvenile dermatomyositis (JDM), and juvenile spondyloarthropathies (JSA) represent the majority of the remaining, though less common, JRD diagnoses. Although differences in presentation exist between the various diagnoses, they are characterized by a variety of similar symptoms, including connective tissue inflammation, pain, restricted ability, and a chronic course (Vandvik & Hoyeraal, 1993).

Across the diagnoses encompassed by JRD, psychological maladjustment among children has been researched extensively, perhaps due to the highly variable and pervasive nature of these diseases and their treatment. Although considerable research has focused on child adjustment to chronic illness, parental adjustment is equally important. Thompson et al. (1993a, b) have highlighted the complex interplay between parental distress and child maladjustment in pediatric chronically ill populations. In

addition to affecting children's psychological adjustment, parent distress can lead to poorer physical outcomes indirectly by influencing treatment compliance (Chaney & Peterson, 1989). Indeed, parent distress appears to be a more important determinant of child maladjustment and future outcomes than disease factors (Chaney et al., 1997; Frank et al., 1998). Similarly, disease severity is less important in predicting parent distress than subjective ratings of illness by the parent. These subjective ratings are particularly salient when they are tied to parents' perceptions of their role as care provider (Walker, Ford, & Donald, 1987).

Parental adjustment is not only deeply tied to child outcomes, but clearly constitutes an important area of investigation apart from child adjustment issues. For example, nearly two-thirds of parents of children with JRD report moderate to severe familial difficulties such as conflict and a lack of sufficient support (Vandvik, Hoyerhaal, & Fagertun, 1989). Similar results from longitudinal studies (i.e., Kupst & Schulman, 1988; Wallander et al., 1989), have indicated that disease variables are significantly poorer predictors of parental difficulties than level of social support, marital difficulties, and other social environment variables. These effects appear to be more pronounced in mothers than fathers (Nagy & Ungerer, 1990). Other studies (e.g., Dahlquist et al., 1993), however, found elevated symptoms of anxiety for both mothers and fathers. Regardless, parents of chronically ill children generally report more emotional distress than parents of healthy children.

Perhaps the most compelling evidence demonstrating the importance of examining parent adjustment in pediatric chronic illness is provided by Epel and colleagues (2004). These investigators found that mothers of chronically children

evidenced greater perceived stress compared to an age-matched comparison group of caregivers. More importantly, both increased caregiver stress and chronicity of their reported stress were associated with multiple biological indices of cellular aging - all of which have been linked to age-related physical illnesses such as cardiovascular disease and immune system dysregulation. These data suggest that the prolonged stress of having a chronically ill child not only has untoward effects on parents' subjective psychological well being, but also places parents at increased risk for adverse health sequelae.

Much like child adjustment to illness, parent adjustment is multi-determined by a host of variables, including cognitive appraisal processes. Except for a few studies, however, cognitive appraisals have been largely overlooked in investigations of parent adjustment to childhood chronic illness, with a few notable exceptions (e.g., Lustig, Ireys, Sills, & Walsh, 1996). Because of the significant level of physical impairment and lifestyle disruptions associated with JRD, one cognitive appraisal variable that appears particularly relevant to parent adjustment in JRD is *illness intrusiveness*. Briefly, illness intrusiveness is conceptualized as a generalized cognitive schema representing perceived "illness-induced barriers" across a wide array of life domains (Devins et al., 1983-84; p. 329). Illness intrusiveness is related to perceived disability, but it is considered conceptually distinct. To illustrate, perceived disability as it is traditionally assessed in the rheumatic diseases represents the degree to which routine activities of daily living such as walking, dressing, etc., are restricted. Illness intrusiveness captures the extent to which perceived illness restrictions (including but not limited to disability) preclude involvement in and/or access to disease-unrelated activities (e.g., family activities, social

relationships, leisure activities). Illness intrusiveness theory suggests that the resulting decrease in rewarding activities is largely responsible for adjustment difficulties.

Because JRD often involve a significant decrease in children's activity levels (Henderson et al., 1995), the role of parents' perceptions of interference with routine activities due to illness may take on particular importance. Although there are no known studies documenting the relationship between parents' perceived intrusiveness of their child's illness and parent adjustment, there is evidence in the adult and child rheumatic literature suggesting the salience of illness intrusiveness in the adjustment process.

High levels of child-reported illness intrusiveness have been shown to magnify the effect of parent distress on child depression (Wagner et al., 2003). Extant literature has only begun to examine the extent to which parent global distress is influenced by parent perceptions of the intrusiveness of their child's illness. Andrews and colleagues (2007) found that high parent-reported illness intrusiveness resulted in increased parental distress. This relationship was moderated by race, suggesting that parents already facing stressors unrelated to illness may be more vulnerable to the additional stress imposed by illness-induced barriers.

Another factor related to the impact of illness intrusiveness involves developmental expectations. This issue has been highlighted by Wallander and colleagues (2003), who clearly state that pediatric research should be conducted as a study of the "effects of challenging circumstances on child development" (p. 153). Importantly, they also note that developmental issues not only affect the individual diagnosed with a chronic illness, but affect the family and its members as well. For example, research on adults with rheumatoid arthritis (RA) indicates that the extent to

which individuals perceive their own illness as interfering with disease-unrelated activities is associated with significant increases in depressive symptoms and decreased overall quality of life (Devins et al., 1992). Importantly, these associations were more relevant for younger individuals with RA, which may indicate that the effects of lifestyle disruptions due to illness may be more pronounced when they are inconsistent with normative expectations regarding reductions in activity level across multiple life domains. This would certainly have implications for youth with JRD and their parents, given increased autonomy and self-reliance normally expected of children and adolescents that accompany increases in age.

Although childhood illness is best conceptualized in a family systems framework (Kazak & Wilcox, 1984), studies in this area are rare. However, Eisner and Berrenberg (1995) do note that arthritis is more limiting for adolescents because they are “forced into extended dependency” on their parents (p. 112) by their decreased mobility. Moreover, parents may be more distressed about the restrictions of their child’s illness than the children themselves. Eisner and Berrenberg (1995) note that this heightened parental awareness and distress may be because their parents’ “wider experiences” make them more aware of what constitutes typical development and abilities (p. 110).

Normative parental expectations of independence in activities of daily living vary across childhood and adolescence, with children assuming greater autonomy as they grow older. The impact of this developmental period is heightened among children diagnosed with chronic illnesses, as parental expectations may be thwarted by poorer disease outcomes (e.g., due in part to declines in compliance) among adolescents (Quittner et al., 2000; Williams, Holmbeck, & Greenley, 2002). This creates a gap between

developmental and environmental needs (Holmbeck & Kendall, 2002), which can lead to increases in illness related and unrelated stressors among both parents and their children. Given the challenges that can arise when a child's abilities and parental expectations do not vary together under ideal conditions (Holmbeck, 2002a), the addition of a significant complicating factor such as a chronic illness only serves to magnify these developmental challenges for both parents and youth.

Rolland's Family Systems Model is an integrated family development perspective, which acknowledges that functioning within the context of a chronic illness is a multigenerational combination of individual, family and illness development (e.g., Rolland, 1987a, b; Rolland & Williams, 2005). Among families not facing a chronic illness, the progression from centripetal (e.g., dependence) to centrifugal (e.g., emancipation) family organization is dictated by the development of the child and their increasing need for independence. Rolland notes that illnesses characterized by flares or relapses (such as JRD) require flexibility upon the part of all family members—this is essential because of the necessity of altering family organization to fit the current state of the illness. Therefore, the natural inclination of parents to behave in a more centrifugal, or disengaging, manner as their children reach adolescence is disrupted by the presence of restrictions and increased vigilance imposed by the child's illness.

Although a growing emphasis on these issues is seen among studies focusing on children, research on parent adjustment has largely not followed the same trajectory. The present study attempts to address these limitations in the extant literature by examining potentially different patterns in the association between illness intrusiveness and global distress among parents of children of varying ages.

To fulfill this aim, a review of the current literature in the area is presented. First, a review of the literature associated with medical and clinical issues in JRD (diagnosis, disease subtypes, prognosis, and treatment issues) will be presented. Second, literature examining socio-emotional issues in pediatric chronic illness from the perspective of parent adjustment is reviewed. Specific factors affecting parents will be discussed: these include cognitive appraisals (namely illness intrusiveness), and the development/age of their child. The effect of developmental expectations will be conceptualized within Rolland's Family Systems Model (e.g., 1987; Rolland & Williams, 2005).

Finally, a study is described that examines whether the illness intrusiveness-parental distress relationship among parents of children and adolescents who have been diagnosed with a JRD varies by the age of the child. In other words, the potential moderating role of child age on the illness intrusiveness-parental distress relationship is examined. It is hypothesized that the illness intrusiveness-parent distress relationship will be significant regardless of the age of the child. However, for parents of older children with JRD, this association will be significantly greater compared to those with younger children.

CHAPTER II

REVIEW OF THE LITERATURE

Medical and Clinical Issues in JRD

Issues Associated with Diagnosis

The Juvenile Rheumatic Diseases (JRD) consist of juvenile rheumatoid arthritis (JRA), systemic lupus erythematosus (SLE), juvenile spondyloarthropathies, and juvenile dermatomyositis (JDMA), and are a series of autoimmune disorders characterized by a highly similar presentation. These symptoms, including connective tissue inflammation, pain, restricted ability, and a chronic course (Vandvik & Hoyeraal, 1993), make differential diagnosis difficult. Unfamiliarity on the part of medical staff often results in delays in both treatment and referrals to specialists, which are associated with poorer long-term outcomes. Further compounding this problem, even for specialists, JRD are highly unpredictable and unstable—symptoms that suggest a diagnosis of JRA immediately after symptom onset may later be more congruent with another rheumatic diagnosis once more specific disease markers appear (Cassidy & Petty, 2001). Thus, attaining an accurate diagnosis may really be due to a process of elimination (Vandvik & Hoyeraal, 1993; Cassidy & Petty, 1990), concluding far after the onset of significant symptoms; even then, misdiagnosis is common. This seems to be especially true for JRA, where 22% of diagnosed children received a replacement diagnosis within ten years (Flato, Aasland, Vinje, & Forre, 1998). Even after obtaining an accurate diagnosis and treatment plan, symptoms may persist and even worsen despite parents' best efforts to the

contrary (Young, 1992). Thus, the process of diagnosis and eventual treatment is frequently highly distressing for parents of children diagnosed with a JRD.

Disease Subtypes

Juvenile rheumatoid arthritis (JRA) is the most commonly diagnosed JRD, and ranks as one of more prevalent chronic childhood illnesses. Distinctive characteristics of JRA include persistent inflammation of joints, restricted functional ability, and pain (Lovell, 1997). Growth is commonly delayed across the disease course, and is due to either corticosteroid use or the presence of progressive symptomatology. Normal development can be anticipated during periods of remission; however, permanent delay in growth and development of secondary characteristics may be seen. Regardless of disease severity and growth delays, some atrophy of the muscles around affected joints is common (Cassidy & Petty, 2001). Generally, onset of inflammatory symptoms occurs prior to age 16 (Kewman, Warschausky, & Engel, 1995), with increases in prevalence at ages 2, 8, and 12 (Cassidy & Petty, 2001), thus illustrating the potential damage to a child's growth. In addition, JRA is more common in girls than boys; however, incidence by gender and age is different for the pauciarticular, polyarticular, and systemic subtypes (Lovell, 1997).

Pauciarticular JRA consists of involvement in fewer than five joints and is the most common subtype among the general population. Occurring in up to 50% of children with JRA (Lovell, 1997), onset is typically before age 10. Gender differences are prominent, as with many rheumatoid diagnoses, with boys affected five times more than girls (Kewman et al., 1995). Large joints are most frequently affected irrespective of gender; knee involvement is most common among children with only one arthritic joint

(Lovell, 1997). Arthritis is typically the only disease manifestation—growth retardation, subcutaneous nodules (painless nodules often on the heel or elbow) and cardiac involvement are usually absent for this subtype (Cassidy & Petty, 2001).

The next most common subtype, polyarticular JRA, is present in approximately 40% of children diagnosed with JRA. Consisting of arthritis in at least five joints, most children have over 20 joints involved, and 75% of these children show symmetric involvement. Early symptoms often include weight loss or failure to make expected gains, low-grade fever, anemia, and growth retardation. Onset is not age-specific, but is three times as common among girls as among boys (Lovell, 1997). Later onset, however, is associated with a more adult symptom pattern of rheumatoid arthritis among rheumatoid factor seropositive girls (Cassidy & Petty, 2001). In addition to the presence of subcutaneous nodules, this late onset is usually indicative of a poor disease course consisting of progressive and deforming disease activity (Calabro, Marchesano & Parrino, 1989).

The onset of systemic JRA is independent of both age and gender, and affects around 10% of children with JRA. This subtype is characterized by attacks of spiked fevers and pink rashes late in the day. These attacks are highly unpredictable, both in onset and in length; half of the children diagnosed with systemic arthritis will have more than one attack. During a remission of these classic systemic symptoms, half of the diagnosed children show symptoms of severe arthritis as well (Lovell, 1997). Complications in severe cases commonly include lymphatic, cardiac, liver, and spleen involvement, as well as uveitis (Cassidy & Petty, 2001).

