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STRESS, FAMILY COPING AND ADJUSTMENT IN ADOLESCENTS WITH JUVENILE RHEUMATOID ARTHRITIS (JRA)

by

PAMELA BUBOLO DEGOTARDI

A dissertation submitted to the Graduate Faculty in Psychology in partial fulfillment of the requirements for the degree of Doctor of Philosophy, The City University of New York

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Abstract

STRESS, FAMILY COPING AND ADJUSTMENT IN ADOLESCENTS WITH JUVENILE RHEUMATOID ARTHRITIS (JRA)

by

Pamela Bubolo Degotardi

Advisor: Professor Tracey A. Revenson

This study examined how family-level coping affects adjustment for families who have an adolescent with juvenile rheumatoid arthritis (JRA). Drawing from family systems theory and a developmental perspective, I proposed a multivariate model of how disease severity, family demographics, and perceived stress influence adjustment outcomes. Family coping and developmental stage were posed as moderators of this relationship.

Following a routine clinic visit the families of 35 early adolescents (10 – 13 years), and 33 late adolescents (14 – 19 years) with JRA were interviewed using a semi-structured protocol. The interview explored family narratives of disease onset, response to JRA-specific stressors, and adaptation across several domains (school, peer group). Interviews were coded for endorsement of 22 family-level coping strategies, using the FCCS (Hauser et. al., 1993), and reduced through factor analysis to five scales: Team Effort, Status Quo, Emotion Processing, Cognitive Flexibility, Seeking Meaning. Following the interview, parents and adolescents completed separate self-report measures of stress and adjustment.

Overall, parents reported significantly more stress than their children. Parents reported concerns regarding their child's future, whereas, adolescents focused on

immediate concerns, e.g., pain, activity restrictions. Adolescents' (but not parents') reports of JRA-specific stress were strongly related to concrete indicators of disease severity (pain, functional limitations). Stress was also associated with poorer adherence, increased internalizing, lower quality of life, and greater parental depression.

Families used many coping strategies (M= 12). Disease severity, stress and family coping were good predictors of most adjustment outcomes, explaining between 44% (internalizing) and 82% (quality of life) of the variance. Moreover, coping moderated the relationship between disease severity and adjustment: Although stress was inversely related to adjustment, at levels of high stress family coping acted as a stress-buffer. In particular, family use of the strategies of team effort or seeking meaning was related to improved quality of life, and efforts to maintain the status quo were related to improved adherence.

Surprisingly, there were few differences between early and late adolescents, and no interactions between the child's age and length of time since diagnosis on any measures. This study demonstrated the utility of a structured family interview for assessing family-level coping and highlighted the primacy of cognitive appraisals of the illness experience in the psychosocial adjustment of adolescents with JRA.

Acknowledgements

I am deeply grateful to the Arthritis Foundation for supporting my dissertation research. By awarding me a Student Summer Fellowship in June 1996, the New York Chapter was instrumental in cultivating my interest in juvenile rheumatoid arthritis. This grant provided the means to begin collecting pilot data. Although I was unsuccessful in my first application for a dissertation grant, extensive feedback from two anonymous reviewers helped to shape (and improve) my proposed research. I incorporated these suggested changes into my final study design and in 1998 I was awarded a two-year Doctoral Dissertation Award for Health Professional from the Arthritis Foundation. As well as validating my research by providing financial support, the Arthritis Foundation has provided a forum to present my findings at national conferences and in arthritis publications. I feel that I have found my research home and I look forward to continuing pediatric arthritis research.

For the past three years, my research has been based at Schneider Children's Hospital under the auspices of Dr. Norman Ilowite (Chief, Division of Pediatric Rheumatology). I am indebted to both Dr. Ilowite and Dr. Beth Gottlieb for teaching me about the medical aspects of JRA, listening to my research ideas, and allowing me to interrupt clinic schedules to interview JRA families. Although at times I felt that I was being a "royal pain" by commandeering scarce office space, monopolizing the secretary's time, and constantly asking questions, the entire pediatric rheumatology team unfailingly supported my research and cheered even minor accomplishments. I am also grateful to all the families who so willingly shared their arthritis stories.

Over the past three years I have been fortunate to develop wonderful professional relationships with many strong, intelligent and compassionate women at Schneider Children's Hospital – especially, Dr. Emily Klass. The friendship, advice, support, shared laughter (and lunches) made the last few years of this dissertation journey fun.

In spite of two major relocations, the community of faculty and students at the CUNY Graduate Center provided a safe forum to debate and explore the unanswerable. Drs. Katherine Nelson, Herb Saltzstein and Joe Glick provided just the right combination of challenge and support, so that I was inspired to question and explore slippery issues of process. My dissertation committee helped frame my questions, and offered a sounding board for research ideas. My friends in dissertation group kept me on track, assuring me that they too had "no clue" that first semester. Together we conquered the *second-year docs* and inspired each other to complete the dissertation marathon.

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My family offered a continual reminder of why family context is important – yes, the stressors of dissertation research did impact the entire family unit. As best they could, my husband and son tried to *normalize* this dissertation experience, and through concerted *team effort* we not only realized our dream but we *thrived*! I couldn't have done this on my own – Michael and Zach, your love and support made this possible.

Dedicated to the loving memory of my mother, who constantly talked about the world of possibilities beyond the pineapple patch....

Lillian Joan Degotardi

1922 - 1989

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CHAPTER 1: INTRODUCTION

Adjustment to a chronic childhood illness, juvenile rheumatoid arthritis (JRA) will be examined in this study within the context of family coping and the child's developmental status. Family coping is as yet 'uncharted territory' in the quest to discover factors that promote the child's adjustment to the stressors of living with a chronic illness such as JRA. Disease characteristics (such as type and severity of illness) and functional limitations that disrupt daily activities have been linked with adjustment; this study proposes that family stress and family coping strategies modify that relationship.

All families with a chronically ill child must adopt strategies and routines that enable them to deal effectively with illness-related stressors. These stressors include frequent medical appointments, invasive medical procedures, pain management, hospitalization, and disruptions to daily routines. In addition to these common illness-related tasks, children with JRA must deal with joint pain and inflammation, morning stiffness, fatigue and irritability, increased sleep requirements, and treatment regimens that often include a combination of pharmacological, physical and occupational therapy. One of the characteristics of JRA is the unpredictability of disease progression. Children can experience "flares" when arthritis symptoms are intense, and they may also experience protracted periods of symptom remission (Cassidy, Levinson & Brewer, 1989; Cassidy & Petty , 1995). Thus, treatment complexity, pain, and unpredictable illness course all contribute to the stress experienced by families of children with JRA.

There have been equivocal findings regarding the psychosocial functioning of children with JRA. Most research has focused on the psychological distress experienced

by the child. In this way it has focused on the patient without attention to the familial context in which the illness is treated and experienced. Family systems theory suggests that all family members are affected by changes in the health or functioning of individual members (Patterson & Garwick, 1994). In fact, a child's chronic illness upsets existing structures within the family, propelling changes in family roles, rituals, and daily routines that help to restore equilibrium.

Although it is apparent to health care professionals that some families are more successful than others in dealing with the demands of JRA (Cole & Reiss, 1993), there has been no systematic study of the type of coping strategies that are associated with better adjustment of children and their families. This study will document both the level of perceived family stress and types of family coping strategies used by JRA families, and examine how these are differentially related to adherence and psychological well-being.

A critical review of current studies of adjustment to JRA reveals several methodological and conceptual flaws. First, these studies rely almost exclusively on the self-reports of mothers; rarely are fathers or siblings included. Second, the research designs generally have been limited to brief self-report questionnaires that constrain the amount and quality of data collected. Third, most of the research does not draw on the rich literature in developmental or pediatric psychology, where development is viewed both in terms of the child's life stage and in term of the developmental progression of the illness. Children's ages reflect vast differences in their cognitive capacity to understand and find meaning in a life challenge, such as JRA. However, most JRA studies have included a wide age range (typically 4 to 17 years) and ignore age-specific developmental

tasks and changes related to disease progression that affect coping and adjustment (Roberts & Wallander, 1992).

Clearly, current research on children with JRA lacks a developmental perspective and overlooks family context. This study addresses these shortcomings by using both qualitative and quantitative methodologies. The combination of the two provides a deeper understanding of the psychosocial issues that are important for family functioning, and allow me to document how the adolescents' developmental status and family circumstances influence perception of stress, use of family coping strategies, and adjustment.

Role of the Family in JRA

Most research on adjustment to JRA has focused on the individual child, overlooking the contributions of family context. Although a philosophy of family-centered care has pervaded pediatric psychology practice, research has yet to catch up. Typically, the ill child is the subject of inquiry, but the illness is not viewed within the broader context of either family, social (e.g., school), or health-care systems. The impact of the illness on family dynamics and functioning has not been explored adequately (Fiese & Sameroff, 1992; Roberts & Wallander, 1992).

The family plays a major role in the management of a chronic pediatric illness, with parents providing both instrumental and emotional support for the ill child (Cole & Reiss, 1993). Parents typically decide when, where, and from whom to seek medical treatment when symptoms are first noticed, and in most families the mother has primary responsibility for monitoring adherence to treatment. Pediatric chronic illness may either strengthen family relationships or disrupt family functioning. Furthermore, dysfunctional

family interaction prior to illness is a predictor of problematic adjustment (Gervasio, 1986; LaGreca & Schuman, 1995).

There are multiple and reciprocal influences among parents, children and health care systems. Flexibility in dealing with complex illness-related demands and challenges is essential for optimal family functioning. Early family systems theorists viewed a child's chronic illness as a perturbation to the family system that necessitated changes to restore equilibrium in the family (Minuchin, 1974). These changes included adjustments in family paradigms (e.g., changes in expectations of the child and assessment of family functioning), the retelling of family stories or myths (especially with regard to the illness of other family members), and the reevaluation of past experiences of coping (Fiese & Sameroff, 1992).

Each family system has unique beliefs and rules regarding the health, development, and illness of family members. These illness attributions influence families' construal of and response to symptom onset (Kazak, Segal-Andrews, & Johnson, 1995). In families when a child develops arthritis symptoms there may be changes in family identity as families construct shared meanings of JRA. Construals and reinterpretations of JRA demands may have either positive or negative biases, with positive appraisals associated with better adaptation. When JRA is construed as a challenge there may be growth or enhancement of family functioning (O'Leary & Ickovics, 1995). Positive family changes may include growth in the child's tenacity and willingness to endure unpleasant procedures such as joint aspiration, and increased empathy and kindness of siblings (Patterson & Garwick, 1994).

Although the stressors of a child's chronic illness initially may disrupt the

family's developmental trajectory and temporarily upset family functioning, balance to the family system is restored over time as the family adjusts to the demands of the illness and its treatment. At the onset of symptoms and during the initial acute phase, well-established family routines and rituals may be compromised. The care-giving system becomes organized around technical aspects of the illness, including seeking appropriate medical care, and learning about JRA and treatment administration. Family energy is focused on the ill family member and his or her special needs. The issue of "loss" may become paramount (Kazak, Segal-Andrews & Johnson, 1995). For children with JRA, there may be loss of normal functioning (e.g., restriction of activities), loss of future dreams (e.g., athletic prowess, career possibilities), loss of established patterns of family routines, and loss of personal control as children experience unpredictable arthritis flares. Families perceive and grieve these losses in a variety of ways. Some families may become locked into cycles of empathy-resentment-guilt (Konkol et. al., 1989), with parents and siblings feeling empathy for the ill child, but also resenting the burdens of illness-related demands. Siblings may be envious of the special considerations the ill child receives. Feelings of loss contribute to the perception of stress and may deplete family resources (Lane & Hobfoll, 1992). The potential for family distress is great.

As the illness settles into a chronic phase, long term management of the arthritis becomes paramount, and the focus shifts to balancing the needs and demands of the illness with the developmental needs of all family members (Reiss, Steinglass & Howe, 1993). Quirk and Young (1990) suggest that family adaptation to JRA should become increasingly differentiated and hierarchically integrated, with an ideal state "characterized by the encouragement and support of the child's exercise therapy ... promote

independence through self-administration and regulation of anti-inflammatory medications, recognize the child's needs, and involve collaboration with the child." (p.40). However, this ideal state is not always achieved and in some instances the family's developmental trajectory is completely disrupted. In these families arthritis becomes a central organizing structure with family identity built around arthritis-related experiences and demands (Cole & Reiss, 1993; Reiss et.al., 1993; Sawa, 1992). Some families adopt the mantle of "JRA family," other families work hard to maintain a semblance of family normality and function with minimal disruptions (Strauss et. al., 1984).

Adolescent Development and JRA

Adolescence can be a critical time for children with chronic illness. Although adolescence is not characterized universally by turmoil or "storm and stress" -- in fact, adolescence is only problematic for 15 - 25% of families (Arnett, 1999; Hill, 1987) -- it is acknowledged as a time of transitions in many domains. Attitudes and styles of behavior develop in social and sexual domains, and there are significant physiological changes and growth. The search for individual identity (Erikson, 1959/1982), and the solidifying of cognitive skills, especially hypothetical reasoning (Piaget, 1963; Piaget & Inhelder, 1969) predominate as the child moves from dependence on parents to stronger affiliation with peers. Life tasks include defining a sense of identity, establishing positive and intimate peer relationships, and working towards independence and autonomy (Chassin et. al., 1995).

For adolescents with chronic illness, this path toward autonomy and identity formation may be compromised by excessive parental involvement and developmental

immaturity (Sayer et. al., 1995). Children with JRA often are physically small in stature and may experience growth retardation (Cassidy & Petty, 1995). Emotional immaturity can be fostered by the over-protectiveness of parents and the restricted involvement in peer group activities necessitated by limited mobility. The complexity of treatment regimens prolongs dependence on parents delaying parent-child separation and the establishment of autonomy. Parents of children with JRA were found to be less encouraging of independent or assertive behavior in their child than parents of healthy children, and this may become problematic as children mature (Harris, Newcomb & Gewanter, 1991). Thus, normative struggles to ward off intrusive parental involvement can be exacerbated by the presence of a chronic illness (Sayer et. al., 1995).

Successful accomplishment of developmental tasks may be compromised by the strain of dealing with a chronic illness. In early childhood, decisions related to JRA treatment and management are primarily the responsibility of parents. Typically, parents supervise and implement medication and exercise regimens, and children's compliance with treatment is associated with obedience to parental directives and the diligence of parents. By middle childhood, children become accustomed to the limitations and malfunctioning of their bodies and learn to control fears associated with the arthritis (Quirk & Young, 1990). Exploration of the social world continues, with social acceptance from parents, siblings and peers becoming paramount. As the child approaches adolescence the developing capacity for independence, self-regulation, and autonomous decision-making brings demands for greater participation in treatment decisions and management (Chassin et. al., 1995). This may conflict with the parent's desire to supervise adherence to prevent joint deformity.

Chronic illness also may impact other important areas of adolescent development. Self-esteem, body image and identity formation may be affected by the loss of competence in valued domains (e.g., physical attractiveness, peer acceptance, athletic ability). Issues of body image become particularly salient for adolescents who have musculoskeletal diseases involving joint deformity (Hinrichsen, Revenson & Shinn, 1985). There is greater dependence on social feedback and peer opinion for verification of self-worth; thus, having a body that chronically malfunctions may contribute to distorted perceptions of body image, or to dissatisfaction with physical appearance. Zeltzer and colleagues (1980) assessed issues relevant to adolescents with chronic illnesses, and found that adolescents (especially females) with arthritis reported more illness and treatment related disruptions of body image and more problems with parents than adolescents with diabetes, cancer, renal disease, or cystic fibrosis. In addition, adolescents may be reluctant to comply with medications that cause undesirable body changes. For example, common side effects of corticosteriods used to treat JRA include weight gain, bloated "moon" face, and skin changes. Thus, adolescents with JRA have to contend with many barriers to emotional, physical and social maturity. Restricted participation in peer group activities, the narrowing of career choices, and the frequent over-involvement of parents all impact on adolescents' development trajectory.

Research Goals

Within a framework that emphasizes adolescent development and family context this study has three specific aims: (1) To examine the role of perceived stress in JRA; (2) To examine the role of family-level coping in JRA, and explore the relation among stress, family coping and adjustment; and (3) To compare the findings for families with early-

and late-adolescents with JRA.

An important research focus will be to describe the personal and disease specific factors that shape the family's perception of stress and family coping response. I will document the demands inherent in dealing with the adolescents' chronic illness and explore which illness-related demands are perceived as most stressful. Similarly, I will document the most frequently endorsed modes of family coping, and investigate to what degree personal and disease characteristics influence use of family coping strategies.

A second major focus is to explore the relation among disease characteristics, stress, family coping, and psychosocial adjustment. I will test if the relationship between perceived stress and adjustment changes depending on the mode of family coping. Although high levels of perceived stress are expected to be associated with poorer adjustment, I anticipate that family coping will modify this relationship. It is expected that family coping will act as a "buffer" in the relationship between perceived stress and adjustment; that is, use of family coping will ameliorate the deleterious effects of highly stressful experiences. Moreover, I will explore which modes of family coping are associated with better psychosocial adjustment.

The final research goal will be to explore if differences in developmental status affect the relationship among stress, coping and adjustment. Empirical findings of a relationship between coping repertoire and developmental status (Aldwin, 1994; Compass et.al., 1992) suggests that families will be influenced by the early adolescent's reliance on unsophisticated strategies such as blaming others, minimizing problems, and inappropriate emotional outbursts. In contrast, older adolescents are expected to have learned to regulate their emotions and have mastered more complex and differentiated

coping strategies. Furthermore, based on the differences between the developmental tasks of early and late adolescence, differences in perception of stress and adjustment to JRA are expected.

Although it is beyond the realm of this study to examine how stress, coping, and adjustment change over time as a function of experience in dealing with the arthritis-related demands, I anticipate that there will be sample variability in the length of time since diagnosis. It is expected that more recently diagnosed children will experience more adjustment problems, and that these families will perceive their child's illness to be more stressful.

A Proposed Model of Disease, Stress and Family Coping

The conceptual model is depicted in Figure 1. Family coping and perceived stress are anticipated to modify the relationship between disease or demographic indicators and psychosocial adjustment. Briefly, characteristics of the illness (e.g., disease severity and functional limitations), and of the family (e.g., child's developmental status) are expected to influence the level of stress perceived by JRA families. High stress is expected to be associated with poorer adjustment; however, this process is expected to be influenced by family coping. This proposed moderational model would identify the explanatory processes underlying the main effects model in most current research whereby psychosocial adaptation to JRA is assumed to be determined by factors such as arthritis severity. Although research in this tradition has predominated, the evidence for direct pathways between disease severity and adjustment to chronic illness has been equivocal.

First, I will give a brief overview outlining each of the pathways depicted in Figure 1. Then, I will examine empirical and theoretical evidence for these pathways.

Relation between Demographic Indicators and Stress (path a). Personal attributes, such as the child's developmental stage or the family's socioeconomic status are expected to be related to illness-related stress. For example, adolescents' developmental status affects the type of demands that confront families dealing with JRA. And, there are financial burdens associated with medical and treatment costs which may contribute to family stress.

Relation between Disease Characteristics and Stress (path b). Disease severity and the extent of the adolescents' limitations in daily functioning are anticipated to affect the level of stress perceived by families. For example, adolescents with severe arthritis in many joints require more aggressive medical intervention, assistance with daily activities, and more active family involvement. Thus, illness severity is expected to be associated with increased stress. Similarly, the length of time since diagnosis of the illness is expected to influence the degree of stress experienced by families. The period immediately following diagnosis is often marked by turmoil as family members learn to accommodate to the rigors of the multi-component treatment regimen. Over time these routines become entrenched, as families develop skills to manage the unpredictable course of arthritis, stress is expected to lessen.

Relation between Demographics Indicators and Adjustment (path c). It is anticipated that there may be differences between younger and older adolescents in terms of adjustment to JRA. Additionally, families with low socioeconomic status are expected to experience more problematic adjustment, as the financial burdens of treatment may exceed their limited resources.

Relation between Disease and Adjustment (path d). Similar to the relationship between

disease status and stress, it is anticipated that severity of the adolescents' JRA will be related to adjustment problems. In particular, adolescents with marked functional limitations are expected to report more social and behavioral difficulties. Less severe JRA is expected to be associated with reports of increased quality of life. Parental depression is also expected to be related to disease severity.

Relation between Stress and Adjustment (path e). Family perceptions of high levels of stress is anticipated to be related to problematic adjustment, such as increased parental depression, poor adherence, and lower quality of life. In contrast, lower stress is expected to be associated with better adjustment.