The other diagnoses contained under the JRD umbrella, although intrinsically different than JRA, frequently contain an arthritic component, thus complicating diagnosis. Systemic lupus erythematosus (SLE) involves abnormal production of antibodies to components of cell nuclei, which causes inflammation, blood vessel abnormalities, and immune changes. It is characterized by a butterfly rash, arthritis, and arthralgias. Unlike JRA, however, arthritis in children with SLE does not destroy the affected bone (Lehman, 1997). Onset primarily occurs in adolescence (Cassidy & Petty, 2001), with girls accounting for more diagnoses by a ratio of 4 to 3.1. Additional symptoms may include nephritis, diffuse tissue lesions, photosensitivity, fever and lymphadenopathy (Lehman, 1997). In more severe cases, complications may include cardiac, gastrointestinal, renal, and vascular difficulties (Cassidy & Petty, 2001), and significant nervous system involvement is present in 19% to 36% of children diagnosed with SLE. Although uncommon, fatalities among SLE patients are usually due to renal failure or general infection (Lehman, 1997).

The juvenile spondylarthropathies are a class of diagnoses, with juvenile ankylosing spondylitis (JAS) as the most common subtype. JAS occurs more often in boys than girls, with onset in late childhood or adolescence (Cassidy & Petty, 2001). Characteristic, although nonspecific, symptoms include large joint arthritis, with asymmetry in the lower extremities. JAS is present in 10% of children with arthritis; common symptoms include back pain and stiffness. Onset typically involves the spine, with peripheral arthritis frequently following—the hips are most often affected. Peripheral involvement extends to polyarticular arthritis in 25% of patients (Lovell, 1997). Although JAS follows a highly unpredictable course of remissions and flares,

prognosis is usually good; however, if complications arise, they are typically of cardiac or gastrointestinal nature (Khan, 1993).

Juvenile dermatomyositis (JDMA) is a disease of the connective tissues characterized by diffuse vasculitis. Affected areas may include the skin, muscle, and gastrointestinal system. Initial symptoms frequently consist of tenderness, fever and a rash on the eyelids, as well as muscle weakness. Additionally, one-fifth of children diagnosed with JDMA have arthritic symptoms, and cardiac symptoms leading to myocarditis are not uncommon. Onset is most frequent among girls from ages five to fourteen, and appears to be precipitated by genetics and infections (Lehman, 1997).

Long-term Prognosis

JRD are, by their very nature, chronic diseases with potential symptoms and effects lasting into adulthood. Adults previously diagnosed with JRA show greater mortality compared with the population at large, as well as increased limitations to their functioning such as pain, and energy level. In addition to these general health issues, 65.9% reported current arthritic symptoms (Peterson et al., 1997). Perhaps illustrating the unpredictable nature of JRD, however, over half of patients with JRA were in remission a decade later (Flato, Aasland, Vinje, & Forre, 1998). Fatalities due to JRA are fortunately rare, and are usually due to cardiac or renal complications. Mortality rates are from 2-4% (Cassidy & Petty, 2001), and are mostly among children diagnosed with systemic arthritis (Petty, 1999). These data are encouraging; however, there is no way to predict specific outcome for children diagnosed with a JRD, as the course is so unreliable (Vandvik & Hoyeraal, 1993).

Issues Associated with Treatment

Pharmacological treatment among children with JRD is highly similar to adult RD, although careful attention to side effects is necessary. This is the most prevalent and efficient method of treating JRD, although physical therapy, maintaining a normal activity level, and symptom management techniques such as the use of electric blankets to decrease morning stiffness may be helpful (Rhodes, 1991). Use of non-steroidal anti-inflammatory medications such as aspirin, ibuprofen, tolmentin, naproxen, and fenoprofen is common, effective, and well tolerated. These medications are popular in part because of the speed of their effect—most children show symptom reduction within a month. Use of aspirin is declining, however, due to the risk of severe disease complications such as Reye's syndrome. If this inexpensive option does not reduce symptoms, such as with more severe cases of polyarticular or systemic arthritis, intramuscular gold therapy or corticosteroids such as prednisone may be used. Oral administration of corticosteroids results in fewer side effects, as frequent injections can damage cartilage within the joint. Treatment options outside of the NSAIDS are problematic, not only because of the severity of their side effects, but also because symptoms are reduced slowly—methotrexate, for example, takes 3-6 months to achieve its maximum effectiveness. As such, these drugs are primarily used with children diagnosed with severe polyarticular or systemic arthritis (Lovell, 1997).

Socio-emotional Issues in Pediatric Chronic Illness

Parent Adjustment

Although considerable research has focused on child adjustment to chronic illness, parental adjustment is equally important. Thompson et al. (1993a, b) have

highlighted the complex interplay between parental distress and subsequent child maladjustment among chronically ill children. Cross-sectional (Daniels, Moos, Billings, & Miller, 1987) as well as longitudinal (Timko, Stovel, & Moos, 1992a, b; Timko, Baumgartner, Moos, & Miller, 1993) results based in JRD populations highlight the lasting importance of parent variables in determining child adjustment. Indeed, parent distress actually seems to be a more important determinant of child maladjustment and psychosocial outcomes than disease-related variables, especially for mothers (e.g., Chaney et al., 1997; Frank et al., 1998). Likewise, objective disease severity is less important in predicting parents' distress than subjective ratings of illness severity by the parent, particularly when parents perceive their child's illness as negatively influencing their ability to parent (Walker, Ford, & Donald, 1987).

Compared to research on children's adjustment to illness, far less research has been conducted examining parents' adjustment to their child's illness. The data that do exist suggest that, in general, parents of children diagnosed with chronic illnesses are at greater risk for a host of psychological difficulties compared to the general population. For example, Timko, Stovel, and Moos (1992a) found that among a sample of 165 children diagnosed with JRD and their parents, levels of parent depression, drinking problems, and negative life events remained stable over a period of one year.

Landolt et al. (2002) examined adjustment in a sample of parents of 38 children diagnosed with Type 1 diabetes. Half of mothers and 40% of fathers displayed clinical or subclinical symptoms of posttraumatic stress; nearly 25% of parents displayed symptoms that met full DSM-IV criteria for Posttraumatic Stress Disorder. Additionally, Dahlquist et al. (1993) found that, among 134 parents of children recently diagnosed with cancer,

25% of mothers and 28% of fathers reported significant marital distress, which was predicted by general emotional distress, anxiety, and maladaptive coping mechanisms.

Similarly, Chaney et al. (1997) found that 50% of fathers and 33% of mothers of children diagnosed with Type 1 diabetes demonstrated significant adjustment difficulties. These levels of distress remained relatively consistent over a one-year period; indeed, only 25% of both mothers and fathers demonstrated changes in psychological symptoms as measured by the SCL-90-R.

Thompson et al. (1994) found that in two longitudinal studies of mothers of children diagnosed with sickle cell disease or cystic fibrosis, a moderate level of maternal distress was found in both samples. Further, both studies found moderate stability in maternal adjustment over time. The first study, with a sample of 57 mothers of children diagnosed with cystic fibrosis, found that the rate of poor maternal adjustment decreased between time 1 and time 2. However, those mothers with stable poor adjustment reported more daily stress and familial conflict than the other mothers in the sample. The second study, with a sample of 60 mothers of children diagnosed with sickle cell disease, also found that the amount of poor maternal adjustment decreased over time. Further, those mothers also reported more daily stress, but also reported higher levels of illness-related stressors and lower levels of family support.

More recently, Sloper et al. (2000) found that, among a sample of 68 mothers and 58 fathers of children diagnosed with cancer, significant levels of distress were present for both parents over time. Variables predictive of parental distress differed by gender, however, with mothers being influenced by appraisals of illness-related strain, family cohesion, and coping strategies. Although fathers' distress was similarly influenced by

these appraisals, cohesion, employment concerns and frequency of hospital admission variables were also important determinants of fathers' distress.

Although the majority of studies have found poorer adjustment for parents, multiple studies have found that parents of chronically ill children do not report added signs of distress beyond those seen in the general population. For example, Cadman, Rosenbaum, Boyle, and Offord (1991) found that among a sample of 1869 households, parents of chronically ill children experienced small elevations in symptoms such as negative maternal affect. Overall, however, there were no significant differences between parents of chronically ill and healthy children. Similarly, in a sample of parents of 64 children diagnosed with JRA, Gerhardt et al. (2003) found no differences in distress between these families and matched controls. Daltroy et al. (1992) also found comparable distress levels for mothers of 102 children diagnosed with arthritis and those seen among the general population. Further, depressed mood scores for the Daltroy et al. sample were significantly lower than normative data for psychiatric outpatients.

Despite a few exceptions (e.g., Cadman et al., 1991; Gerhardt et al., 2003), it is apparent that parents of children diagnosed with a chronic illness are generally at an increased risk for psychological difficulties; however, there appear to be extenuating factors that increase or decrease the potential for emotional and adjustment problems. Thus, adjustment difficulties do not seem to be inevitable, but instead seem to be related to a number of evaluative variables, such as perceived stress, illness impact, and other cognitive factors (e.g., Sharpe, Brown, Thompson, & Eckman, 1994).

Factors Affecting Parent Adjustment

Cognitive appraisals. Many studies have found that a variety of cognitive appraisal variables are key determinants of adjustment for parents of chronically ill children. For example, in a sample of 53 mothers of children ages two to eleven diagnosed with JRA, Lustig, Ireys, Sills, and Walsh (1996) found that both biological and functional severity aspects of the disease contributed to the mothers' level of distress. Importantly, however, the effects of disease variables on maternal distress were not direct, and were partially mediated by maternal appraisals of the extent to which the child's illness impacted the functioning of the family as a whole. In other words, despite the direct effects of child illness on maternal adjustment, this relationship was mediated by the perceived impact of the illness.

The influence of maternal appraisals extends to perceived level of stress. For example, Thompson et al. (1994) found that, among two longitudinal samples totaling 117 mothers of children diagnosed with either cystic fibrosis or sickle cell disease, ratings of daily stress differentiated mothers with varying levels of adjustment. Independent of initial maternal adjustment as measured by the SCL-90-R, maternal levels of daily stress were predictive of maternal adjustment at the one-year follow-up. Additionally, mothers demonstrating poorer adjustment also endorsed greater levels of illness-related stress.

Further, Manuel (2001) found that, among a sample of 92 mothers of children diagnosed with JRA, mothers' perceived control (as measured by the Multidimensional Health Locus of Control Scale; Wallston, Wallston, & Devellis, 1978) over their child's illness was associated with lower parent distress. This relationship remained significant even when illness-related stress was high. Thus, appraisal style acted as a moderator

between stress and psychological symptomatology. Similarly, Sloper (2000) found that, among a sample of 68 mothers and 58 fathers of children diagnosed with cancer, both subjective appraisals of the child's illness and their perceived ability to handle associated stressors (as described by Thompson et al., 1992) were predictive of parent distress.

These studies suggest that, much like child adjustment to illness, parent adjustment is regulated in a complex manner—this includes child, demographic, family, and other systemic variables, including the parents' cognitive appraisal processes. Despite this, research on parents' cognitions in response to their child's illness is relatively scarce.

Illness intrusiveness. Because of the significant level of physical impairment and subsequent limitations that frequently accompany a JRD diagnosis (Henderson et al., 1995), one cognitive appraisal variable that seems to be particularly salient to parents of children diagnosed with a JRD is *illness intrusiveness*. On the surface, illness intrusiveness appears to be highly similar to perceived disability due to illness. Devins et al. (1983-84) state that although illness intrusiveness includes perceived disability due to illness, illness intrusiveness represents a broader concept that attempts to capture perceptions of the extent to which participating in daily, non-disease related activities such as church and school attendance are restricted due to the disease and its effects. This is illustrated by Devins and colleagues' characterization of illness intrusiveness as consisting of wide-ranging and generalized "illness-induced barriers" (1983-84, p. 329).

These barriers are hypothetically linked to increased maladjustment and depressive symptoms both by reducing positive, reinforcing experiences, and by decreasing perceived control over important personal outcomes (Devins, Edworthy,

Guthrie, & Martin, 1992). Thus, it is not the disease itself that determines the intrusiveness of the illness; rather, it is the individual's subjective appraisal of how the disease interferes with the person's life across multiple domains that constitutes illness intrusiveness and subsequent distress (Devins, Seland, Klein, Edworthy, & Saary, 1993).

Although there appears to be conceptual relevance of illness intrusiveness to parents' adjustment in JRD, most studies examining the intrusiveness-adjustment link in adult populations have focused on adults' cognitive appraisals of their own illness and the relation of those appraisals to emotional adjustment. Indeed, a number of studies have documented the importance of perceived illness intrusiveness to psychological adjustment. For example, among a sample of 78 adults diagnosed with multiple sclerosis, Mullins et al. (2001) found that self-ratings of illness intrusiveness related to illness outcomes exerted a significant negative effect on adjustment (as measured by the SCL-90-R; Derogatis, 1993). Importantly, this effect was found to be independent of physical difficulties. In other words, even though participants with multiple sclerosis reported physical challenges, perceived illness barriers contributed significant incremental variance to adjustment beyond the influence of physical impairment due to the disease. Further, Franche et al. (2004) found that, among a sample of 81 men and 67 women who had experienced a cardiac event, depression assessed during their initial hospitalization predicted increased illness intrusiveness one year later. These may suggest that illness intrusiveness and depressive symptoms influence each other over time.

In addition to its direct influence on distress, illness intrusiveness also has indirect effects in combination with individuals' perceived psychosocial resources and other cognitive variables. For example, Devins, Stam, and Koopmans (1994) found that among

a sample of 51 cancer patients who had recently undergone a laryngectomy, the combined influence of illness intrusiveness and perceived stigma associated with the illness had a significant effect on psychosocial well being. More specifically, highly stigmatized self-perceptions enhanced the impact of illness intrusiveness on maladjustment. Further, among a sample of 90 adult recipients of a bone marrow transplant, individuals who endorsed increased illness intrusiveness also demonstrated a significantly greater tendency to incorporate the *transplant patient identity* into their self-definition (Beanlands et al., 2003). Specifically, this finding was heightened when patients perceived higher levels of hopelessness, helplessness, and distress. Thus, patients who were experiencing increased illness-related restrictions were more likely to be negatively influenced by those restrictions, and were more likely to have a disease-oriented self-concept.

Much like the adult illness intrusiveness literature, existing research examining illness intrusiveness in pediatric chronic illness populations has largely targeted the relation between children's perceptions of intrusiveness and their own adjustment. For example, Wagner et al. (2003) found that children's perceived illness intrusiveness moderated the relationship between parent and child distress among a sample of 45 children ages 9-17 diagnosed with JRD. Specifically, children's heightened perceptions of illness intrusiveness increased the impact of parent distress on child depressive symptoms. This study not only highlighted the important role of parent adjustment in determining child adjustment, but also the indirect effect of perceived illness intrusiveness in this process.

Although illness intrusiveness appears to play an important role in child adjustment to JRD, only one known study has addressed illness intrusiveness from a parenting or family systems perspective. Andrews and colleagues (2007) found that, among a sample of 40 Native American and Caucasian parents of children diagnosed with a JRD, parent-perceived illness intrusiveness was significantly associated with poorer parent adjustment. Moreover, this intrusiveness-adjustment relationship was moderated by ethnic minority status. Illness intrusiveness was related to parent adjustment in the Native American sample of parents, but not in the Caucasian sample. These results suggest that the degree of illness-induced disruptions (i.e., illness intrusiveness) may be particularly relevant for ethnic minority families and other groups already at risk for health and social difficulties.

It is clear that the relationship between illness intrusiveness and parent adjustment is affected by a variety of variables within the family system; however, these indirect relationships have not been researched to any significant degree. Given the interplay between parent and child adjustment, it seems that a clearer understanding of how the parents perceive the intrusiveness of their child's illness would be vital to appropriately treating children and their families. The relationship between parent adjustment and parent-reported intrusiveness, however, is likely to be further complicated when the developmental status of the child and associated changes in family structure and responsibilities are taken into account.

Development and age of the child. Another factor that may increase the impact of illness intrusiveness involves developmental expectations. For example, research on 110 adults between the ages of 24 and 78 who had been diagnosed with rheumatoid arthritis

(RA) indicates that the extents to which individuals perceive their own illness as interfering with disease-unrelated activities is associated with significant increases in depressive symptoms and decreased overall quality of life (Devins et al., 1992). For younger patients, this decrease in physical capacity and rewarding events is in direct contrast to normal developmental expectations; this is vastly different than for older patients who are more likely to perceive that the physical limitations due to their illness are more appropriate for their age (Rolland, 1987). It seems that this incongruity would be even more devastating for pediatric patients and their caregivers given the expectations and activity levels of same-age peers.

For all children, the expectations of independence and autonomy that are placed upon them change dramatically with increasing age. Indeed, parents of adolescents typically alter their expectations and behaviors as their children achieve greater freedom and potential for self-care (Steinberg & Silverberg, 1986). Among parents and adolescents, however, the timetable for these milestones may not match. Among a sample of 508 families with children ages 12-18, parents reported later dates for the achievement of developmental milestones than did their adolescent children, although the reported sequence of events was very similar (Dekovic, Noom, & Meeus, 1997). For both parents and children, the expected time of mastery for these events was determined by a variety of variables: age, gender, pubertal timing, and temperament.

Studies in this area are rare among pediatric chronic illness populations; however, Ungerer and colleagues (1988) showed that, among 363 children diagnosed with JRA, reported restrictions were at their lowest during the preschool years and increased to a high point in adolescence. Interestingly, although a significant relationship between

disease severity and family problems was observed among the older children and adolescents, the same effect was not observed for younger children. The authors hypothesized that this was because younger children have a social life that is focused within the family and because their physical care is easier to cope with (due to the size of the children and a shorter duration of symptoms).

Further, Eisner and Berrenberg (1995) note these findings demonstrate that adolescents are “forced into extended dependency” (p. 112) by their lowered mobility, and that these restrictions have an increased social impact in comparison to younger children. Thus, the adolescent diagnosed with a chronic illness may need to rely on his/her parents for emotional and illness-related support. Unfortunately, this occurs at a time when both parents and child expect the adolescent to have more autonomy and social experiences outside of the family. This creates a gap between developmental and environmental needs (Holmbeck & Kendall, 2002), which can lead to increases in illness related and unrelated stressors among both parents and their children.

However, a specific investigation of how this lack of independence is perceived by parents has not been conducted. This paucity of research is unfortunate, given that parents may be more distressed about the restrictions of their child’s illness than the children themselves; their “wider experiences” (p. 110) make them more aware of what constitutes typical development and abilities (Eisner & Berrenberg, 1995). This is likely to be more applicable for younger children, whose social experiences and contacts outside the family are fewer.

However, among a sample of 197 adolescents ages 11-17 years who had been diagnosed with epilepsy, Heimlich and colleagues (2000) found that older teenagers had

more negative attitudes towards their illness as measured by the Child Attitude Toward Illness Scale (CATIS; a measure developed by Heimlich for children diagnosed with epilepsy and asthma). This was found to be especially true for girls and for children with a more severe disease course. The authors noted that this finding may have been due to having to delay developmental expectations of autonomy, while watching their healthy peers achieve those same goals. Similarly, Lebovidge and colleagues (2005) found that, among a sample of 75 children ages 8-18 diagnosed with chronic arthritis, older adolescents were more likely to be experiencing higher levels of depression and anxiety. Again, attitude towards the illness was a significant predictor of adjustment for the children in the sample, regardless of age. For the older children, adolescents whose disease symptoms are more overt become aware that their independence is progressing differently than their peers; this experience will likely lead to increased difficulties such as stigmatization and distress.

Despite this potential delay in autonomy, an increasing level of independence in administering treatment is often expected. For example, among a sample of 222 children diagnosed with insulin-dependent diabetes mellitus, Holmes and colleagues (2006) found that parents used the age of their child as the primary factor in determining levels of responsibility for self-care. Unfortunately, without the presence of increased knowledge and self-efficacy, older age was actually related to a poorer level of self-care. Similarly, among a sample of 106 children diagnosed with asthma, age predicted responsibility for treatment management, as well as increased knowledge and reasoning about asthma. It should be noted that in this study, adherence did not increase as youth assumed greater

treatment responsibility or gained increased disease-related information (McQuaid et al., 2003).

The finding of poor adherence among adolescents is a common one, as teenagers are less rule-oriented about their disease-related care than younger children (e.g., Johnson & Meltzer, 2002); this effect seems to be more significant among those adolescents who are primarily oriented towards their peers rather than to adults (Weisz & Hawley, 2002). Regardless, the impact of this decrease in rule orientation is heightened among children diagnosed with chronic illnesses, as disease and health outcomes across the lifespan may be altered (Williams, Holmbeck, & Greenley, 2002). Thus, the need for parental supervision with treatment increases, which should simultaneously increase parent perceptions of illness intrusiveness due to the restrictions imposed by the disease and its management.

Despite a pattern of poor adherence, however, children and adolescents do appear to need a measure of autonomy to be successful and well-adjusted; indeed, problem behaviors may arise when the child's abilities and parental assistance do not vary together as the child grows older (Williams, Holmbeck, & Greenley, 2002). More specifically, providing a developmentally inappropriate level of assistance may lead the adolescent to be overly dependent, and to have low levels of self-efficacy. In a theoretical discussion of this, Anderson and Coyne (1991) refer to inappropriate support that inadvertently undermines the child's independence as "miscarried helping" (p. 167); this is an apt term for any assistance that is poorly timed or excessive in nature, yet offered with genuine concern for the child's health and well-being. This inappropriate pattern of helping may take a variety of forms (e.g., Thomasgard, 1998), that include perceptions of

vulnerability, which may predispose the parent to behave in an indulgent manner, and overprotective parents, who attempt to regulate the health of their child through increased control. Regardless of what issue is behind parents' helping behavior, however, the nature and consequences of that behavior is likely to change based on the developmental needs of the child.

Excessive overprotection, for example, appears to be directly in conflict with normative adolescent development. Being overly protective for the child's developmental age has been shown to be more common among parents of chronically ill children; Holmbeck and colleagues (2002a) found that 68 parents of 8-9 year old children diagnosed with spina bifida were significantly more overprotective than parents of matched, healthy controls. Further, overprotection was found to not only be associated with lower levels of child-reported autonomy, but also with a lower likelihood of parents granting the child more autonomy in the future. Among the spina bifida sample in this study, the relationship between parental overprotection and externalizing behavior problems was mediated by behavioral autonomy. Although this study used a narrow age range, it seems that this relationship would become even more important (and problematic) as children approach adolescence due to their expectations as well as to increased autonomy on the part of healthy peers.

Providing an overly high level of support is not the only way in which a mismatch between abilities and assistance may be conceptualized; an overly strict parenting style may lead to adverse outcomes as well. For example, Davis and colleagues (2001) have found that, among a sample of 55 parents of children ages 4-10 who had been diagnosed with Type I diabetes, restrictive parenting was associated with poorer metabolic control.

Thus, parental warmth and support, as well as family cohesion, were associated with greater treatment adherence for young children; the authors noted that this effect may be due to increased self-control on the part of authoritatively parented children.

Unfortunately, if parents perceive their child's disease or other factors to be threatening, they may engage in a more restrictive parenting style, thereby inhibiting the ability of their child to progress in independent disease management.

Adjustment among children and adolescents is clearly impacted by low levels of autonomy and independence; however, higher levels of independence may induce stress as well. Among a sample of 94 children ages 7-20 who had been diagnosed with JRA, autonomy was unrelated to illness-related worries for girls. Among boys, however, greater autonomy was associated with increased illness related worries (Helgeson et al., 2003). The authors hypothesize that this is because increased autonomy is developmentally appropriate and contributes to self-esteem, especially for boys. Conversely, autonomy in treatment management simultaneously provides the new stress of accountability for adolescents who may be unaccustomed to this added responsibility.

At a certain level, increases in parental supervision and protection are adaptive for parents of children facing an unpredictable and chronic disease course. As Anderson and Coyne (1991) discuss, parents have a delicate balancing task ahead of them—to integrate the need to develop their child's independence and self-control with the desire to insure that their child remains adherent to treatment. Although difficult across childhood and adolescence, the struggle between these two issues is likely not as important in early to middle childhood, when a certain degree of dependence is expected. However, as the

child transitions to adolescence, it seems that parents would be torn by these conflicting responsibilities, thus resulting in more symptoms of distress.

Rolland's Family Systems Model

Although not often investigated in a clinical context, an integrated theoretical perspective of these issues is well-illustrated by Rolland's Family Systems Model, which acknowledges that functioning in the context of a chronic illness is a multigenerational combination of individual, family and illness development (e.g., Rolland, 1987; Rolland & Williams, 2005). This impact extends beyond the simple relationship between the parent and chronically ill child, as parent adjustment has additionally been found to have an impact on the psychosocial well being of healthy siblings of chronically ill children (e.g., Jaworski, 1993).

Among families not facing a chronic illness, the progression from centripetal (or engaging) to centrifugal (or disengaging) family organization is dictated by the development of the child and their increasing need for independence (Rolland, 1987; Rolland & Williams, 2005). He notes that chronic illnesses (such as JRD) that are characterized by flares or relapses require flexibility upon the part of all family members—this is essential because of the necessity of altering family organization to fit the current state of the illness. Therefore, the natural inclination of parents to behave in a more centrifugal, or *disengaging*, manner as their children reach adolescence is disrupted by the presence of restrictions imposed by the child's illness. Therefore, the expectation of independence is subsumed under more immediate, illness-related concerns.

Other researchers have noted that this familial flexibility is essential to surmounting the obstacles inherent in developmental change. Although Levinson's

(1986) work is focused on adults (indeed, the initial phase of life in his model encompasses all of childhood through the age of 20), he notes that passage to each new “era” in a human life is marked by lengthy transitional periods of up to five years, and that each era and transitional period consist of a variety of developmental tasks. This distinguishes his model from others wherein a child simply reaches maturity and promptly becomes an adult.

According to Levinson, the *Early Adult Transition*, beginning at around age seventeen and lasting until the age of twenty-two, requires great flexibility on the part of adolescents and their families as they negotiate the process of individuation. Without this increase in autonomy, Levinson states that life can “stagnate” and prevent the adolescent from forming the concepts of life necessary to become a productive adult. Therefore, the transition from childhood to adulthood (in addition to other transitional periods) is essentially an era of vulnerability and discontinuous change that is difficult to navigate even without the presence of additional factors such as illness. In Rolland’s terminology, the presence of centripetal relationships among children diagnosed with a chronic illness should lead to difficulties in fully reaching maturity.