Influence of Family Coping on the Relationship between Stress and Adjustment (path f). The final pathway in the model is the relation among stress, family coping and adjustment to JRA. It is expected that family-level coping strategies will modify the relationship between stress and psychosocial adjustment. Although it is anticipated that perceptions of high stress will be associated with problems of adjustment, use of specific family coping strategies are expected to ameliorate the potentially negative influences of high stress, that is, family coping will act as a stress-buffer.

Review of the Evidence for these Paths

The following sections will examine the empirical and theoretical evidence for these paths in the model (Figure 1) outlined above. Each section will review the current literature concerning relationships among major variables in the model.

Demographics and person attributes

Person attributes such as the child's age, gender, and the family's socioeconomic status are expected to be related to the family's perception of stress (Figure 1, path a).

Child's Age

Given the vast literature detailing developmental changes throughout childhood, it is surprising that research on the psychosocial adjustment of children with JRA lacks a developmental perspective. Nor does it address the difficulty measuring and comparing health and functional status across a diverse age range. Illness construals, and the normative life tasks encountered, depend on the developmental status of the child (Berry et.al., 1993; Biback & Walsh, 1980; Quirk & Young, 1990; Roberts & Wallander, 1992). With increasing age there is a trend towards more sophisticated understanding of disease pathology and increasing independence in all domains including management of JRA (Singsen, 1991). However, few studies have considered age as an independent variable or considered developmental issues, and it not unusual to see children with extremely diverse ages analyzed as a single group. Two notable exceptions were an Australian study (Ungerer et. al., 1988) that compared elementary school age, high school age, and young adults, and an American study (Daltroy et.al., 1992) comparing children aged 4-5, 6-11, and 12-16. In the Australian study younger children reported lower self-concept, greater loneliness, spending more leisure time alone, and being teased more often about their JRA. In the American study, adolescents were less socially competent than the younger children. One possible explanation is that peer group activities become increasingly important for older children and JRA affects participation in sports and social activities. These two studies highlight the influence of age on social functioning and confirm the need for a developmental perspective.

Child's Gender

Although proportionally more females than males are diagnosed with JRA, few

studies address gender issues. A single study directly compares boys and girls: Daltroy and colleagues (1992) found that adolescent males reported more aggressive and acting-out behaviors than females. It is likely that some of the more serious complications of JRA -- in particular, growth retardation and delayed sexual maturity -- may affect the perception of body image and self-concept differently for male and female adolescents. In addition, parental over-protectiveness and expectations of competency may differ.

Structural Variables

Similarly, the JRA literature does not explore the influence of ethnicity and socioeconomic status, two variables that are powerfully linked to health outcomes (e.g., Adler et.al., 1994; Singsen, 1991). Financial security, education, and the ability to access and understand complex medical information about JRA are considered to be important family resources.

The Nature of JRA

Disease severity is expected to affect the level of stress perceived by families (Figure 1, path b), and the psychosocial adjustment of adolescents (Figure 1, path e). The following section will describe the nature of JRA, and detail the medical aspects of JRA that comprise disease severity.

Prevalence and etiology. JRA is an autoimmune disease of unknown etiology, and it is one of the most common rheumatic diseases of childhood. Although the actual frequency of JRA is unknown, it is estimated that approximately 285,000 children in the USA have some form of arthritis (Arthritis Foundation, 1999). JRA is found in all races (although proportionally fewer Black than White children have JRA) and all geographic regions of the country. Data from a 1983 Mayo Clinic survey estimated an incidence of

13.9 new cases per 100,000 per year; and prevalence rate of 113.4 per 100,000 children (Cassidy & Petty, 1995).

JRA has been arbitrarily defined as arthritis that begins before the age of sixteen. It is a chronic but not life-threatening disease. Although it is difficult to predict arthritis course, approximately 10% - 30% of children develop moderate to severe functional disability. Arthritis symptoms can occur at any age; however, the age of symptom onset is often quite young, peaking between the ages of one and three years. This early onset is most pronounced in girls. Girls are also more likely to have JRA than boys, with a ratio close to 3 : 1 for the two most common forms of JRA (Cassidy & Petty, 1995).

Research efforts to establish JRA etiology have focused on both physiological and psychological predisposing factors. There is some evidence of JRA occurring in families, but investigations of genetic predisposition have been inconclusive (Cassidy & Petty, 1995). Recent medical research efforts have been on a cellular level and are based on evidence of altered immunity in children with JRA, the association between specific immunodeficiencies and rheumatic disease, and the close relationship between immune reactivity and joint inflammation (Cassidy & Petty, 1995).

Symptoms and diagnosis. JRA is a disease of the joints but it may also affect other body organs (e.g., eyes). Arthritis is characterized by four major changes in the joints: inflammation, contracture, damage, and altered growth. The most common symptom of JRA is joint inflammation. The joint lining (synovium) becomes thickened and produces too much synovial fluid, resulting in swelling, pain, stiffness, and sensations of warmth. To minimize discomfort, children may restrict use of an inflamed joint or hold the joint in a flexed position. Over a period of time this can result in a joint

contracture. Constant effusion and swelling of the joint damages the synovial tissues and joint surfaces, resulting in demineralization of the bones and erosions (i.e., pitting) of the bone surface. Chronic inflammation may either accelerate or retard growth centers in the bone.

At onset, JRA symptoms may be confused with those of other diseases, and children may be referred to orthopedists or other specialists before referral to a rheumatologist is made (Arthritis Foundation, 1999). Diagnosis of JRA is made by the physician after taking a complete health history, physical examination (to assess joint inflammation, range of motion, and the presence of rash, rheumatoid nodules, or eye problems), laboratory tests (e.g., erythrocycte sediment rate, rheumatoid factor test [RH], antinuclear antibody test [ANA], HLA-B27 typing), radiographs or MRI to identify abnormalities and bone changes (i.e., erosions, calcification, or widening of the joint space), and joint aspiration (to test for infection in the synovial fluid). Only a small percentage of children are positively diagnosed with JRA through blood work; however, these tests are important because RF seropositivity is a hallmark of poor prognosis and these children require more aggressive medical intervention (Cassidy, Levinson & Brewer, 1989). A set of five diagnostic criteria proposed by the American College of Rheumatology (ACR) includes: (1) Age of symptom onset less than 16 years; (2) Arthritis (swelling or effusion, limitation, and increased heat) in one or more joints; (3) Duration of symptoms for at least six weeks; (4) Onset type defined by the type of the disease in the first six months (polyarticular, five or more inflamed joints; pauciarticular, less than five inflamed joints; systemic, arthritis with characteristic fever); and (5) Exclusion of other forms of juvenile arthritis (Cassidy & Petty, 1995).

Clinical manifestations and complications. One of the early symptoms of JRA is fatigue, particularly for children with pauciarticular or systemic onset. Fatigue -- lack of energy, increased sleep requirements and irritability -- also may be evident during arthritis flares. Joint pain at night may disrupt sleep and contribute to the child's tiredness. Anorexia, weight loss and failure to thrive are common in children with systemic onset. In older children, active arthritis may delay sexual maturation.

The arthritic joint usually exhibits cardinal signs of inflammation (swelling, pain, warmth, redness, and restricted range of motion or loss of function). Children may describe the joint pain as either a stretching or aching sensation, and the pain may be reported as ranging from mild to moderate. Reports of pain vary with age (Ilowite, Walco & Pochaczevsky, 1992). Very young children may not complain of pain, but refuse to use the affected joint in play. Psychological factors such as child anxiety, family harmony, and maternal anxiety have also been found to influence pain reports (Ross et. al., 1993). Any joint may be affected by arthritis, but the large joints (knee, elbow, wrist) are most commonly involved (Cassidy & Petty, 1995).

Variations of the clinical characteristics of JRA at onset are the basis for the classification of JRA into three major subtypes: polyarticular; pauciarticular; and systemic onset. Half the children have four or fewer inflamed joints (pauciarticular onset) during the first six months of the disease. Pauciarticular arthritis most commonly involves joints of the knees, ankles or elbows. In many children only a single joint is affected. Approximately 20% of children with pauciarticular onset -- especially young girls with ANA seropositivity -- may develop iritis (asymptomatic eye inflammation).

Ten percent of children have severe systemic involvement prior to the

development of overt arthritis symptoms. The diagnostic hallmark of systemic onset JRA is a high spiking fever that usually occurs in the early evening, often accompanied by a migratory pink rash. Fifty percent of children with systemic JRA recover completely, while others show progressive involvement of additional joints. Pericarditis (heart problems) occurs in three to nine percent of children with systemic onset (Cassidy & Petty, 1995).

Polyarticular onset (five or more affected joints) is often insidious, but may be acute. There tends to be symmetric involvement of the large joints of the knees, wrists, elbows, and ankles. The inflammation of the cervical spine, temporomandibular joint (the jaw), and the small joints of the hands and feet often occurs. Adolescent girls who have late onset of arthritis and who test positive for rheumatoid factor (RF+) have a poorer prognosis, with symptoms more like adult rheumatoid arthritis. This group may develop rheumatoid nodules (firm or hard lumps under the skin) and experience a more chronic disease course (Cassidy et. al., 1989).

Children with polyarticular or systemic onset are more likely to experience difficulties, as more joints are affected and symptoms are more severe than pauciarticular onset. These children have increased school absences, are more likely to have problem with daily activities, and may require special education services (Kewman, Warschausky & Engel, 1995). In addition, noticeable symptoms such as nodules, joint swelling, deformity, or skin discoloration may exacerbate body image problems.

The most frequent complications of JRA are abnormalities of growth and development. Linear growth may be retarded during active systemic disease, and during periods of remission accelerated growth may occur. The maturity of the skeletal frame at

the time of inflammatory insult determines whether the long-term result will be lengthening or shortening of the limb. There may be asymmetry of development if there are localized growth disturbances. Arthritis in one knee is often associated with accelerated bone growth and this can result in discrepancies in leg length. Additionally, atrophy and weakness of the muscles around inflamed joints can result in shortening of the muscles and tendons (joint contracture) exacerbating growth abnormalities. Severe contracture may require splints, casts or surgery (i.e., soft tissue release). In older fully grown adolescents, joint replacement can reduce pain and improve the function of badly damaged joints.