Summary. Parents of children who have been diagnosed with a JRD face an uncertain disease course that may delay the mastery of a variety of their child’s developmental milestones, including the establishment of autonomy. Research on adults with rheumatoid arthritis (RA) suggests that increased illness intrusiveness is associated with depressive symptomatology and lower quality of life, especially among young adults (Devins et al, 1992). This indicates that the effects of lifestyle disruptions due to illness may be more pronounced when they are inconsistent with normal developmental

expectations regarding unanticipated reductions in functional independence and physical activity levels (e.g., Rolland, 1987, 1988). It seems clear that a similar (although reversed) relationship would be observed among pediatric populations.

Clearly, for older children and their parents, the limitations imposed by a chronic and debilitating illness are indeed in marked contrast to developmental expectations typical to adolescence such as independence. Therefore, although the age of the child has not often been investigated as a significant predictor of parent adjustment in response to chronic illness, it seems likely that it would affect the relationship between illness intrusiveness and adjustment for parents of children diagnosed with a JRD. Specifically, it seems that parents of older children would experience more distress at high levels of intrusiveness due to the incongruence between developmental expectations and their child's abilities.

CHAPTER III

THE PRESENT STUDY

Based on the preceding review of the literature, it is apparent that children diagnosed with a juvenile rheumatic disease and their families face a variety of challenges across multiple domains. Parent adjustment, for example, is presumed to be multi-determined by a host of variables, including cognitive appraisal processes. Except for a few studies (e.g., Lustig, Ireys, Sills, & Walsh, 1996), however, cognitive appraisals have been largely overlooked in investigations of parent adjustment to childhood chronic illness.

One such cognitive appraisal variable, illness intrusiveness, appears to be particularly relevant to the illness process in rheumatic diseases (Devins & Edworthy, 2000). However, the vast majority of research on illness intrusiveness in pediatric populations has focused exclusively on children's perceptions of intrusiveness and its relation to child adjustment—extant literature has largely ignored parents' intrusiveness perceptions as a determinant of their own distress levels. Because parent adjustment influences children's adjustment to chronic illness, such information could prove useful in developing interventions to modify parents' illness perceptions, their adjustment, and ultimately, children's adjustment.

Also conspicuously absent in the pediatric chronic illness literature are investigations examining potential differences in the manner in which cognitive appraisals influence adjustment across age groups. It seems likely that age would have an

indirect effect on parent adjustment through the disruptions that these diseases may cause in their children's mastery of certain developmental milestones, such as independence. The effect of lifespan expectations on illness intrusiveness-adjustment relations has been noted in an adult population (e.g., Devins et al., 1992). This study indicated that the extent to which individuals perceive their own illness as interfering with disease-unrelated activities was significantly related to increases in depressive symptoms and decreased overall quality of life, particularly among the younger participants with RA. The authors interpreted these findings as an indication that the effects of lifestyle disruptions due to illness may be more pronounced when they are inconsistent with normative lifecycle expectations regarding reductions in activity level. These results appear to be highly relevant for families of chronically ill children, given the changes in normative developmental expectations across childhood and into adolescence. However, similar studies have not been carried out among a pediatric population.

The present study attempted to address these limitations in the literature by investigating the association between parent-reported illness intrusiveness and parent distress in a sample of children ages 9-18 who have been diagnosed with JRD. Specifically, the present study examined the potential moderating influence of child age on the relationship between parent-reported illness intrusiveness and parent distress. Due to the increases in expected autonomy for older children, it was expected that for parents of older children with JRD, the association between parent-perceived illness intrusiveness and distress would be significantly greater compared to parents with younger children.

Primary Hypotheses

Hypothesis 1. It was anticipated that greater parents' perceived illness intrusiveness would be related to poorer parent adjustment (i.e., increased distress) for parents of all children, regardless of the age of the child. It was hypothesized that this relationship would remain after controlling for demographic and disease variables.

Hypothesis 2. The association between illness intrusiveness and parent adjustment was expected to be significantly greater for parents of older children. Thus, it was expected that children's age would moderate the relationship between parents' perceptions of illness intrusiveness and parent adjustment, such that illness intrusiveness would be more closely related to poorer parent adjustment (i.e., increased distress) among parents of older children.

CHAPTER IV

METHODS

Participants and Procedures

Participants were 52 parents (23 Caucasian, 15 Native American, 5 Hispanic, 4 African American, 1 Asian, 4 Biracial) of children ages 9-17 ($M = 13.75$ yrs.; $SD = 2.42$) who had been diagnosed with JRA ($N = 30$), SLE ($N = 12$), JSA ($N = 3$), or JDM ($N = 7$). The families were recruited from the pediatric rheumatology clinic at Children's Hospital of Oklahoma at the University of Oklahoma Health Sciences Center. Institutional review board (IRB) approval for protection of human participants was granted, and written informed consent and assent were obtained from each participant. Inclusion criteria included the following: 1) diagnosis of one of the above-mentioned illnesses, 2) living at home and between the ages of nine and 17, and 3) the duration of the child's symptoms had been at least one year, irrespective of the time of diagnosis. Illness duration, however, was defined as the subtraction of the date of diagnosis from the date of participation, and ranged from .00 – 15.73 years ($M = 2.54$ yrs.; $SD = 3.29$). For complete information regarding disease, demographic, and psychosocial variables, see tables 1 and 2. Exclusion criteria consisted of: 1) the child had comorbid cognitive deficits such as mental retardation, 2) the child had a comorbid chronic illness, and 3) either the child or the parent were non-fluent English speakers. The primary rheumatologist verified the inclusion criteria before eligible participants were contacted.

Eligible participants were recruited in either of the two following ways. The majority of participants were recruited during a routine visit to the rheumatology clinic (N = 35). If the family chose to participate, parents completed a questionnaire packet. This packet was either returned to the clinic or to researchers via postage-paid mail. Participants not scheduled for upcoming appointments in the rheumatology clinic were contacted by phone and the packet was sent by mail (N = 17). These families had previously met the researchers prior to telephone contact. All those recruited, regardless of method, agreed to participate. Parent psychological adjustment did not differ as a function of recruitment method, $F(1, 51) = .72, p = .40$. Once participants returned the completed packet, they received \$10 compensation in the form of a gift card.

Instruments

Parent and Child Report

The *Brief Symptom Inventory* (BSI; Derogatis, 1993) is a 53-item questionnaire that assesses overall psychological adjustment (see Appendix A). Respondents rated the degree to which they were distressed by each psychological symptom over the past week. Rating was done on a Likert scale, where responses ranged from 1 (not a lot) to 4 (extremely). The global severity index (GSI) is the average score of the items and was used as the measure of parent distress ($M = .58; SD = .57$). The BSI has been previously found to have acceptable internal consistency; alpha coefficients range from .71 to .85 (Derogatis, 1993). For this study, Cronbach's alpha was .97.

The *Illness Intrusiveness Scale-Parent* (IIS-P) The IIS-P used in the present study was adapted from the original IIS (Devins et al., 1983). For this study, the original 13 items remain the same; directions were changed from “rate the extent to which your

illness...” to “*rate the extent to which your child’s illness interferes with your ability to perform as well as you would like to*”. The IIS-P is a 13-item measure that assesses the degree to which parents perceive their child’s illness as interfering with their own ability to engage in activities across a variety of life domains, such as work, relationships, and recreation (see Appendix B). Parents were asked to respond on a scale from 1 (*affects performance a little*) to 7 (*affects performance a lot*). Items were summed to achieve a total intrusiveness score, with higher scores indicating greater levels of illness intrusiveness ($M = 22.97$; $SD = 14.42$). Data from adult RA and lupus samples reflecting intrusiveness of adults’ own illness symptoms indicate that internal consistency estimates range from .87 to .94 and test-retest reliability indexes range from .79 to .85 (Devins & Edworthy, 2000). In the present sample, Cronbach’s alpha was acceptable, at .92.

The *Juvenile Arthritis Functional Assessment Report–Parent* (JAFAR-P; Howe et al., 1991) is a 23-item parent rating intended to cover a variety of subjective estimates of their child’s functional ability (see Appendix C). Questions address the frequency that their child is able to perform specific tasks related to daily functioning, such as reaching above their head, button a shirt, and brush their own teeth. Responses were made on a three-point Likert scale, where 0 refers to being able to perform the task *all the time*, and 2 refers to being able to perform the task *almost never*. Responses were summed, so that higher scores on the JAFAR-P indicate a perception of greater disability ($M = 4.66$; $SD = 6.04$). The JAFAR has demonstrated good construct validity and acceptable internal consistency for the parent-report (.93) version of the scale (Howe et al., 1991). Again, Cronbach’s alpha for this study was adequate at .92.

The *Children's Depression Inventory* (CDI; Kovacs, 1983; 1992) is a 27-item scale that measures depressive symptoms over the previous two weeks. Each of the items measures the severity of a depressive symptom on a 0 to 2 scale, and is scored by summing the 27 items for an overall index. Higher scores indicated greater child distress ($M = 8.69$; $SD = 8.33$). Although this study is an investigation of parent adjustment, previous studies (e.g., Thompson et al., 1992) have highlighted the reciprocal nature of the relationship between parent and child adjustment. In light of the transactional relation between child and parent adjustment, child adjustment is included as a covariate to provide a more conservative test of the relation between illness intrusiveness and parent adjustment. Cronbach's alpha for this study was adequate at .88.

Physician-Report

Functional Disability estimates were provided by the pediatric rheumatologist following a routine visit. Participants were classified into one of four categories, ranging from Class I (limited to no disability) to Class IV (severe disability) (cf. Hochberg et al., 1992). Average functional disability for this sample was 1.50 ($SD = .61$). This measure has been shown to be a valid indicator of functional disability in JRDs (e.g. Gerhardt et al., 2003) and was significantly correlated with parent-rated JAFAR-P scores in the present sample, $r = .44$, $p = .001$.

CHAPTER V

RESULTS

Preliminary Analyses and Selection of Covariates

Selection and entry of demographic and disease covariates were guided by multivariate models of adjustment to chronic illness (e.g., Thompson et al., 1993a), as well as by significant relationships between the covariates and the criterion variable. First, bivariate correlations were computed to identify significant relationships between key demographic and disease variables and GSI parent distress. Mean comparisons were also utilized to test for differences in GSI across demographic and disease variables.

Zero-order correlations revealed no significant relation on GSI with demographic (maternal and paternal education, marital status, gender) or disease (duration, parent and physician-perceived disability) variables. Further, one-way analyses of variance (ANOVA) revealed no significant gender or diagnosis differences for GSI. Although gender, diagnosis, parent-related disability and physician-rated disability (i.e., functional class) did not demonstrate a direct relationship to scores on the GSI, these variables were included as covariates to provide a more conservative test in accordance with extant psychosocial and medical literature.

Primary Analyses

Hypothesis 1. It was anticipated that greater parents' perceived illness intrusiveness on the IIS-P would be related to lower parent adjustment as measured by the GSI for all parents, regardless of the age of the child. Further, it was predicted that

this relationship would remain after controlling for demographic and disease variables. Prior to investigating this hypothesis, age and IIS-P were centered by subtracting the sample mean from each score. Then, a regression equation was constructed in which disease subtype, gender, ethnicity, disease years, functional disability, JAFAR-P, and total CDI were entered as a block on Step 1. On Step 2, child age and parent report of illness intrusiveness (IIS-P) were entered. Results revealed a significant main effect of IIS-P on GSI scores, $t = 3.06$, $p = .004$, thus demonstrating the direct relationship between illness intrusiveness and parent distress for the entire sample (See Table 2).

Hypothesis 2. The association between illness intrusiveness on the IIS-P and parent adjustment on the GSI was expected to be significantly greater for parents of older children in comparison to parents of younger children. Thus, it was expected that child age would moderate the relationship between parent perception of illness intrusiveness and global parent adjustment, such that illness intrusiveness would be more closely related to parent adjustment in the sample of parents with older children compared to the sample of parents with younger children. To investigate this hypothesis, a regression equation was constructed in which disease subtype, gender, ethnicity, disease years, functional disability, JAFAR-P, and total CDI were entered as a block on Step 1. On Step 2, child age and parent report of illness intrusiveness (IIS-P) were entered. On Step 3, the interaction term of child age x IIS-P was entered. Again, centered variables were used for child age and IIS-P, as well as the interaction term. Results revealed a significant age x IIS-P interaction, which accounted for an additional 5.55% of incremental variance in parental distress $F(1, 41) = 3.73$, $p = .001$. Observed power for the interaction was 0.89. The significant interaction and positive value of β (See Table 3) indicates that child age

moderated the relationship between parent perception of illness intrusiveness and global parent adjustment in the predicted direction. Specifically, illness intrusiveness was more closely related to parent adjustment among the parents of older children.

Post-hoc Probes

As suggested by Holmbeck (2002), post-hoc probes were conducted to determine the nature of the significant interaction found in the primary analyses. Conditional moderators were created for the centered age variable, so that HI_AGE equals zero when centered age is one standard deviation above the mean. Similarly, LO_AGE equals zero when centered age is one standard deviation below the mean. New interaction terms were then created by multiplying each conditional moderator with the centered IIS-P variable.