Treatment of JRA. Although JRA cannot be cured, spontaneous remissions occurs in many children. Control of active arthritis symptoms incorporates many treatment components including pharmacological, physical, and psychosocial management. The goal of comprehensive management of JRA is "to control inflammation, prevent joint deformities, maximize function and promote psychosocial adjustment" (Kewman, Warschausky & Engel, 1995, p. 385). Long-term treatment goals include: minimizing the effects of the disease and treatment; promoting normal growth and development; rehabilitation; and education. Typically, children with active JRA require monthly appointments with the pediatric rheumatologist, and frequent physical or occupational therapy (2 - 3 times/week) which may or may not be provided through the educational system under PL 94-14.

Pharmacological treatment often includes nonsteroidal anti-inflammatory drugs (NSAIDS), or more potent corticosteroids and disease-modifying anti-rheumatic drugs such as methotrexate. A new class of medication known as a biologic agent (e.g., Enbrel)

is now used for those who fail trials with more traditional medications.

As an adjunct to medication, therapeutic exercises are prescribed to help maintain or restore joint function (range-of-motion exercises), and prevent contractures (strengthening exercises). Children are encouraged to participate in as many normal activities as possible but to avoid activities such as contact sports that may stress inflamed joints. The complexity of the treatment regimen contributes to difficulties adhering to treatment recommendations. It is important for children to adhere to the treatment regimen, as failure to do this may lead to long-term consequences such as joint deformity. Not all families are equally competent in managing treatment-related demands.

Perceived Stress and Family Coping in JRA

Family perception of stress is anticipated to be associated with psychosocial adjustment (Figure 1, path e), and it is expected that family coping will be a moderator of this relationship (Figure 1, path f). The following section will describe sources of family stress in adolescents with JRA and the types of coping strategies used to deal with this stress. Empirical evidence describing the relationship among stress, coping and adjustment will be examined.

Sources of stress

Families who have a child with JRA encounter a unique configuration of illness-related stressors and tasks. The early peak onset of JRA, the unpredictable disease course, the lack of one-to-one relationship between treatment adherence and symptom relief, and the chronic nature of JRA shape the coping tasks encountered by families. There are many competing or conflicting demands on the family's resources, and JRA

may place additional strains on families with few financial or social resources. The need for frequent clinic visits may tax parental resources in terms of taking time off work, traveling long distances for specialist services, or having adequate medical insurance for a chronic condition. These financial and time burdens increase the strain on family resources and may magnify perceptions of stress.

The complexity of a treatment regimen that involves exercise, pharmacological, and psychosocial components, and the need to incorporate medication and exercise schedules into daily routine can pose extra stress for families. Pre-existing family problems may be exacerbated by treatment demands (Chaney & Peterson, 1989; LaGreca et.al., 1992). Family routines, meals and activities often need to be reorganized as medication is often taken with food to avoid gastric problems. In most pediatric chronic illness there are compliance difficulties for regimens that are time consuming, complex, expensive, or require life style changes (LaGreca & Schuman, 1995; Rapoff, 1989; Thompson et. al., 1995). The JRA treatment regimen involves all these factors.

Pain is a major source of stress for most children with arthritis. Children must learn to deal with the persistent joint pain associated with inflammation. Pain management is achieved through a combination of drugs, exercise, and cognitive-behavioral self-regulatory techniques (e.g., progressive muscle relaxation). All require a major investment of time and effort, and parents often are conflicted about enforcing physical therapy as stretching inflamed joints is often painful (Brewer & Angel, 1995). Although pain reduction using self control techniques has shown promise of success, it is difficult to maintain child interest and motivation to use the strategies frequently enough to ensure pain relief (Walco, Varni & Ilowite, 1992; Wallander & Varni , 1995).

A further disruption to family functioning is the need to modify family routines and activities to adapt to the fluctuating status of the child's arthritis symptoms. Pain experience is unpredictable and can vary from morning to afternoon. This necessitates a degree of flexibility in planning family activities or outings. Stress may be experienced by the entire family if the child is unable to participate in sports or family activities they had previously enjoyed. When the child is experiencing arthritis flares, parents may need to assist more with activities of daily life (e.g., dressing), and siblings may have to assume the ill child's share of household chores. This has potential to cause problems of sibling resentment, and overprotective behavior by parents even during periods where the child is functioning well.

There is some evidence that children with JRA do not achieve as well academically as healthy peers. Fatigue, distractibility, inattention due to pain interference, and limitations of mobility, combined with higher rates of absenteeism than national norms contribute to lower academic performance (Lovell et al., 1990). Children may also have poor gross- and fine-motor coordination that interferes with writing and gym participation (Bartholomew et. al., 1994; Stoff, Bacon & White, 1989).

Arthritis is typically an 'invisible' disease as most children do not have obvious joint deformity, thus, the child's discomfort is not objectively evident. This may lead to teachers having unrealistic expectations of the student. Children with arthritis in finger or wrist joints may have difficulty writing, and carrying books or lunch trays. They may need extra time to complete tests and require an extra set of books to keep at home. In addition, classroom routines may have to be adapted to accommodate the arthritis as children may need to move about during class time to prevent joint stiffness, or need to

rest inflamed joints during gym (Arthritis Foundation, 1999). In spite of these special needs, a large multicenter study found that only 47% of children with JRA received necessary services (mandated under PL 94-142) to facilitate academic achievement and minimize the impact of physical limitations (Lovell et. al., 1990). Thus, parenting a child with JRA may involve acting as an advocate to ensure individualized education programs that include appropriate physical and occupational therapy with the goal of keeping the child within the mainstream of educational and social activities.

Family Coping

Coping is a psychological mechanism that enables individuals to manage stress and it involves a complex interaction of cognitive, behavioral and emotional components. Although there are several theoretical models of coping, most incorporate the distinction between problem-focused coping (i.e., taking concrete action to reduce or solve the problem), and emotion-focused coping (i.e., regulating emotional responses to relieve the distress of the situation), originally described by Richard Lazarus (e.g., Lazarus & Launier, 1978). In addition, most theories include cognitive, behavioral and affective responses. Cognitive coping involves efforts to reinterpret or reappraise situations so they are perceived as less threatening (see also Carver, Scheier & Weintraub, 1989; Pearlin & Schooler, 1978). Another type of coping that doesn't appear in most models but is specific to illness, is the use of normalizing strategies. Normalizing is defined as refusing to adopt a sick role and making efforts to hide or cover-up symptoms and maintain as normal a lifestyle as possible (Strauss et. al., 1984).

In families where a child has JRA, both individual and family coping strategies may used, as JRA impacts on the both the child and the family. Much of the research on

coping focuses on the coping strategies of individuals; however, individual attempts to manage stressful situations have repercussions for the family unit. Family members interact in complex ways to jointly manage stressors, and the functioning and coping skills of each individual affects the responses of other family members in addressing the common problem of "dealing with the arthritis." For example, poor functioning and ineffective coping by parents, as evidenced by maternal depression or anxiety, is associated with the poorer adjustment of chronically ill children (Daniels et. al., 1987; Timko et.al., 1992; Vandvik & Eckblad, 1991).

Coping is connected to family dynamics in a variety of ways (Compass et. al., 1992). First, family members provide both tangible and emotional support for the child with JRA. Second, family members may either facilitate the coping process, for example, provide appropriate coping models, or impede it. For example, Thoits (1986) conceptualized social support from others as coping assistance, and suggested that if there is empathic understanding and a 'good fit' between the needs of the supportee and the support offered, then adaptive functioning can be enhanced. This suggests the importance of families to be empathic to the needs of the ill child and offer appropriate support that will aid the child in his or her efforts to deal with arthritis-related demands. Easier said than done! Third, families have rules, rituals and belief systems that influence the coping strategies utilized by individual members. For example, if families believe in the value of "keeping a stiff upper lip" in the face of adversity, then the coping responses of the child may be constrained in expressing concerns, fears and anxieties. Constraints within the family environment may lead to feelings of isolation, loneliness and poorer health outcomes (Lepore, 1997).

Extrapolating from a transactional view of stress and coping, whereby there are mutually reciprocal influences between the person and the environment that affect both perception of stress and coping responses (e.g., Aldwin 1994, Lazarus, 1981), family coping responses reflect a dynamic transaction between family and situational factors. For families who have a child with arthritis, family factors (e.g., beliefs regarding JRA controllability and self-efficacy) and situational factors (e.g., arthritis severity, and the availability of medical and supportive resources) interact to determine the family coping responses. This interaction between family and situation is multidirectional and includes feedback loops, for example, if the family's coping resources are inadequate to meet the demands of managing a complex treatment regimen, this influences the family's beliefs in their ability to deal with future stressors.

Stuart Hauser emphasized this characterization of coping as a family process in his seminal work on family-level coping processes among families of diabetic children. Hauser defined family coping as " the thoughts and behaviors that the family expresses in attempting to handle or control the effects of immediate and long-term stressful situations. Such responses include ways of observing, defining, acting and experiencing" (1993, p. 309). Hauser and colleagues developed a coding scheme to document these cognitive, behavioral and affective aspects of family coping, and found that adolescent's treatment compliance was associated with family cohesion and coordinated family efforts to solve problems. Highly compliant families expressed feelings of optimism and mastery, and were active in seeking both support and information about their child's diabetes from the health care team. In contrast, noncompliance was associated with family use of denial and expression of feelings of helplessness (Hauser et.al., 1986,

1993).

Family coping responses may change as a function of the cognitive and emotional maturity of the ill child. Compass, Worsham and Ey (1992) found evidence of a positive relationship between children's reports of emotion-focused coping and age. Although rudimentary emotion-focused coping (i.e., self-comforting behaviors such as thumb-sucking) was evident from infancy, more sophisticated verbalizing of feelings did not emerge until later in development. During early childhood (i.e., pre-operational and concrete operational stages) children have less access to internal states and may fail to realize that emotions can be self-regulated, however they are able to model the overt (i.e., problem-focused) coping behavior of adults. By the stage of formal operations, the repertoire of coping strategies increases and becomes more complex and differentiated. Early adolescents relied on unsophisticated strategies such as blaming others, minimizing the significance of problems, and inappropriate ventilation of emotion. By late adolescence they had learned to regulate emotions to deal with stressful situations, and reported using a combination of problem-focused and emotion-focused coping strategies (Compas et.al., 1992). Aldwin (1994) notes that all children use some form of avoidant strategies, with younger children relying on escapism and older children utilizing more sophisticated cognitive strategies such as distraction.