One regression equation was then computed to generate the slope for the high condition (HI_AGE) and another for the low condition (LO_AGE). For each equation, step 1 remained the same as in the primary analyses, while the main effects and new interaction term were entered simultaneously as step 2. Significance tests indicated that the simple slope of the older regression line was significant, $p = .001$; the simple slope of the younger regression line was nonsignificant, $p = .06$. Thus, post-hoc tests indicated that illness intrusiveness was significantly associated with distress for parents of older children, but was unrelated to parent distress among parents of younger children in this sample.

CHAPTER VI

DISCUSSION

The present study was designed to address two key limitations in the extant literature on adjustment to pediatric chronic illness. First, despite findings in the pediatric psychology literature indicating that parents of chronically ill children are at increased risk for emotional adjustment problems such as anxiety (e.g., Dahlquist et al., 1993) and familial conflict (e.g., Vandvik, Hoyeraal & Fagertun, 1989), little is known about the precise mechanisms responsible for this. Some data indicate that a variety of variables, including children's adjustment, influence parent adjustment (Thompson et al., 1993a, b); other data suggest that parents' perception of their child's illness and its management are major contributors to parent adjustment issues. Specifically, evidence suggests that parents' perceptions of the impact of their child's illness can have negative effects on parents' adjustment (Walker, Ford, & Donald, 1987). However, there are few empirical data to support this. One particular variable that seems to be particularly salient to parents of children diagnosed with a JRD is *illness intrusiveness*. On the surface, illness intrusiveness appears to be highly similar to perceived disability due to illness; however, it represents a broader concept that attempts to capture perceptions of the extent to which participating in daily, non-disease related activities such as church and school attendance are restricted due to the disease and its effects.

This variable, which represents perceived "illness-induced barriers" across a wide array of life domains (Devins et al., 1983; p. 329), was examined in the present study

because JRD often involve a significant decrease in children's activity levels (Henderson et al., 1995). Because of this, the role of parents' perceptions of interference with routine activities due to illness assumes particular importance.

Another shortcoming in the literature is the lack of data addressing adjustment issues among parents of children of varying ages. Given the close link between parent and child adjustment, parental expectations of the child and developmental changes in family structure and responsibilities introduce a further layer of complexity to this relationship. Indeed, Devins and colleagues (1992) demonstrated that age moderated the relationship between intrusiveness and distress among adults diagnosed with rheumatoid arthritis. This finding can be interpreted through the lens of changing expectations across the lifespan. To illustrate, older individuals were more likely to perceive that illness-induced limitations were appropriate for their age. Conversely, younger individuals were more likely to experience higher levels of distress because of the incongruity between their limitations imposed by their illness and age-appropriate expectations.

Among pediatric populations, it seems that issues of changing expectations would be equally, if not more important to the adjustment process. Certainly, all children experience (and parents expect) experience radically different levels of independence and autonomy as children progress into adolescence and then adulthood. As children achieve greater potential for self-care, parents' expectations for their child's independence and self-care typically increase (Steinberg & Silverberg, 1986).

Older chronically ill youth, however, are often "forced into extended dependency" by decreased mobility and increased illness demands (Eisner and Berrenberg, 1995; p. 112), resulting in the potential for greater social impact relative to younger children.

Thus, the adolescent with a chronic illness may experience increase reliance on his/her parents for emotional and illness-related support at a time when age-appropriate parental expectations are the exact opposite (i.e., increased independence). Under these conditions, illness-induced barriers and limitations would be at odds with parents' evolving expectations and lead to greater distress among parents of older children.

To address these gaps in the literature, two hypotheses were proposed for this study. First, greater parents' perceived illness intrusiveness on the IIS-P would be related to greater GSI parent distress for all parents irrespective of youth age. Second, it was anticipated that the youth age would moderate the relationship between illness intrusiveness and parent distress. Specifically, it was anticipated that illness intrusiveness would be more closely related to parent distress among parents of older youth.

Results of the primary analyses were consistent with the first hypothesis; namely, greater parents' perceived illness intrusiveness was related to greater distress across age groups. This finding is consistent with other studies of the relationship between parent-perceived intrusiveness and distress (e.g., Andrews et al., 2007). Studies on parent adjustment to chronic illness largely suggest that these parents are at risk for increased distress and symptoms of depression (for exceptions, see Cadman et al., 1991; Gerhardt et al., 2003) and that adjustment difficulties are affected by a host of cognitive and familial processes, including perceived illness impact (e.g., Sharpe, Brown, Thompson & Eckman, 1994).

This model is perhaps even more clearly relevant to understanding the results of the second primary hypothesis, namely that child age acted as a moderator in the illness intrusiveness-parent distress relationship. More specifically, illness intrusiveness was

related to parent adjustment among parents of older children, but not among parents of older children in this sample. This finding is relevant to Rolland's Family Systems Model, which acknowledges that functioning in the context of a chronic illness is a multigenerational combination of individual, family and illness development (e.g., Rolland, 1987; Rolland & Williams, 2005). He notes that chronic illnesses (such as JRD) that are characterized by flares or relapses require flexibility upon the part of all family members—this is essential because of the necessity of altering family organization and activities to fit the current state of the illness. Therefore, the natural inclination of parents to behave in a more centrifugal, or *disengaging* manner as their children reach adolescence is disrupted by the presence of restrictions imposed by the child's illness. Therefore, the expectation of independence is subsumed under more immediate, illness-related concerns; parent behavior then shifts to a centripetal, or *engaging* type of family organization (Rolland, 1987; Rolland & Williams, 2005). This then disrupts the natural familial progression toward independence and individuation.

Results of this study are therefore consistent with a family lifespan interpretation wherein families of younger children are experiencing the kind of restrictions inherent to parenting any small child—social relationships are embedded within the family, an “engaging” parenting style, physical assistance with tasks that are developmentally above their level. However, for parents of adolescents, the normal developmental process of individuation, of shifting to a more “disengaging” parenting style, has been disrupted. The parent data are consistent with the notion that these parents may be more likely to experience their child's illness-related restrictions as a direct violation of their

expectations for both child and family development, resulting in increased parental distress.

Although a more precise developmental variable could have been used as a moderator rather than age in the present study, Holmes and colleagues (2006) found that that parents of chronically ill children used the age of their child as the primary variable in determining levels of expected responsibility for self-care and *not* demonstrated knowledge or competence. Unfortunately, without the presence of increased knowledge and self-efficacy, older age was actually related to a poorer level of self-care in that study. Apparently, these parents were using age as a proxy for their child's true developmental capabilities. In the present study, a similar mismatch between age and developmental expectation could have accounted for the enhanced effect of illness intrusiveness on distress among parents of older youth. The resulting moderating effect of age may represent the largest illness-induced restriction of all; namely, that children and parents experience difficulties in negotiating typical developmental expectations in an appropriate manner.

Strengths and Limitations

The present study has several strengths. First, this study represents one of the only investigations examining the role of specific parent-reported appraisals of their child's illness in determining parent distress. Given the importance of parent (mal)adjustment to both child and parent physical and psychological outcomes (Thompson et al., 1993 a, b), an analysis of parent-reported illness intrusiveness represents a valuable addition to the pediatric chronic illness literature.

Similarly, while studies have been conducted examining the impact of child age on adjustment and treatment processes (e.g., adherence; see Johnson & Meltzer, 2002, and Weisz & Hawley, 2002 for examples), and on the difficulties of coping with a given chronic illness during the adolescent years (e.g., Coakley et al., 2002). However, the variable impact of child age on parents' cognitive appraisals such as illness intrusiveness has been largely ignored in the pediatric chronic illness literature.

An associated strength of the study is the inclusion of both objective, physician-rated disease severity and subjective, parent-rated perceptions of disease severity. Inclusion of both variables is theoretically important because, although estimates of objective disease severity are necessary to understanding disease processes, they are less important in predicting parent distress relative to subjective ratings of illness by the parent. Especial importance is attached to these subjective ratings when they are tied to parents' perceptions of their role as care provider (Walker, Ford, & Donald, 1987). Because of this, including only parent-report disease severity would likely have resulted in shared method variance. Using physician-report functional class as a covariate accounts for additional variance beyond that accounted for by subjective measures. Indeed, as Sztajn bok and colleagues (2007) note, objective and subjective ratings of functional ability are independently related to outcomes, with parents frequently providing a lower rating of children's health status.

However, this study has multiple limitations as well. With respect to the design of the study itself, self-report inventories were used exclusively to assess both parent adjustment and cognitive appraisals. Thus, the significant association observed between illness intrusiveness (IIS-P) and adjustment (GSI) may have been due to shared method

variance and not to the directional relationships predicted in the hypotheses. However, the fact that parent-reported perceptions of illness intrusiveness did not demonstrate a significant relationship with parent adjustment among the parents of younger children lessens the concern that shared method variance was solely responsible for the observed relationship between these variables among parents of older children in the sample.

A further limitation involves the modest sample size used in this study. A small sample may misrepresent relationships between predictor and criterion variables across the study by including a narrow group of patients' families that were overly homogenous with respect to disease and psychological variables. Families experiencing more distress may have chosen to take part in this study, creating a selection bias. This threat to internal validity is minimized due to the extremely high rate of participation; however, bias could have been introduced by increased clinic attendance among those families experiencing higher levels of intrusion and distress (thus making it more likely that they would be recruited for the study). Further, specific diagnoses were almost universally underrepresented, with JRA comprising over half of the sample. However, it should be noted that this simply indicates low sample sizes for some diagnoses, and not an issue with proportions of children diagnosed with different JRDs. As indicated in the introduction, JRA generally accounts for around 50% of new diagnoses. Nonetheless, generalizability and power are limited for this study due to the modest sample size.

Finally, this study did not incorporate developmental measures, either regarding autonomy and independence, or the developmental general status of the child. Although previous studies have demonstrated that parents of chronically ill children actually base

decisions about self-care and independence primarily on the chronological age of their child (Holmes et al., 2006), such measures could have been included, and would likely have added a greater degree of precision to the interpretation of the data.

Future Studies

Many of these issues could be addressed by future studies. For example, obtaining a greater number of subjects would likely include more a representative sample, especially among the less-common JRD diagnoses. This would then increase generalizability across the population of families of children diagnosed with a JRD. Increasing generalizability could also be achieved by designing the study to decrease selection bias among the participants. This issue could be addressed by designing the study so that it would be possible to compare potential differences between participating and non-participating families. Further design changes could incorporate a variety of assessment methods, rather than relying exclusively on parent-report measures. The incorporation of structured interviews, for example, would alleviate concerns regarding shared method variance (see Chaney et al., 1997).

In terms of measuring developmental level and expectations, a variety of approaches could be utilized. Measures specific to parental expectations, such as the Autonomy scale from the Perceptions of Parents Scale (Grolnick et al, 1991) include items such as “my mother, whenever possible, allows me to choose what to do”. Although this measure (as well as other similar instruments) is child report, information regarding developmental or adaptive abilities could be completed in a parent-report format. Extant measures from the pediatric psychology literature largely relate to children’s competence to manage their own treatment, or to parents’ level of behavioral

monitoring. Although specific to diabetes care, these measures could be adapted for use with JRD. As a recent example, the Parental Monitoring of Diabetes Care Scale (PMDC) has five domains: supervision of medical care (diet, checking blood glucose levels, and insuring adequate medical/pharmacological supplies), parental knowledge of noncompliant episodes, and whether the parents supervised their children via direct oversight (e.g., “How often are you present in the room when your child takes insulin?”; Ellis et al., 2008). Importantly, this measure was used in conjunction with blood glucose meters, as well as parent and adolescent-report versions of the Diabetes Management Scale, which evaluates a range of disease management behaviors (DMS; Frey et al., 2004). Incorporation of similar measures would allow future studies to examine a) whether parents’ perceptions of their child’s independence and self-care abilities vary as a function of chronological age, and b) whether developmental status or perceived autonomy contributes variance to parent distress beyond what is accounted for by age.

Clinical Implications

The findings of the present study highlight the importance of parental perceptions of the intrusiveness of their child’s illness in determining parent distress. More importantly, this study provides data indicating the increased salience of this variable to parent distress among chronically ill children who are transitioning into adolescence and adulthood. Clinical interventions should focus on educating parents on realistic expectations for their child (Power et al., 2003) and in coping with alterations in their child’s developmental trajectory due to illness. These interventions could be especially helpful in a group format focusing on appropriate expectations for children in terms of autonomy, self-care, and other similar concerns. Although not accessible to everyone,

arthritis camps have been shown to assist families in normalizing their experiences with JRD and in developing appropriate goals as well (Hagglund et al., 1996).

REFERENCES

- Anderson, B.J. & Coyne, J.C. (1991). "Miscarried helping" in the families of children and adolescents with chronic diseases. In J.H. Johnson & S.B. Johnson (Eds.), *Advances in Child Health Psychology* (pp. 167-177). Gainesville, FL: University of Florida Press.
- Andrews, N.R., Chaney, J.M., Mullins, L.L., Wagner, J.L., Hommel, K.A., & Jarvis, J.N. (2007). Brief report: Illness intrusiveness and adjustment among Native American and Caucasian parents of children with Juvenile Rheumatic Diseases. *Journal of Pediatric Psychology, 32*, 1259-1263.
- Beanlands, H.J., Lipton, J.H., McCay, E.A., Schimmer, A.D., Elliott, M.E., Messnes, H.A., & Devins, G.M. (2003). Self-concept as a "BMT patient", illness intrusiveness, and engulfment in allogenic bone marrow transplant recipients. *Journal of Psychosomatic Research, 55*, 419-425.
- Cadman, D, Rosenbaum, P., Boyle, M. & Offord, D.R. (1991). Children with chronic illness: Family and parent demographic characteristics and psychosocial adjustment. *Pediatrics, 87*, 884-889.
- Calabro, J.J., Marchesano, J.M., & Parrino, G.R. (1989). Juvenile rheumatoid arthritis: Long-term management and prognosis. *The Journal of Musculoskeletal Medicine, 6*, 17-32.
- Cassidy, J.T., & Petty, R.E. (2001). *The textbook of pediatric rheumatology (4th ed.)*. Philadelphia: W.B. Saunders.

- Cassidy, J.T., & Petty, R.E. (1990). *The textbook of pediatric rheumatology*. New York: Churchill Livingstone.
- Chaney, J.M., Mullins, L.L., Frank, R.G., Peterson, L. Mace, L.D., Kashani, J.H., & Goldstein, D.L. (1997). Transactional patterns of child, mother, and father adjustment in Insulin-dependent diabetes mellitus: A prospective study. *Journal of Pediatric Psychology, 2*, 229-244.
- Chaney, J.M., & Peterson, L. (1989). Family variables and disease management in juvenile rheumatoid arthritis. *Journal of Pediatric Psychology, 13*, 389-403.
- Coakley, R.M., Holmbeck, G.N., Friedman, M.A., Greenley, R.N., & Thill, A.W. (2002). A longitudinal study of pubertal timing, parent-child conflict, and cohesion in families of young adolescents with spina bifida. *Journal of Pediatric Psychology, 25*, 461-473.
- Dahlquist, L.M., Czyzewski, D.I., Copeland, K.G., Jones, C.L., Taub, E. & Vaughan, J.K. (1993). Parents of children newly diagnosed with cancer: Anxiety, coping, and marital distress. *Journal of Pediatric Psychology, 18*, 365-376.
- Daltroy, L.H., Larson, M.G., Eaton, H.M., Partridge, A.J., Pless, I.B., Rogers, M.P., & Liang, M.H. (1992). Psychosocial adjustment in juvenile arthritis. *Journal of Pediatric Psychology, 17*, 277-289.
- Daniels, D., Moos, R.H., Billings, A.G., & Miller, J.J. (1987). Psychosocial risk and resistance factors among children with chronic illness, healthy siblings, and healthy controls. *Journal of Abnormal Child Psychology, 15*, 295-308.
- Davis, C.L., Delamater, A.M., Shaw, K.H, La Greca, A.M., Eidson, M.S., Perez-

- Rodriguez, J.E., & Nemery, R. (2001). Parenting styles, regimen adherence, and glycemic control in 4- to 10-year-old children with diabetes. *Journal of Pediatric Psychology, 26*, 123-129.
- Dekovic, M., Noom, M.J., & Meeus, W. (1997). Expectations regarding development during adolescence: Parental and adolescent perceptions. *Journal of Youth and Adolescence, 26*, 253-272.
- Derogatis, L.R. (1993). *The Brief Symptom Inventory: Administration, Scoring, and Procedures Manual*. Minneapolis, MN: National Computer Systems, Inc.
- Devins, G.M., Binik, Y.M., Hutchinson, T.A., Hollomby, D.J., Barre, P.E. & Guttman, R.D. (1983-1984). The emotional impact of end-stage renal disease: Importance of patients' perception of intrusiveness and control. *International Journal of Psychiatry in Medicine, 13*, 327-343.
- Devins, G.M. & Edworthy, S.M. (2000). Illness intrusiveness explains race-related quality-of-life differences among women with systemic lupus erythematosus. *Lupus, 9*, 534-541.
- Devins, G. M., Edworthy, S. M., Guthrie, N. G., & Martin, L. (1992). Illness intrusiveness in rheumatoid arthritis: Differential impact on depressive symptoms over the adult lifespan. *Journal of Rheumatology, 19*, 709 –715.
- Devins, G.M., Seland, T.P., Klein, G., Edworthy, S.M., & Saary, M.J. (1993). Stability and determinants of psychosocial well-being in multiple sclerosis. *Rehabilitation Psychology, 38*, 11-26.
- Devins, G.M., Stam, H., & Koopmans, J.P. (1994). Psychological impact of

- laryngectomy mediated by perceived stigma and illness intrusiveness. *Canadian Journal of Psychiatry*, 39, 608-616.
- Eisner, C. & Berrenberg, J.L. (1995). Assessing the impact of chronic disease on the relationship between parents and their adolescents. *Journal of Psychosomatic Research*, 39, 109-114.
- Ellis, D.A., Templin, T.N., Podolski, C.L., Frey, M.A., Naar-King, S., & Moltz, K. (2008). The parental monitoring of diabetes care scale: Development, reliability, and validity of a scale to evaluate parental supervision of adolescent illness management. *Journal of Adolescent Health* 42, 146-153.
- Epel, E.S., Blackburn, E.H., Lin, J., Dhabhar, F.S., Adler, N.E., Morrow, J.D., & Cawthon, R.M. (2004). Accelerated telomere shortening in response to life stress. *Proceedings of the National Academy of Sciences*, 101, 17323-17324.
- Flato, B., Aasland, A., Vinje, O., & Forre, O. (1998). Outcome and predictive factors in juvenile rheumatoid arthritis and juvenile spondyloarthropathy. *Journal of Rheumatology*, 25, 366-375.
- Franche, R., Abbey, S., Irvine, J., Shnek, Z.M., Grace, S.L., Devins, G.M., & Stewart, D.E. (2004). Sex differences in predictors of illness intrusiveness 1 year after a cardiac event. *Journal of Psychosomatic Research*, 56, 125-132.
- Frank, R.G., Hagglund, K.J., Schopp, L.H., Thayer, J.F., Vieth, A.Z., Cassidy, J.T., Goldstein, D.E., Beck, N.C., Clay, D.L., Hewett, J.E., Johnson, J.C., Chaney, J.M., & Kashani, J.H. (1998). Disease and family contributors to adaptation in juvenile rheumatoid arthritis and juvenile diabetes. *Arthritis Care and Research*, 11, 166-176.

- Frey, M., Ellis, D., Naar-King, S., & Greger, N. (2004). Diabetes management in adolescents in poor metabolic control. *Diabetes Education, 30*, 647-657.
- Gerhardt, C.A., Vannatta, K., McKellop, M., Zeller, M., Taylor, J., Passo, M. et al. (2003). Comparing parental distress, family functioning, and the role of social support for caregivers with and without a child with juvenile rheumatoid arthritis. *Journal of Pediatric Psychology, 28*, 5-16.
- Grolnick, W.S., Ryan, R.M., & Deci, E.L. (1991). Inner resources for school achievement: Motivational mediators of children's perceptions of their parents. *Journal of Educational Psychology, 83*, 508-517.
- Hagglund, K.J., Doyle, N.M., Clay, D.K., & Frank, R.G., et al. (1996). A family retreat as a comprehensive intervention for children with arthritis and their families. *Arthritis Care and Research, 9*, 35-41.
- Heimlich, T.E., Westbrook, L.E., Austin, J.K., Cramer, J.A., & Devinsky, O. (2000). Brief report: Adolescents' attitudes towards epilepsy: Further validation of the Child Attitude Towards Illness Scale. *Journal of Pediatric Psychology, 25*, 339-345.
- Helgeson, V.S., Janicki, D., & Lerner, J. (2003). Brief Report: Adjustment to Juvenile Rheumatoid Arthritis: A family systems perspective. *Journal of Pediatric Psychology, 28*, 347-353.
- Henderson, C.J., Lovell, D.J., Specker, B.L., & Campaigne, B.N. (1995). Physical activity in children with juvenile rheumatoid arthritis: Quantification and evaluations. *Arthritis Care and Research, 8*, 114-119.
- Hochberg, M.C., Chang, R.W., Dwosh, I., Lindsey, S. Pincus, & T. Wolfe, F. (1992). The

- American College of Rheumatology 1991 revised criteria for the classification of global functional status in rheumatoid arthritis. *Arthritis and Rheumatism*, 35, 498-502.
- Holmbeck, G.N. (2002a). A developmental perspective on adolescent health and illness: An introduction to the Special Issues. *Journal of Pediatric Psychology*, 27, 409-415.
- Holmbeck, G.N. (2002b). Post-hoc probing of significant moderational and mediational effects in studies of pediatric populations. *Journal of Pediatric Psychology*, 27, 87-96.
- Holmbeck, G.N. & Kendall, P.C. (2002). Introduction to the special section on clinical adolescent psychology: Developmental psychopathology and treatment. *Journal of Counseling and Clinical Psychology*, 70, 3-5.
- Holmes, C.S., Chen, R., Streisand, R., Marschall, D.E., Souter, S., Swift, E.E., & Peterson, C.C. (2006). Predictors of youth diabetes care behaviors and metabolic control: A structural equation modeling approach. *Journal of Pediatric Psychology*, 31, 770-784.
- Howe, S., Levinson, J., Shear, E., Hartner, S., McGirr, G., Schulte, M., & Lovell, D. (1991). Development of a disability measurement tool for juvenile rheumatoid arthritis. *Arthritis and Rheumatism*, 34, 873-880.
- Jaworski, T.M. (1993). Juvenile rheumatoid arthritis: Pain-related and psychosocial aspects and their relevance for assessment and treatment. *Arthritis Care and Research*, 6(4), 187-196.
- Johnson, S.B. & Meltzer, L.J. (2002). Disentangling the effects of current age, onset

- age, and disease duration: Parent and child attitudes toward diabetes as an exemplar. *Journal of Pediatric Psychology*, 27(1), 77-86.
- Kahn, M.A. (1993). Seronegative spondylarthropathies: Ankylosing spondylitis. In H.R. Schumacher, Jr., J. H. Klippel, & W.J. Koopman (Eds.), *Primer on the rheumatic diseases: Tenth edition* (pp. 171-174). Atlanta, GA: Arthritis Foundation.
- Kazak, A.E. & Wilcox, B.L. (1984). The structure and function of social support networks in families with handicapped children. *American Journal of Community Psychology*, 12, 645-661.
- Kewman, D.G., Warschausky, S.A., & Engel, L. (1995). Juvenile rheumatoid arthritis and neuromuscular conditions: Scoliosis, spinal cord injury, and muscular dystrophy. In Roberts, M.C. (Eds.), *Handbook of pediatric psychology: Second edition* (pp. 384-402). New York, NY: The Guildford Press.
- Kovacs, M. (1992). *Children's Depression Inventory Manual*. Toronto, Ontario: Multi-Health Systems, Inc.
- Kupst, M.J. & Schulman, J.L. (1988). Long-term coping with pediatric leukemia: A six-year follow-up study. *Journal of Pediatric Psychology*, 13, 7-22.
- Landolt, M.A., Ribl, K., Laimbacher, J., Vollrath, M., Gnehm, H.E., & Sennhauser, F.H. (2002). Posttraumatic stress disorder in parents of children with newly diagnosed Type 1 diabetes. *Journal of Pediatric Psychology*, 27, 647-652.
- LeBovidge, J.S., Lavigne, J.V., & Miller, M.L. (2005). Adjustment to chronic arthritis of childhood: The roles of illness-related stress and attitude toward illness. *Journal of Pediatric Psychology*, 30, 273-286.
- Lehman, T.J.A. (1997). Connective tissue diseases and nonarticular rheumatism. In J. H.

- Klippel (Ed.), *Primer on the rheumatic diseases: Eleventh edition* (pp. 398-403). Atlanta, GA: Arthritis Foundation.
- Levinson, D.J. (1986). A conception of adult development. *American Psychologist, 41*, 3-13.
- Lovell, D.J. (1997). Juvenile rheumatoid arthritis and juvenile spondyloarthropathies. In J. H. Klippel (Ed.), *Primer on the rheumatic diseases: Eleventh edition* (pp. 393-398). Atlanta, GA: Arthritis Foundation.
- Lustig, J.L., Ireys, H.T., Sills, E.M., Walsh, B.B. (1996). Mental health of mothers of children with juvenile rheumatoid arthritis: Appraisal as a mediator. *Journal of Pediatric Psychology, 21*, 719-733.
- Manuel, J.C. (2001). Risk and resistance factors in the adaptation in mothers of children with juvenile rheumatoid arthritis. *Journal of Pediatric Psychology, 26*, 237-246.
- McQuaid, E.L., Kopel, S.J., Klein, R.B., Fritz, G.K. (2003). Medication adherence in pediatric asthma: Reasoning, responsibility, and behavior. *Journal of Pediatric Psychology, 28*, 323-333.
- Mullins, L.L., Cote, M.P., Fuemmeler, B.F., Jean, V.M., Beatty, W.W., & Paul, R.H. (2001). Illness intrusiveness, uncertainty, and distress in individuals with multiple sclerosis. *Rehabilitation Psychology, 46*, 139-153.
- Nagy, S. & Ungerer, J.A. (1990). The adaptation of mothers and fathers to children with cystic fibrosis: A comparison. *Children's Health Care, 19*, 147-154.
- Peterson, L.S., Mason, T., Nelson, A., O'Fallon, M.W., & Gabriel, S.E. (1997). Psychological outcomes and health status of adults who have had juvenile rheumatoid arthritis. *Arthritis and Rheumatism, 40*, 2235-2240.