Family coping responses also change over time as a function of changing demands of the illness (Cole & Reiss, 1993). Prior to diagnosis families may use denial to manage the overwhelming emotional responses to their child's pain and discomfort. Following diagnosis, denial would no longer be an effective strategy, as the situational demands have changed and the family now needs to seek information about treatment

and become involved in appropriate long-term medical care. Similarly, coping strategies that are effective during periods of arthritis flares may differ from the strategies used when symptoms are in remission.

All JRA families need to develop ways of helping the child manage pain. Psychological factors can modify the pain experience. Maternal anxiety is associated with greater pain, and for some families the expression of pain (e.g., complaining, crying) may be unwittingly reinforced as it is attended to by concerned parents (Ross et. al., 1993). Studies of coping with acute pain, such as stressful medical procedures, have found that active coping (e.g., seeking information) was associated with higher pain tolerance (e.g., Peterson, 1989); however, little is know about the coping strategies used by children to deal with chronic arthritis pain. Although interventions that included family social support were found to reduce pain perceptions for adults with arthritis (Radojevic et. al., 1992), the role of family support in modifying children's pain has not been studied.

Appraisal processes

Lazarus and his colleagues (Folkman et.al., 1986) have suggested that two interconnected processes -- the cognitive appraisal of the stressful situation and the use of coping strategies -- mediate stressful person-environment relations and psychological outcomes. Differences in cognitive appraisal of an illness influenced the type of coping strategies used. If the stressor was perceived to be a challenge, adolescents used more logical analysis, positive reappraisal, engaged in active problem solving and sought alternative rewards (Ebata & Moos, 1991; Folkman, 1984).

Appraisal of the stressful situation is an important component of many coping

theories. Parents, children, and health professionals may have divergent perceptions of JRA-related stressors and use different criteria to gauge successful outcome. Physicians may be primarily concerned with symptom reduction and compliance, while families focus on incorporating JRA-related demands into everyday routines (Rudolph, Dennig & Weisz, 1995). Konkol and colleagues (1989) explored the stress appraisals of 50 JRA families. Parents and children had similar perceptions about the general effect of JRA -- that is, they all associated arthritis with symptoms of pain, swelling and stiffness -- but differed in their perception of the impact of JRA on family and social functioning. Parents reported the psychological stress of dealing with the uncertainty of arthritis was the greatest burden. In contrast, children and their siblings were more likely to regard emotional stress, physical limitations and dealing with pain as more problematic. In terms of school difficulties, parents focused on the physical limitations (e.g. difficulty writing) and the need for communication with school personnel, whereas children expressed anxiety about peer acceptance, especially the reaction of peers to obvious signs of disability. There were also discrepancies between parents and children's concerns regarding disease progression. Parent's fears focused on the possibility of their child becoming disabled and unable to maintain normal social and vocational roles. Children worried about becoming a family burden and feared their arthritis might worsen. Many children expressed fears of dying.

Appraisal of the controllability of the stressor has been found to be associated with the type of coping strategy used. When the stressor is perceived as controllable, problem-focused coping is more likely to be used; if the stressor is appraised as resistant to active intervention then emotion-focused coping is preferred (Aldwin, 1994; Carver,

Scheier & Weintraub, 1989; Compas et.al., 1992; Folkman et al., 1986). Strong beliefs in the controllability of JRA may be associated with active family interventions such as researching treatment options or monitoring adherence. In contrast, where families believe that they have no control over fluctuations of arthritis symptoms, coping strategies may involve encouraging family members to express feelings, and empathize with the child. Alternatively families may "give-up" if they believe that nothing they do will improve the situation, and they may either passively accept the JRA or feel overwhelmed or angry at the demands of the child's illness (Lane & Hobfoll, 1992).

Family coping and adjustment

Which coping strategies are associated with better psychosocial adjustment? On the individual level, for children with different types of chronic illness (e.g., diabetes, sickle cell), adjustment has been associated with active coping, distraction and seeking support (Lewis & Kliewer, 1996). On the family-level, coordinated family efforts to deal with problems, seeking information about the illness, optimism, and openly expressing feelings have been associated with better adjustment for families with a diabetic child (Hauser et.al., 1993). Similar coping strategies have been found to be associated with better adjustment for children with JRA. Timko, Stovel and Moos (1992) found that cognitive strategies to manage the appraisal of JRA-related stress, and direct behavioral attempts to deal with the problem were associated with better adjustment. Konkol and colleagues (1989) found that maintaining a positive attitude, and having social support from friends, family and health professionals were cited by children as helping them to understand and deal with problems related to arthritis. Ebata and Moos (1991) found that adolescents with JRA who used more approach coping -- logical analysis and reappraisal

of the problematic situation, and seeking support to solve problems -- had higher levels of psychological well-being and lower distress. In contrast, adolescents who were either depressed or conduct disordered were more likely to use strategies of cognitive avoidance, seek alternative fulfillment (e.g., drugs or alcohol), and were more apt to have emotional outbursts.

In an extension of the Folkman and Lazarus' theory of coping and adaptation, Wallander and Varni (1988, 1992, 1995) developed a "disability-stress-coping model of adjustment" in pediatric chronic illness. In this model, risk and resistance factors are thought to interact to produce child adjustment. Risk factors such as, disease severity, functional independence and psychosocial stress are thought to be primarily responsible for adjustment difficulties. However,

resistance factors are believed to influence this risk-adjustment relationship. Resistance factors such as, intrapersonal factors (e.g., temperament, competence), stress processing (cognitive appraisals and coping strategies), and social-ecological factors (e.g., family environment, social support) are believed to have both a direct effect and a moderational influence on adjustment. All components of this complex model have not yet been fully tested, however, preliminary data suggests that psychosocial stress is a significant risk factor for chronically ill children and social support is an important resistance factor (Wallander & Varni, 1992, 1995). My study contributes to this research tradition by examining the role of family coping and perceptions of stress in pediatric adjustment.

Psychosocial Adjustment in JRA

Most JRA research has focused on either relationships between disease severity and adjustment (Daltroy et. al., 1992; Frank et. al. 1998; Ungerer et. al. 1988) or coping

and adjustment (Ebata & Moos, 1991; Harris et. al., 1991) overlooking the role of perceived stress. Recent meta-analyses and reviews of adjustment among chronically ill children have found that despite generally good overall levels of adjustment, some children with chronic illness are at risk of developing adjustment difficulties (Cadman et. al., 1991; Lavinge & Faier-Routman, 1992; Thompson & Gustafson, 1996). A review sponsored by a committee of the American Academy of Pediatrics states, "Large community based studies and national surveys ... suggest that the majority of children and adolescents with chronic health conditions do not have identifiable mental health, behavioral or educational difficulties... Nevertheless, these same studies show that children and adolescents with chronic conditions do have about twice the prevalence of psychological symptoms as compared to children without a chronic condition." (1993, p. 876). Additionally meta-analyses of pediatric adjustment studies do not suggest a direct relationship between having an illness and lowered psychosocial adjustment, but point to intervening factors. Accordingly, many studies have explored variables that could make a difference in children's adjustment to chronic illness, including maternal depression (Lustig et. al., 1996; Vandvik & Eckblad, 1991), use of specific coping modes (Ebata & Moos, 1991; Harris, Newcomb & Gewanter1991; Hauser et. al., 1993), and family functioning (Blechman & Delmater, 1993; Chaney & Peterson, 1989; Timko, Stovel & Moos, 1992).

Good adjustment among pediatric populations has been defined as normative, healthy, and age-appropriate behavior that follows a trajectory towards positive adult functioning (Wallander & Thompson, 1995). Studies of pediatric chronic illness propose that markers of good adjustment include treatment adherence, acknowledging the

challenges and responsibilities of illness, pursuit of age-appropriate peer and family relationships, academic achievement, and success with developmental tasks. In contrast, maladaptive adjustment is characterized by poor peer and family relationships, low self-esteem, anxiety, passive dependence or manipulativeness, regressive behavior, and highly noncompliant self-care (Billings et. al., 1987; Daltroy et. al., 1992; Daniels et. al., 1987; Ennett et. al., 1991; Harris et al., 1991; Jaworski. 1993; Timko et. al., 1992; Wallander & Thompson, 1995; Wertlieb, Jacobson & Hauser, 1990). Psychosocial adjustment is not unidimensional; in fact there are many components of adjustment. Therefore, multiple measures are needed to capture family adjustment. Wallander and Varni (1992) used measures of mental health, physical health, and social functioning to assess the psychosocial adjustment of chronically ill children. Stuart Hauser and colleagues used measures of adherence, family cohesiveness, and ego development as indicators of family adaptation (1993). Specific dimensions of family adjustment will be discussed in the following sections.

Treatment adherence. There is general agreement that adherence is an important issue in the treatment of pediatric populations (Bearison, 1994, 1996). Estimates of adherence for children with JRA range from 55% (Litt & Cuskey, 1981; Rapoff, 1989) to a high of 95% (Hayford & Ross, 1988). Typically, lower rates of adherence are reported for range-of-motion exercises compared to adherence to medication (43% vs. 60%; Rapoff, 1989).

The Health Belief Model (Becker et. al., 1972, 1978) suggests that willingness to adhere to treatment can be predicted by weighing perceived benefits and costs of treatment. The benefits of good adherence to JRA treatment, such as, reduced morbidity,

pain, and joint deformity, are often obscured by the absence of a direct relationship between taking medication and symptom relief. Delayed response to pharmacological treatment and unpredictable cycles of arthritis flares may strengthen the adolescents' belief that medication is ineffective (Rapoff, 1989). This diminishes reinforcement for adherence behaviors. Additionally, the costs of adherence can be substantial -- unpleasant side effects of medication, and the extensive time demands of range-of-motion exercises. Thus, for adolescents with JRA costs may outweigh any perceived benefits of compliance.

For children, beliefs regarding treatment benefits are related to age and cognitive development. Beales and colleagues (1983) found that younger children (7 - 11 years) viewed both JRA and treatment in concrete terms, with little understanding that treatments that are immediately unpleasant could have long term beneficial effects. In contrast, older children (12 - 17 years) viewed JRA as an internal pathological state and understood the relationship between treatment and later benefits. There are also age-related changes in assuming responsibility for remembering the medication or exercise. Parents almost always supervise the treatment regimen of younger children; however, responsibility for adherence is transferred as children become older. Hayford and Ross (1988) found that parents expect children to begin assuming responsibility for medication by 11 to 13 years of age, however, at this age there is a concurrent decline in adherence.

For all chronic illnesses, adherence declines over time and when the patient is asymptomatic (LaGreca & Schuman, 1995; Melamed, 1984; Rapoff, 1989). Therefore it is expected that adolescents with long-term arthritis and those in a quiescent phase (i.e., no active symptoms but still requiring medication) may be less compliant. In general,

poor treatment adherence is believed to be associated with frequent arthritis flares, the family's poor understanding of arthritis, complexity of treatment regimen, adverse side effects of drugs, and conflict between health team members and the family. Cassidy and Petty (1995) suggest that compliance could be increased by positive reinforcement of the desired adherence behavior, efforts by the physician to educate families about disease and treatment approaches, and agreement among patient, family and physician regarding treatment goals (also see suggestions by Christopher, 1994; LaGreca & Schuman, 1995; Thompson et. al., 1995).

Family dynamics play a crucial role in adherence, as noncompliance may help to maintain customary roles in the family. Compliance is harder to achieve when adhering to the regimen requires extensive changes in family roles, tasks or interaction patterns (Gervasio; 1986). Chaney and Peterson (1989), in a study of 25 families with JRA, found that family dysfunction was related to lower compliance. Specifically, maladaptive families who were less cohesive and had experienced more life stressors had children who were less compliant with their treatment regimen. In contrast, father's satisfaction and mother's reported use of a greater number of coping strategies were closely associated with more adaptive functioning and better medical compliance. Studies of other pediatric chronic illnesses have also found that adherence is influenced by family interaction, with effective communication and family problem solving associated with better adherence; and confrontative, emotionally charged and negative interactions associated with poorer adherence (Blechman & Delmater, 1993; Hauser et.al., 1993).

Adolescents' social and behavioral adjustment. In spite of the generally good adjustment of chronically ill pediatric populations, meta-analyses and reviews of the

literature concur that children with a chronic illness are more likely to have adjustment problems than healthy children (Lavinge & Faier-Routman, 1992; Thompson & Gustasfson, 1996). A large and methodologically sound study of 270 chronically ill children found that these children were less socially competent and reported more behavioral problems (e.g., withdrawn or acting-out behaviors) than a healthy community sample (Wallander et.al., 1988). Similarly, most studies of psychosocial adaptation to JRA, report increases in psychological problems for children with arthritis (Daltroy et.al., 1992; Daniels et.al.,1987; Harris et.al., 1991; Jaworski, 1993; McAnarney et.al., 1974; Timko et.al., 1992; Ungerer et.al., 1988; Vandik, 1990). Only one study reported no differences in behavioral or emotional problems between children with JRA and healthy peers (Billings et.al., 1987).

One factor that has been examined in attempts to account for discrepant findings in adjustment is the effect of disease severity on adjustment. In several studies, disease severity was associated with increases in psychosocial and behavioral problems. Children with systemic or polyarticular onset JRA were found to have more problems related to physical limitations and school functioning (Daltroy et. al., 1992; Kewman, Warschausky & Engel, 1995). Billings and colleagues (1987) found that children with more severe JRA reported more physical problems (e.g., pain and stiffness) and more psychological problems (e.g., anxiety and depression), but did not report more behavioral problems. Paradoxically, one early study found that children with the least severe arthritis had the most psychological problems and made more negative self statements such as feeling "different", "inferior", or "unworthy" (McAnarney et. al., 1974). In a comprehensive review of the literature, Jaworski (1993) concludes that "the preliminary

evidence suggests that severity of illness and functional loss, children's perception of JRA, child self-concept, parent's level of psychosocial disturbance, and family stressors, problems and social support may be related to the psychosocial adaptation and functioning of JRA patients." (p. 194). Clearly there is no simple or direct link between illness severity and children's adjustment to arthritis.

There is as yet no satisfactory account of the processes that lead to either successful or unsuccessful adjustment to the stressors of JRA. Children with chronic illness have been identified as a group at risk for poor psychosocial adjustment (Pless, 1983; Varni et. al., 1988; Wallander & Thomas, 1995), yet considerable variability exists within groups of children with respect to their psychological functioning. Not all children experience adverse outcomes, and successfully dealing with the stressors of illness may even promote personal growth. These children are thought to be resilient or stress-resistant and they thrive on the challenges inherent in the management of chronic arthritis. Resilient individuals (and families) are thought to have developed coping skills that enable them to respond to challenges and counteract the negative impact of stressors (Cowan, Cowan & Schultz, 1996; Hetherington & Blechman, 1996; O'Leary & Ickovics, 1995).

This concept of thriving in the face of adversity could explain the findings of one study (Harris, Newcomb & Gewanter, 1991) where children with JRA were not at a psychosocial disadvantage and in fact had a greater repertoire of coping strategies than a comparison group of healthy children. Children who predominantly used active, problem-focused coping strategies reported less stress. In another study (Ebata & Moos, 1991) adolescents who relied on proportionally more approach coping (i.e, cognitive and

behavioral attempts to define, understand and resolve the stress) and less avoidance coping (i.e., attempts to avoid thinking about the stressor) reported increased psychological well-being and less stress. In sum, it appears that use of coping strategies and perception of stress play an important role in the adjustment to pediatric chronic illness. However, the precise role of stress and coping in developing resilience has not yet been defined.

Parental depression in JRA. Several studies have explored the link between parental depression and the adjustment of chronically ill children. A longitudinal study of the psychological functioning of parents of children with JRA found that mothers were more depressed than fathers, and maternal depression was associated with poorer psychosocial functioning of the child (Timko, Stovel & Moos, 1992). Furthermore, maternal drinking problems and social isolation predicted more child psychosocial problems one year later. In contrast, more adaptive functioning was associated with children engaging in more peer activities, being socially integrated at school, and attaining higher grades. Another recent study of the mental health of 53 mothers of young children with JRA (2- 11 years) found that over half the mothers reported psychological symptoms and poor maternal mental health was associated the child's social isolation (Lustig et. al., 1996).

Summary

Using a family systems and a developmental framework, this proposed study aims to examine the role of perceived stress in adjustment to JRA; examine the role of family coping, and explore the relation among stress, coping and adjustment; and to compare findings for families with early and late adolescents. Although a number of studies have

explored children's adjustment to JRA, mechanisms accounting for adjustment have not been explained fully. I anticipate that disease severity and perceived stress will be related to adjustment, furthermore, I expect that family coping strategies will modify this relationship.

CHAPTER 2: METHOD

Participants

Sixty-eight families of children diagnosed with JRA participated in the study. The sample included 35 families with early adolescents (10 – 13 years) and 33 families with late adolescents (14 – 19 years). [1] The majority of children were diagnosed with either pauciarticular onset JRA (50%) or polyarticular onset JRA (43%); only seven percent of the children had systemic onset JRA. Nineteen percent of the children were in remission and currently receiving no treatment, and another nine percent were quiescent (that is, children showed no physical signs of arthritis while on medication). However, the majority of children (72%) had active arthritis symptoms and were prescribed a treatment regimen that included one or more medications as well as range-of-motion exercises. Children with active JRA were assessed by the pediatric rheumatologist to have mild (35%), moderate (28%) or severe (9%) arthritis symptoms. There were no differences between early and late adolescents in diagnostic category, disease severity, or prescribed treatment regimen (see Table 1).

Time since diagnosis of JRA ranged from 1 month to 14.5 years, with average disease duration of 4.3 years (early adolescents) and 5.6 years (late adolescents). Nearly half the sample (49%) had been dealing with JRA for an extended period (greater than 5 years), 31% of children were recently diagnosed (less than 12 months), and 21% had arthritis between 1 and 5 years. Compared to late adolescents significantly more early adolescents had been recently diagnosed (Table 1).

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[1] This sample size yields sufficient power (.80) to detect significant differences (alpha = .05) between the two age groups when a medium effect size is expected (Cohen, 1992).

Not surprisingly, given the gender ratio (3:1) for onset of JRA symptoms, 74% of the children were female (Table 2). Family composition for the majority of families (81%) was "two parents with at least one child." Most families were White (75%), with the remainder being African-American (13%), Hispanic (7%), and Asian (4%). There were no differences between early and late adolescents in family composition or ethnicity.

Father's occupation ranged widely: professionals and business managers (36%), to administrative, clerical or skilled work (44%), to semi-skilled or unskilled workers (16%). Most fathers had at least some college education (64%), with 22% graduating from either professional or graduate school. Families of younger adolescents reported higher socioeconomic status (Table 2).

Procedures

Children with JRA and their families were recruited from the Pediatric Rheumatology Division of Schneider Children's Hospital, where a multidisciplinary team (pediatric rheumatologist, nurse specialist, physical therapist, social worker) provides comprehensive specialist care for children with arthritis. All children who were scheduled for regular clinic visits during the 18-month data collection period had their medical files reviewed to assess eligibility for inclusion in the study. [2]

Multiple inclusion criteria were used. Only established patients of the clinic between the ages of 10 and 19 years, who met the American College of Rheumatology (ACR) diagnostic criteria for JRA were eligible for the study. Co-morbidity (i.e., additional medical or psychiatric conditions) or unconfirmed diagnoses were grounds for

exclusion.[3] The first 70 eligible children with scheduled clinic appointments were recruited. These families were sent letters signed by their pediatric rheumatologist explaining the purpose of the study (see Appendix A). In a subsequent phone call, study details were discussed and an interview was scheduled to coincide with the child's next clinic visit. All family members were encouraged to participate in the interview. The response rate was high (97 %), with only two families refusing to participate. One family cited the mother's work commitments; in the other, the adolescent did not feel comfortable discussing her arthritis.

Following the child's medical examination, families were escorted to a small conference room. Family members present at the interview varied. Although efforts were made to encourage participation by both parents, mothers were over-represented in this sample. Clinic hours usually conflicted with parental work schedules and, typically, the mother accompanied the child on regular clinic visits. For the majority of interviews (59%) mothers and their child with arthritis were interviewed together. Fathers participated in 31% of the interviews. Additionally, siblings were present for 15% of these interviews. All families completed informed consent forms and gave permission for the session to be audiotaped before the interview began (see Appendix A). Family interviews lasted from 30 to 65 minutes (M = 44, SD = 5.8).