- Petty, R.E. (1999). Prognosis for children with rheumatic diseases: Justification for consideration of new therapies. *Rheumatology, 38*, 739-742.
- Quittner, A.L., Drotar, D., Iveres-Landis, C., Slocum, N., Seidner, D., & Jacobsen, J. (2000). Adherence to medical treatments in adolescents with cystic fibrosis: The development and evaluation of family-based interventions. In D. Drotar (Ed.), *Promoting Adherence to Medical Treatment in Chronic Childhood Illness: Concepts, Methods, and Interventions* (pp. 383-407). Mahwah, NJ: Lawrence Erlbaum Associates.
- Rhodes, V.J. (1991). Physical therapy management of patients with juvenile rheumatoid arthritis. *Physical Therapy, 71*, 910-919.
- Rolland, J.S. (1987a). Family illness paradigms: Evolution and significance. *Family Systems Medicine, 5*, 482-503.
- Rolland, J.S. (1987b). Chronic illness and the life cycle: A conceptual framework. *Family Process, 26*, 203-221.
- Rolland, J.S. & Williams, J.K. (2005). Toward a biopsychosocial model for 21st-century genetics. *Family Process, 44*, 3-24.
- Sharpe, J.N., Brown, R.T., Thompson, N.J. & Eckman, J. (1994). Predictors of coping with pain in mothers and their children with sickle cell syndrome. *Journal of the American Academy of Child and Adolescent Psychiatry, 33*, 1246-1255.
- Sloper, P. (2000). Predictors of distress in parents of children with cancer: A prospective study. *Journal of Pediatric Psychology, 25*, 79-91.
- Steinberg, L. & Silverberg, S.B. (1986). The vicissitudes of early adolescence. *Child Development, 57*, 841-851.

- Sztajnbok, F., Coronel-Martinez, D.L., Diaz-Maldonado, A., Novarini, C., Pistorio, A., Viola, S., Ruperto, N., Buoncompagni, A., Martini, A., & Ravelli, A. (2007). Discordance between physician's and parent's global assessments in juvenile idiopathic arthritis. *Rheumatology*, *46*, 141-145.
- Thomasgard, M. (1998). Parental perceptions of child vulnerability, overprotection, and parental psychological characteristics. *Child Psychiatry and Human Development*, *28*, 223-240.
- Thompson, R.J, Gil, K.M., Burbach, D.J., Keith, B.R., & Kinney, T.R. (1993a). Psychological adjustment of mothers of children and adolescents with sickle cell disease: The role of stress, coping methods, and family functioning. *Journal of Pediatric Psychology*, *18*, 549-559.
- Thompson, R.J, Gil, K.M., Burbach, D.J., Keith, B.R., & Kinney, T.R. (1993b). Role of child and maternal processes in the psychological adjustment of children with sickle cell disease. *Journal of Consulting and Clinical Psychology*, *61*, 468-474.
- Thompson, R.J, Gil, K.M., Gustafson, K.E., George, L.K., Keith, B.R., Spock, A. & Kinney, T.R. (1994). Stability and change in the psychological adjustment of mothers of children and adolescents with cystic fibrosis and sickle cell disease. *Journal of Pediatric Psychology*, *19*, 171-188.
- Thompson, R.J., Gustafson, K.E., Hamlett, K.W., & Spock, A. (1992). Psychological adjustment of children with cystic fibrosis: The role of child cognitive processes and maternal adjustment. *Journal of Pediatric Psychology*, *17*, 741-755.
- Timko, C., Baumgartner, M., Moos, R.H. & Miller, J.J. (1993). Parental risk and

- resistance factors among children with juvenile rheumatic disease: A four-year predictive study. *Journal of Behavioral Medicine*, 16, 571-588.
- Timko, C., Stovel, K.W., Moos, R.H. & Miller, J.J. (1992a). A longitudinal study of risk and resistance factors among children with juvenile rheumatic disease. *Journal of Clinical Child Psychology*, 21, 132-142.
- Timko, C., Stovel, K.W. & Moos, R.H. (1992b). Functioning among mothers and fathers of children with juvenile rheumatic disease: A longitudinal study. *Journal of Pediatric Psychology*, 17, 705-724.
- Ungerer, J.A., Horgan, B., Chaitow, J., & Champion, G.D. (1988). Psychosocial functioning in children and young adults with juvenile arthritis. *Journal of Pediatrics*, 81, 195-202.
- Vandvik, I.H. & Hoyeraal, H.M. (1993). Juvenile chronic arthritis: a biobehavioral disease. Some unsolved questions. *Clinical and Experimental Rheumatology*, 11, 669-680.
- Vandvik, I.H., Hoyeraal, H.M. & Fagertun, H. (1989). Chronic family difficulties and stressful life events in recent onset juvenile arthritis. *The Journal of Rheumatology*, 16, 1088-1092.
- Wagner, J.L., Chaney, J.M., Hommel, K.A., Page, M.C., Mullins, L.L., White, M.M., & Jarvis, J.N. (2003). The influence on parental distress on child depressive symptoms in Juvenile Rheumatic Diseases: The moderating effect of illness intrusiveness. *Journal of Pediatric Psychology*, 28, 453-462.
- Walker, L.S., Ford, M.B. & Donald, W.D. (1987). Cystic fibrosis and family stress: Effects of age and severity of illness. *Pediatrics*, 79, 239-246.

- Wallander, J.L., Thompson, R.J., & Alriksson-Schmidt, A. (2003). Psychosocial adjustment of children with chronic physical conditions. In M.C. Roberts (Ed.), *Handbook of Pediatric Psychology: Third edition* (pp. 141-158). New York: Guilford Press.
- Wallander, J.L., & Varni, J.W. (1989). Social support and adjustment in chronically ill and handicapped children. *American Journal of Community Psychology, 17*, 185-201.
- Wallston, K.A., Wallston, B.S., & Devellis, R. (1978). Development of the Multidimensional Health Locus of Control (MHLC) scales. *Health Education Monographs, 6*, 160-170.
- Weisz, J.R. & Hawley, J.M. (2002). Developmental factors in the treatment of adolescents. *Journal of Consulting and Clinical Psychology, 70*, 21-43.
- Williams, P.G., Holmbeck, G.N., & Greenley, R.N. (2002). Adolescent Health Psychology. *Journal of Counseling and Clinical Psychology, 70*, 828-842.
- Young, L.D. (1992). Psychological factors in rheumatoid arthritis. *Journal of Consulting and Clinical Psychology, 60*, 619-627.

APPENDICES

Appendix A

Brief Symptom Inventory (BSI)

	NOT AT ALL	A LITTLE BIT	MODERATELY	QUITE A BIT	EXTREMELY	
1	1	2	3	4	5	Nervousness or shakiness inside
2	1	2	3	4	5	Faintness or dizziness
3	1	2	3	4	5	The idea that someone else can control your thoughts
4	1	2	3	4	5	Feeling others are to blame for most of your troubles
5	1	2	3	4	5	Trouble remembering things
6	1	2	3	4	5	Feeling easily annoyed or irritated
7	1	2	3	4	5	Pains in heart or chest
8	1	2	3	4	5	Feeling afraid in open spaces or on the streets
9	1	2	3	4	5	Thoughts of ending your life
10	1	2	3	4	5	Feeling that most people cannot be trusted
11	1	2	3	4	5	Poor appetite
12	1	2	3	4	5	Suddenly scared for no reason
13	1	2	3	4	5	Temper outbursts that you could not control
14	1	2	3	4	5	Feeling lonely even when you are with people
15	1	2	3	4	5	Feeling blocked in getting things done
16	1	2	3	4	5	Feeling lonely
17	1	2	3	4	5	Feeling blue
18	1	2	3	4	5	Feeling no interest in things
19	1	2	3	4	5	Feeling fearful
20	1	2	3	4	5	Your feelings being easily hurt
21	1	2	3	4	5	Feeling that people are unfriendly or dislike you
22	1	2	3	4	5	Feeling inferior to others
23	1	2	3	4	5	Nausea or upset stomach
24	1	2	3	4	5	Feeling that you are watched or talked about by others
25	1	2	3	4	5	Trouble falling asleep
26	1	2	3	4	5	Having to check and double-check what you do
27	1	2	3	4	5	Difficulty making decisions
28	1	2	3	4	5	Feeling afraid to travel on buses, subways, or trains
29	1	2	3	4	5	Trouble getting your breath
30	1	2	3	4	5	Hot or cold spells
31	1	2	3	4	5	Having to avoid certain things, places, or activities because they frighten you
32	1	2	3	4	5	Your mind going blank
33	1	2	3	4	5	Numbness or tingling in parts of your body
34	1	2	3	4	5	The idea that you should be punished for your sins
35	1	2	3	4	5	Feeling hopeless about the future
36	1	2	3	4	5	Trouble concentrating
37	1	2	3	4	5	Feeling weak in parts of your body
38	1	2	3	4	5	Feeling tense or keyed up
39	1	2	3	4	5	Thoughts of death or dying
40	1	2	3	4	5	Having urges to beat, injure, or harm someone
41	1	2	3	4	5	Having urges to break or smash things
42	1	2	3	4	5	Feeling very self-conscious with others
43	1	2	3	4	5	Feeling uneasy in crowds, such as shopping or at a movie
44	1	2	3	4	5	Never feeling close to another person
45	1	2	3	4	5	Spells of terror or panic
46	1	2	3	4	5	Getting into frequent arguments
47	1	2	3	4	5	Feeling nervous when you are left alone
48	1	2	3	4	5	Others not giving you proper credit for your achievements
49	1	2	3	4	5	Feeling so restless you couldn't sit still
50	1	2	3	4	5	Feelings of worthlessness
51	1	2	3	4	5	Feeling that people will take advantage of you if you let them
52	1	2	3	4	5	Feelings of guilt
53	1	2	3	4	5	The idea that something is wrong with your mind

Appendix B

Illness Intrusiveness Scale-Parent (IIS-P)

IIS-Parent

Name _____

For each of the items below, rate the extent to which your child's illness "interferes with" your ability to perform as well as you would like to. (Circle the Number for each item.)

	A Little					A Lot	
1. Work	1	2	3	4	5	6	7
2. Active recreation (e.g., golf, tennis)	1	2	3	4	5	6	7
3. Passive recreation (e.g., playing cards)	1	2	3	4	5	6	7
4. Financial status	1	2	3	4	5	6	7
5. Relationship with your spouse/lover	1	2	3	4	5	6	7
6. Sex Life	1	2	3	4	5	6	7
7. Relationships with your family	1	2	3	4	5	6	7
8. Relationships with other persons	1	2	3	4	5	6	7
9. Self-expression/ self-improvement	1	2	3	4	5	6	7
10. Religious expression	1	2	3	4	5	6	7
11. Community/civic involvement	1	2	3	4	5	6	7
12. Health	1	2	3	4	5	6	7
13. Diet	1	2	3	4	5	6	7

Appendix C

The Juvenile Arthritis Functional Assessment Report for Parents (JAFAR-P)

Over the past week, how often has your child been able to perform each of the activities in the list below?

	All the time	Sometimes	Almost never
1. Take shirt off hanger	_____	_____	_____
2. Button shirt	_____	_____	_____
3. Pull on sweater over head	_____	_____	_____
4. Turn on water faucet	_____	_____	_____
5. Climb into bathtub	_____	_____	_____
6. Dry back with towel	_____	_____	_____
7. Wash face with washcloth	_____	_____	_____
8. Tie shoelaces	_____	_____	_____
9. Pull on socks	_____	_____	_____
10. Brush teeth	_____	_____	_____
11. Stand up from chair without using arms	_____	_____	_____
12. Get into bed	_____	_____	_____
13. Cut food with knife and fork	_____	_____	_____
14. Lift empty glass to mouth	_____	_____	_____
15. Reopen previously opened food jar	_____	_____	_____
16. Walk 50 feet without help	_____	_____	_____
17. Walk up 5 steps	_____	_____	_____
18. Stand up on tiptoes	_____	_____	_____
19. Reach above head	_____	_____	_____
20. Get out of bed	_____	_____	_____
21. Pick up something from floor from standing position	_____	_____	_____
22. Push open door after turning knob	_____	_____	_____
23. Turn head and look over shoulder	_____	_____	_____

Appendix D

Children's Depression Inventory

Kids sometimes have different feelings and ideas.

This form lists the feelings and ideas in groups. From each group, pick one sentence that describes you best for the past two weeks. After you pick a sentence from the first group, go on to the next group.

There is no right answer or wrong answer. Just pick the sentence that best describes the way you have been recently. Put a mark like this X next to your answer. Put the mark in the box next to the sentence that you pick.

Here is an example of how this form works. Try it. Put a mark next to the sentence that describes you best.

EXAMPLE:

- | | |
|-------|------------------------------|
| _____ | I read books all the time |
| _____ | I read books once in a while |
| _____ | I never read books |

Remember, pick out the sentence that describes your feelings and ideas in the PAST TWO WEEKS.