The Principal Investigator conducted all family interviews. After establishing rapport, family members were questioned about the circumstances surrounding the initial JRA diagnosis. Questions were designed to elicit sequential phases of the family's

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[2] The Human Subjects Committee of the Graduate Center of the City University of New York approved the procedures for this study on July 7, 1995; July 30, 1996; and July 2, 1997.
[3] Co-morbidity information was obtained from children's medical files.

response to the child's diagnosis with arthritis, including: the family's framing of the problem; search for information about JRA etiology and treatment; planning, organization and implementation of solutions to JRA-related stressors; and emotional reactions. Questions regarding the adolescent's functioning in three social domains (family, school, and peer group) were included. Probes were designed to elicit family members' narratives of specific JRA-related experiences, including successes and failures in family adaptation to JRA, rather than using forced-choice questions about specific coping strategies. Throughout the interview there was an emphasis on how all family members had been affected and on the family's functioning as a unit. Questions were directed to all family members in an effort to ensure that a single participant didn't dominate the interview.

After the completion of the interview, the parents and the child with arthritis were asked to complete some written measures. Respondents were asked to refrain from discussing their written responses with each other. The questionnaire packets included questions about disease status, prescribed treatment, illness-related stressors, family functioning, and the child's psychosocial adjustment. Twenty-two families (32%) were unable to complete the written instruments within the time constraints of the clinic visit. Instructions for completing the instruments and a stamped addressed envelope in which to return them were given to these families. Follow-up letters were sent two to three weeks later, and phone calls were made a week after that to discuss any problems in completing the instruments. Slightly over half the families (55%) who didn't complete the written instruments at the clinic returned them within two months. In all, 58 families (85%) had complete interview and questionnaire data.

At the end of the interview session, family members were thanked for their participation and children were given a small gift (e.g., candy, and sports or cosmetic packs). Letters summarizing study findings were sent to families at the conclusion of data analysis.

Measures

The interview protocol was based on the Family Life Events Interview developed by Stuart Hauser (1993) to quantify the ways in which a child's chronic illness disrupts or changes the usual activities of the family. As Hauser's work was with families of diabetics, specific questions and topic areas regarding JRA were developed during pilot interviews with five JRA families. These questions were refined into a structured interview protocol that assessed specific JRA knowledge, treatment compliance, family stress and family coping mechanisms (Degotardi, Revenson & Ilowite, 1999). Interview topics are summarized in Table 3.

Multiple measures were used to assess the variables outlined in the conceptual model presented in Figure 1. The specific measures for each of the major constructs (i.e., personal characteristics, medical characteristics, family stress, family-level coping, family psychosocial adjustment) will be described in detail in the following sections. All measures are summarized in Table 4, and descriptive statistics for all scalar measures are presented in Table 5. To clarify, with the exception of the family-level coping variable that was coded from the transcripts of the family interview, all the measures were self-report. As shown in Table 4 informants included parents, children with JRA, and the pediatric rheumatologist.

Personal characteristics. Parents provided the information on family

demographics, including parents' current occupation and highest educational level attained, marital status, number and age of children in the family, and ethnicity (see Appendix B). A single indicator of socioeconomic status that combined occupational prestige and education was formulated based on the Hollingshead and Redlich (1958) criteria.

Disease characteristics. Following routine examination of the child, the pediatric rheumatologist provided medical information on a standardized form. (A copy of this form can be found in Appendix B). A number of variables were used to reflect arthritis severity. Disease duration, current treatment regimen (no current treatment, medication only, medication and physical therapy [4]) and JRA diagnosis (pauciarticular, polyarticular or systemic onset), were based on criteria described by Cassidy and Petty (1995). A global assessment of arthritis activity was reported: currently in remission (no active symptoms for over two months with no medication); quiescent (no arthritis symptoms while medication was maintained); mild (some episodes of joint swelling and pain); moderate; or severe (unremitting and severe episodes of joint swelling and pain, with restricted joint movement). These criteria comprise the Disease Activity Index (DAI; Varni et. al., 1987), and are based on clinical manifestations of arthritis symptoms (e.g., the presence of joint swelling, warmth, pain, and the range of joint movement). The pediatric rheumatologist also estimated the child's functional ability (fully functional, minimum assistance required, moderate assistance required, or fully incapacitated). Unless otherwise noted the age variable used in analyses was dichotomous: early adolescence and late adolescence.

[4] No children were prescribed physical therapy only.

Functional health. Parents rated their child's ability to complete everyday tasks in eight functional areas using the Child Health Assessment Questionnaire (CHAQ; Singh et. al., 1994). Using a four-point scale ("can do without any difficulty" to "unable to do") parents noted if their child could perform specific tasks (e.g., "turn faucets on and off"), and if their child need devices or aids (such as built up pencils) to perform them. Overall, parents' ratings of their child's level of functioning was high (M = 0.35), on a 0 to 3 scale where 0 indicates no functional limitations. Over half the families (52%) reported that their child was able to perform all activities of daily living without difficulty. Internal consistency for the CHAQ was high (α = .92).

Adolescents reported their current level of pain using a 10cm visual analogue scale anchored by 0 (no pain) and 100 (very severe pain). Average pain reported was 17.4, with a range from 0 to 76.

Family stress. The amount of the stress experienced by families was reported separately by parents and children, using the same measure, the short form of the Perceived Stress Scale (PSS4; Cohen et. al., 1983). Respondents were asked to estimate how frequently they perceived their lives to be unpredictable, uncontrollable, and overloaded using a five-point scale ("never" to "very often") -- for example, "In the past month, how often have you felt difficulties were piling up so high that you couldn't overcome them?" The internal consistency of this 4-item scale was acceptable for parents (α = .68), but the internal consistency was low for children (α = .48). Using the Spearman-Brown Prophecy Formula (Ghiselli, 1977) the scale would need to be 16 times longer (66 items) to achieve an acceptable reliability of .80. The low reliability may be due to the small number of items or, more likely children may be having trouble

understanding the meaning of items. The findings using children's PSS4 scores should be interpreted with caution.

A measure of JRA-specific stress was developed from the five pilot interviews and from stressors reported by Dunkel-Schetter and colleagues (1992) in a sample of cancer patients. The illness-related stressors most frequently cited during the pilot interviews (e.g., "unpredictability of arthritis flares," "restriction of sports or activities due to arthritis") were presented as an 11-item checklist (see Appendix B). Parents and children were asked to indicate (separately) the amount of stress they experienced for each of the 11 items on a five-point scale, ranging from "not at all stressful" to "extremely stressful". Item scores were summed, with higher scores indicating higher levels of JRA-related stress. Internal consistency was acceptably high for both parent's and children's report of JRA-specific stress (Table 5).

Family-level coping. The transcribed family interviews were coded using the Family Coping Coding Scheme (FCCS; Hauser et. al., 1993) to produce the quantitative variables used in analyses. In his original research with families of diabetic children, Hauser coded for 20 family coping strategies that mirror the basic modes of coping found in the adult coping literature (e.g., Lazarus & Folkman, 1984). Two additional strategies were added to the FCCS coding scheme as they appeared important to JRA populations: normalizing and prayer. Normalizing is particularly relevant for adolescents with JRA when obvious arthritis symptoms may make the adolescent feel different from his or her peers. Normalizing strategies include attempts to keep up with normal activities, covering up arthritis symptoms, and rejection of a "sick role" (Weiner 1984). Prayer and holding spiritual beliefs were frequently mentioned coping strategies in the pilot study;

moreover, previous research indicated that prayer was an important source of strength for people with chronic illness, including arthritis (Abraido-Lanza, Guier & Revenson, 1996; Samuelson et. al., 1992). Table 6 provides a brief description of all 22 family-level coping strategies that were used to code the interview transcripts; a more in-depth analysis of family coping is presented in the Results section.

The family was the unit of analysis for coding. The Principal Investigator listened to the audiotape while reading the transcript, marked off key sections, reread the transcript, and coded each speech segment for the coping strategies mentioned by individual family members. In other words, each interview segment was coded for each of the 22 strategies for each family member. Individuals received a score of 0 if they never used the strategy and a score from 1 to 4 if they did use it, depending on the frequency and strength of endorsement of the coping strategy as found in the text. If explicit and numerous examples of the strategy were described then individuals were given a high score for that strategy; if a coping strategy was inferred from the text individuals received a low score. For example, "*I have a great support system in my husband. I never feel like I am alone. We go through everything together.*" is an explicit example of seeking social support and would be given a high score. In contrast, "*My friends know about it (arthritis)*" alludes to the child discussing the JRA with friends and possibly receiving social support from peers, therefore, this phrase would be given a low score for seeking social support. The specific phrases and key words from the text that exemplified each strategy were recorded on summary worksheets. (An example of a summary worksheet can be found in Appendix C).

From these data a global family score, a weighted average, was computed for

each coping strategy. (Coding guidelines are presented in the FCCS manual, Hauser et. al., 1993.) As all family interviews were coded by the Principal Investigator, inter-rater reliability using the Kappa statistic (Cohen, 1960) was calculated for 15% (10) of the transcripts that were coded by a second trained rater. Reliabilities were found to be acceptable (coefficient kappa = .92), and consistent with previous studies using this coding system (Degotardi et. al., 1999; Hauser et. al., 1986, 1993).

The strength of endorsement of each strategy was calculated by determining the mean score. Over half (59%) had mean scores greater than 1.5, indicating moderate use. Means ranged from 0.49 (alternative reward) to 2.38 (seeking information). In addition, to determine whether families used a particular coping strategy at all, each strategy was recoded into a dichotomous variable. A score of 0 indicated that families did not endorse the strategy at all, and a score of 1 indicated the coping strategy was used. Table 7 shows that the most frequently endorsed family coping strategies were "normalizing" (82%), "seeking support" (79%), "seeking information" (78%), and "self reliance" (78%). Less than 30% of families used "diminished awareness of other's feelings", "prayer", or "alternative rewards" at all. Generally, the strategies most frequently endorsed by families were also the most strongly endorsed.

Although it was beyond the scope of this study to undertake a thematic field analysis as outlined by Rosenthal (1993), I did review all family interview transcripts after completing the FCCS coding to search for insights into aspects of family functioning that the family coping codes may have missed. This cursory inspection of the family interviews was interpretative, and involved identifying patterns of regularities in family narratives of the illness experience and common themes or topics. Examples of

topics that were not adequately addressed in Hauser's coding scheme were the impact of the illness on siblings, sources of stress, and issues regarding transfer of responsibility for treatment and management of adherence. These qualitative data were used to illustrate findings and to shed light on surprising results. Exemplary quotes were chosen by noting all families who scored at least one standard deviation above the mean on the measure of interest (e.g., team effort), and rereading those family interviews to selected the best (i.e., succinct and unambiguous) examples.