- | | |
|-------|--------------------------|
| _____ | I am sad once in a while |
| _____ | I am sad many times |
| _____ | I am sad all the time |
- | | |
|-------|--|
| _____ | Nothing will work out for me |
| _____ | I am not sure if things will work out for me |
| _____ | Things will work out for me O.K. |
- | | |
|-------|-----------------------|
| _____ | I do most things O.K. |
|-------|-----------------------|

- _____ I do many things wrong
- _____ I do everything wrong
4. _____ I have fun in many things
- _____ I have fun in some things
- _____ Nothing is fun at all
5. _____ I am bad all the time
- _____ I am bad many times
- _____ I am bad once in a while
6. _____ I think about bad things happening to me once in a while
- _____ I worry that bad things will happen to me
- _____ I am sure that terrible things will happen to me
7. _____ I hate myself
- _____ I do not like myself
- _____ I like myself
8. _____ All bad things are my fault
- _____ Many bad things are my fault
- _____ Bad things are not usually my fault
9. _____ I do not think about killing myself
- _____ I think about killing myself but I would not do it
- _____ I want to kill myself

10. _____ I feel like crying every day
_____ I feel like crying many days
_____ I feel like crying once in a while
11. _____ Things bother me all the time
_____ Things bother me many times
_____ Things bother me once in a while
12. _____ I like being with people
_____ I do not like being with people many times
_____ I do not want to be with people at all
13. _____ I cannot make up my mind about things
_____ It is hard to make up my mind about things
_____ I make up my mind about things easily
14. _____ I look O.K.
_____ There are some bad things about my looks
_____ I look ugly
15. _____ I have to push myself all the time to do my school work
_____ I have to push myself many times to do my school work
_____ Doing school work is not a big problem
16. _____ I have trouble sleeping every night
_____ I have trouble sleeping many nights

- _____ I sleep pretty well
17. _____ I am tired once in a while
_____ I am tired many days
_____ I am tired all the time
18. _____ Most days I do not feel like eating
_____ Many days I do not feel like eating
_____ I eat pretty well
19. _____ I do not worry about aches and pains
_____ I worry about aches and pains many times
_____ I worry about aches and pains all the time
20. _____ I do not feel alone
_____ I feel alone many times
_____ I feel alone all the time
21. _____ I never have fun at school
_____ I have fun at school only once in a while
_____ I have fun at school many times
22. _____ I have plenty of friends
_____ I have some friends but I wish I had more
_____ I do not have any friends

23. _____ My school work is all right
_____ My school work is not as good as before
_____ I do very badly in subjects I used to be good in
24. _____ I can never be as good as other kids
_____ I can be as good as other kids if I want to
_____ I am just as good as other kids
25. _____ Nobody really loves me
_____ I am not sure if anybody loves me
_____ I am sure that somebody loves me
26. _____ I usually do what I am told
_____ I do not do what I am told most times
_____ I never do what I am told
27. _____ I get along with people
_____ I get into fights many times
_____ I get into fights all the time

THE END

THANK YOU FOR FILLING OUT THIS FORM

Appendix E

Subject # _____

Date: _____

Provider Questionnaire

1. Patient's name: _____
2. Patient's Diagnosis (if multiple diagnoses, please list 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	5	6	7
Not Active or In Remission		Mild		Moderate		Severe

6. 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

7. 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, & 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 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):**

Nicole Andrews
215 North Murray
Stillwater, OK 74078
Molly White
407 N. Murray
Stillwater, OK 74078

Janelle Wagner
215 N. Murray
Stillwater, OK 74078
John M. Chaney
215 N. Murray
Stillwater, OK 74078

James Jarvis
OUHSC
Oklahoma City, OK 73104

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, Chair
Institutional Review Board

Appendix G

University of Oklahoma Health Sciences Center
Dr. James N. Jarvis

- Consent Form

This is a research study being conducted at the University of Oklahoma Health Sciences Center. Research studies involve only individuals who choose to participate. Please take your time to make your decision. Discuss this with your family and friends. You are being asked to take part in this study because your child has a diagnosis of a juvenile rheumatic disease.

I, _____ (name of participant's parent/legal guardian), voluntarily consent to allow my child to participate in the investigation of psychological factors and juvenile rheumatic diseases (JRD).

WHY IS THIS STUDY BEING DONE?

The purpose of the study is to examine psychological factors associated with JRD disease processes.

HOW MANY PEOPLE WILL TAKE PART IN THIS STUDY?

About 100 children and caregivers will participate in this study.

WHAT IS INVOLVED IN THIS STUDY?

The research requires the completion of several paper-and-pencil measures in the Pediatric Rheumatology Clinic at the Children's Hospital of Oklahoma that address psychological factors and perceptions of life events, both in general and with respect to JRD. Some items on the questionnaires contain sensitive issues (e.g., depression, relationships, etc.)

My child's participation is voluntary; there is no penalty for refusal to participate, and my child is free to withdraw his/her consent and participation in this project at any time without penalty, after notifying the project director.

WHAT ARE THE RISKS OF THE STUDY?

There is virtually no risk associated with completing questionnaires. It is possible that your child may experience some negative emotions during the completion of the questionnaires, but these will be short-lived and have no long-term effects.

WHAT ARE THE BENEFITS OF TAKING PART IN THE STUDY?

Although my child's participation may not necessarily be personally beneficial to my child, the information derived from this project may have important implications for others who have JRD. The information gained may contribute to a better understanding of the cognitive/emotional functioning and overall treatment of individuals with JRD.

WHAT ABOUT CONFIDENTIALITY?

Any data collected as part of my child's participation in this experiment will be treated as confidential and will receive a code number so that they will remain confidential. In no case will any use be made of these data other than as research results. If data from my child's participation is ever displayed, my child's identity will remain confidential. You will be asked to sign a separate authorization form for use or sharing of your protected health information.

WHAT ARE THE COSTS?

There are no costs to your child for participation in this study.

WILL I BE PAID FOR PARTICIPATING IN THIS STUDY?

I understand that my child and I will receive \$10.00 compensation in the form of gift certificates for approximately one hour of participation, and there is no risk of injury as a result of this study.

I may contact Dr. John M. Chaney, Oklahoma State University, Psychology Department, 215 North Murray Hall, Stillwater, Oklahoma 74078, at (405) 744-5703 should I wish further information about the research. I may also contact the Institutional Review Board (IRB) executive assistant, Sharon Bacher, Oklahoma State University, 203 Whitehurst, Stillwater, Oklahoma 74078, (405) 744-5700. Should any problems arise during the course of the study I may take them to Dr. Maureen Sullivan, Psychological Department Head, Oklahoma State University, Department of Psychology, 215 North Murray Hall, Stillwater, Oklahoma, 74078, at (405) 744-6027.

I have read and fully understand the consent form, and the option to receive a copy of this consent form has been give to me. I sign it freely and voluntarily.

Signed: _____ Date: _____ Time _____ (A.M./P.M.)
(Signature of participant's parent/legal guardian)

Child's Assent _____ Witness(es) if required: _____

I certify that I have personally explained all elements of this form to the subject before requesting the subject to sign it.

Signed _____
(Project director or his/her authorized representative)

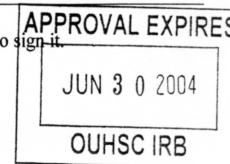
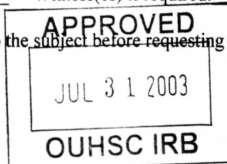


Table 1.

Disease, Demographic, and Psychosocial Variables: Means

Variables	Parents of Older Youth		Parents of Younger Youth	
	<u>M (SD)</u>	<u>Range</u>	<u>M (SD)</u>	<u>Range</u>
Child's Age	15.48 (1.12)	14-17	11.19 (1.25)	9-13
Duration	2.57 (3.81)	0-15.73	2.50 (2.42)	.16-7.70
PRFD	1.38 (0.50)	1-2	1.67 (0.73)	1-3
JAFAR-P	4.08 (5.19)	0-18	5.52 (7.16)	0-22
CDI	10.20 (9.60)	0-44	6.52 (5.61)	1-21
IIS-P	22.60 (13.27)	12-55	23.52 (16.29)	12-67
GSI	0.67 (.67)	0-3.13	0.45 (0.36)	.04-1.32

Variables	Total Sample	
	<u>M (SD)</u>	<u>Range</u>
Child's Age	13.75 (2.42)	9-17
Duration	2.55 (3.29)	0-15.73
PRFD	1.50 (.61)	1-3
JAFAR-P	4.47 (6.04)	0-22
CDI	8.69 (8.33)	0-44
IIS-P	22.97 (14.42)	12-67
GSI	.58 (.57)	.00-3.13

Note: PRFD = Physician-rated functional disability; Duration = Disease Years; JAFAR-P = Juvenile Arthritis Functional Assessment Report; CDI = Child Depression Inventory Total; IIS-P = Illness Intrusiveness Scale – Parent; GSI = Global Severity Index.

Table 2.

Hierarchical Regression Analyses of Parent-Reported Illness Intrusiveness on GSI

Step	Variable	β	t	R ² Change	Cumulative R ²	F Change
1	Diagnosis	0.04	0.31	.24	.24	1.93
	PRFD	-0.09	-0.61			
	Gender	0.01	0.06			
	Ethnicity	0.03	0.24			
	Duration	0.10	0.72			
	JAFAR-P	0.28	1.80			
	CDI	0.40	2.78**			
2	IIS-P	0.50	3.06***	.14	.38	9.34***

Note: PRFD = Physician-rated functional disability; Duration = Disease Years; JAFAR-P = Juvenile Arthritis Functional Assessment Report; CDI = Child Depression Inventory Total; IIS-P = Illness Intrusiveness Scale – Parent; GSI = Global Severity Index.

* $p < .05$ ** $p < .01$ *** $p < .005$

Table 3.

Hierarchical Regression Analyses of Parent-Reported Illness Intrusiveness on GSI as moderated by Child Age

Step	Variable	β	t	R ² Change	Cumulative R ²	F Change
1	Diagnosis	0.04	0.31	.24	.24	1.93
	PRFD	-0.09	-0.61			
	Gender	0.01	0.06			
	Ethnicity	0.03	0.24			
	Duration	0.10	0.72			
	JAFAR-P	0.28	1.80			
	CDI	0.40	2.78**			
2	Age	0.26	1.90	.19	.43	6.77***
	IIS-P	0.53	3.31***			
3	IIS-P X Age	1.34	2.07*	.06	.49	4.26***

Note: PRFD = Physician-rated functional disability; Duration = Disease Years; JAFAR-P = Juvenile Arthritis Functional Assessment Report; CDI = Child Depression Inventory Total; IIS-P = Illness Intrusiveness Scale – Parent; GSI = Global Severity Index.

* $p < .05$ ** $p < .01$ *** $p < .005$

VITA

Nicole R. Andrews

Candidate for the Degree of

Doctor of Philosophy

Thesis: CHILDREN'S AGE AS A POTENTIAL MODERATOR OF THE
ILLNESS INTRUSIVENESS-DISTRESS RELATIONSHIP IN JUVENILE
RHEUMATIC DISEASE

Major Field: Psychology

Biographical

Education: Graduated from Putnam City West High School, Oklahoma City, OK, May, 1999; Received Bachelor of Science degree in Psychology and graduated Magna Cum Laude from the University of Oklahoma, Norman, OK, July, 2002; Completed the requirements for the Doctor of Philosophy degree in Psychology at Oklahoma State University, December, 2009.

Experience: Clinical practica experience through OSU Psychological Services Center, August 2002-July 2005; Clinical practica experiences through the A Better Chance (ABC), pediatric Hematology/Oncology, and adult Neuropsychology clinics at the University of Oklahoma Health Sciences Center, June 2004-September 2009; Clinical practica experience at the Veterans Affairs Medical Center of Oklahoma City, September 2006-August 2007; Predoctoral internship at the Mailman Center for Child Development at the University of Miami, September 2008-September 2009; Postdoctoral fellowship at the Hospital for Sick Children in Toronto, Ontario, September 2009-September 2011; Instructor of Introductory Psychology, August 2003-June 2004; Teaching Assistant, August 2002-June 2003; Research Assistant in Dr. John Chaney's Health Psychology Research Lab at OSU, August 2002-December 2009; Outcome Evaluator at the Infant Parenting Program at the University of Oklahoma Health Sciences Center; Undergraduate honors psychology research at the University of Oklahoma, August 2001-May 2002.

Name: Nicole Andrews

Date of Degree: December, 2009

Institution: Oklahoma State University

Location: Stillwater, Oklahoma

Title of Study: CHILDREN'S AGE AS A POTENTIAL MODERATOR OF THE
ILLNESS INTRUSIVENESS-DISTRESS RELATIONSHIP IN
JUVENILE RHEUMATIC DISEASE

Pages in Study: 77

Candidate for the degree of Doctor of Philosophy

Major Field: Clinical Psychology

This study examines possible differences in coping among the parents of children with Juvenile Rheumatoid Disease (JRD). The vast majority of research on illness intrusiveness in pediatric populations has focused exclusively on children's perceptions of intrusiveness and its relation to child adjustment—extant literature has largely ignored parents' intrusiveness perceptions as a determinant of their own distress levels. This study investigates the illness intrusiveness-parental distress relationship among parents of children diagnosed with a JRD, and how the age of the child affects that relationship. In other words, the potential moderating role of child age on the illness intrusiveness-parental distress relationship is examined. Results demonstrated a significant main effect for parents of all children in the sample. However, while illness intrusiveness was found to be significantly related to distress among parents of older children, only a marginal relationship was seen among parents of younger children.

Advisor's Approval: John M. Chaney, Ph.D.