Psychosocial adjustment. Adaptational outcomes included measures of parent, child and family functioning. Four measures of family psychosocial adjustment were included: treatment adherence, adolescent psychological adjustment, adolescent quality of life, and parental depression.

(1) Treatment adherence. Measures of adherence were obtained from multiple informants -- parents, children, and the pediatric rheumatologist.[5] Questions eliciting a global estimate of adherence to the treatment regimen were adapted from a measure developed by Hayford and Ross (1988). Respondents used a five-point scale (from "never" to "always") to indicate the degree of adherence to medication (e.g., "My child [I/this child] takes medication as often as prescribed") and range-of-motion exercises (e.g., "My child [I/this child] does physical therapy exercises as often as recommended"). These two questions were self-report measures for parents and children, the pediatric rheumatologist's estimate was based on the child's medical history and questioning of the child during the medical examination. Parents and children were asked three additional

[5] Although more accurate measures of medication adherence are obtained by either biological assays (Bearison, 1996) or Track-Caps (pill bottles with a computer chip that records exact time of medication use, Rapoff, 1996), these methods were too costly to be feasible for this dissertation study.

questions regarding treatment adherence: "Please estimate how often your child [you] forgot to take medication in the past week", "Please estimate how often your child [you] forgot to do physical therapy exercises in the past week" both used the response format (never, once, 2 or 3 times, 4 or more times), and "My child [I] remembers to take medication on his/her [my]own". Space was provided for informants to list aspects of the treatment regimen that the child found unpleasant but few wrote responses (see Appendix B).

Adherence to medication and to range-of-motion exercises were examined separately. First, there are important conceptual differences in children's adherence to medication compared with adherence to treatments that necessitate lifestyle changes, such as incorporating exercise into daily schedules. Second, previous studies have shown that adherence to physical therapy is typically lower than adherence to medication (Christophersen, 1994; Hayford & Ross, 1988; LaGreca & Schuman, 1995; Rapoff, 1989). Third, examination of the family interview transcripts indicated that treatment adherence was not a unitary phenomena. Rather, many children reported adherence to medication and then proceeded to discuss reasons for not adhering to range-of-motion exercises (e.g., children found these exercises too time consuming, repetitious, boring, or painful).

Correlations were used to determine the level of agreement among parent, child and pediatric rheumatologist reports of adherence (Tables 8). There were patterns of significant positive correlations (at the $p < .01$ level) among the three sources in estimating adherence to both medication (average correlation = .53, with a range from .48 to .66), and exercise (average correlation = .61, with a range from .39 to .73). Given this

high level of agreement a decision was made to average across sources (parent, child and pediatric rheumatologist) and form two new variables: (average) adherence to medication; and (average) adherence to range-of-motion exercises. Means, standard deviations and item reliabilities are shown in Table 5. Reflecting the literature, mean scores were significantly higher for adherence to medication ($M = 3.7$, $SD = 0.7$), than for adherence to range-of-motion exercises ($M = 2.9$, $SD = 0.9$), $t(56) = 7.3$, $p < .01$. Coefficient alpha was high (>.80) for both scales. In addition, there was a significant positive correlation between adherence to medication and adherence to exercise ($r = .55$), that is, children who took their medication as prescribed were also more likely to report doing their range-of-motion exercises.

(2) Adolescents' psychological adjustment. Social competencies and behavioral problems were assessed by the 112-item Youth Self Report (YSR; Achenbach, 1991). Items describe actual activities, behaviors, and friendship patterns rather than perceptions of competence (e.g., "I would rather be with younger children than children my own age."). Adolescents indicated whether each statement was "very true or often true", "sometimes true", or "not true" of themselves. Potential inflation of YSR internalizing scores due to inclusion of somatic symptoms – in this case, the fatigue, pain, and nausea typically associated with JRA and its treatment-- has been identified (Bearison, 1996; Perrin, Stein & Drotar, 1991). In an attempt to reduce this source of bias, item 41 was modified from "I have the following physical problems with NO known medical cause" to "I have the following physical problems with NO known medical cause (i.e., NOT related to my JRA)". This minor change did not affect the psychometric properties of the YSR and reduced the potential misinterpretation of the question, particularly for those

adolescents reporting somatic symptoms associated with arthritis flares.

Following standard procedures for the YSR, age and gender standardized T-scores for Internalizing (withdrawn, depressed, anxious) and Externalizing (acting-out, aggressive) behavioral problems were calculated and compared with published norms (Achenbach, 1991). The majority of adolescents had scores within the normal range of behavioral and social functioning (see Table 5), and did not differ from those found in previous studies of children with JRA (Daltroy et al., 1992; Harris et al., 1991). Internal consistency was acceptable ($\alpha > .80$) for both Internalizing and Externalizing.

(3) Adolescents' quality of life. Adolescents completed the Satisfaction with Abilities and Well-being Scale (SAWS; Katz & Alfieri, 1997). Respondents used a 5-point likert scale to indicate their level of satisfaction with their ability to perform 13 different tasks. This scale was developed for adults with rheumatoid arthritis and was adapted with minor rewording for adolescents. For example, the item "Ability to shop and do errands" was changed to "Ability to do chores." These changes were discussed with the scale's author (P. Katz, phone conversation May, 1996), and she agreed that this modified scale would be suitable for adolescents. Internal consistency was high ($\alpha = .91$), and these adolescents reported a high level of satisfaction with their ability to engage in routine activities and with their general well-being ($M = 54.4$, $SD = 8.2$).

(4) Parental depression. Parents completed the Beck Depression Inventory (BDI; Beck & Steer, 1988). The BDI consists of 21 statements answered on a four-point scale (e.g., "I get as much satisfaction out of things as I used to" to "I am dissatisfied or bored with everything"). Items probed for cognitive, affective, vegetative and somatic symptoms of depression, and included questions concerning appetite, sleep, feelings of

sadness or guilt, and self-appraisals. Although initially designed as a screening tool, the BDI has been used extensively in research to measure depressive symptoms. Concerns regarding social desirability bias were addressed in a recent review (Conoley & Impara, 1992) and it concluded there was substantial support for the reliability and validity of the BDI in assessing symptoms of depression. In this study, internal consistency for the BDI was good (α = .85).

Summary

This study used a combination of two methodologies to obtain data about stress, family coping, and adjustment in JRA families. Quantitative data regarding family stress and adaptation were obtained from self-report questionnaires administered to parents and children separately. A list of specific measures used is shown in Table 4. The pediatric rheumatologist provided medical information. Qualitative data concerning coping processes were obtained from the family interview. Interviews were transcribed and coded for use of family-level coping strategies. Exemplary quotes from family interviews were used to illustrate findings.

CHAPTER 3: RESULTS

This study proposes that families who have a child with a chronic illness such as arthritis will experiences multiple stressors, and that family-level coping processes are vital in determining whether successful psychosocial adjustment will occur. The model representing these processes is presented in Figure 1. Briefly, demographic characteristics, such as socio-economic status, may affect resources available to families thereby contributing to the levels of perceived stress (path a) and family psychosocial adjustment (path c). Disease status, such as severity or length of illness, is also anticipated to affect family stress levels (path b) and adjustment (path d). Perceived stress is expected to contribute to psychosocial adjustment (path e). This process, in turn, is expected to be influenced by family-level coping (path f).

The findings testing these paths are presented in three sections. First, I present results pertaining to the factors that predict family stress (paths a and b in Figure 1), and analyses that explore whether perceived stress mediates the relationships between disease, personal variables, and adjustment (paths c, d and e). The relative contributions of disease, stress, and coping to adjustment will be examined. Second, the nature of family level coping will be described, and the role of coping in dealing with arthritis-related stress (path f) will be examined. The final section will focus on developmental questions of age differences in the conceptual model. In each section, I briefly review the model focusing on the underlying the paths being tested, describe data analysis procedures, present the statistical findings, and provide relevant qualitative data from the family interviews, where appropriate.

Perceived Stress in Juvenile Rheumatoid Arthritis

This section will describe the types and levels of JRA-specific stress and global stress faced by JRA families, examine the congruence between parent and child reports, and explore the predictors of family stress (Figure 1, paths a and b). It will also examine the relationship among disease and demographic variables, perceived stress, and adjustment (Figure 1, paths c, d and e).

What types of stress do JRA families report?

Perception of global stress was low to moderate: On a scale ranging from 0 – 15, the mean score for parents was 5.88 and for adolescents, 3.96. However, parents perceived higher levels of global stress than did a normative group of healthy adults studied by Cohen and Williamson (1988), $t(2, 296) = 3.8$, $p < .01$.[6] As shown in Table 9, parents reported significantly more global stress than their children. At the same time, there was congruence between parents' and adolescents' reports of total global stress ($r = .43$, $p < .01$). If parents perceived global stress to be high, their children also reported high levels of global stress.

A similar pattern emerged for JRA-specific stress. Parents reported more JRA-specific stress than their children did and again, there was congruence between parent and child reports of total JRA-specific stress ($r = .67$, $p < .01$). On all items except restriction in sports activities, which was high for all respondents, parents reported greater stress than their children. Most parents (over 80%) reported fears regarding disease progression, concern about their child's joint pain and the unpredictability of arthritis flares as sources of considerable stress. In contrast, adolescents identified joint

[6] Normative data on the PSS4 are not available for adolescents.

pain (66%) and restriction in sports participation (62%) as predominant sources of stress. The qualitative data illustrate these different concerns expressed by parents and their children. Parents tended to focus predominantly on future concerns, whereas, their children expressed more immediate and concrete concerns.

> *My first concern was: "Will she [my daughter] be crippled? Is it going to get so bad that she'll be in a wheelchair?" I'm just concerned about all the medicines over the years ... like the methotrexate ... you don't want that around in your system during your childbearing years. I also think, "What is it going to be like for her later in life?" I don't think that at her age she even thinks about later on – even a week from now, maybe what is the next dance, or the next time she is going out with her friends.*

Contrast this mother's quote with that of a young girl (not her daughter):

> *You can't do as many sports that you like – you can't do baseball the way that you have to slide ... and soccer you have trouble kicking. You have to be more careful – you have to run away from your opponents when they are coming!*

In sum, although congruence between parent and child reports was found for both global and JRA-specific stress, there are important differences in what they perceive as stressful.

What are the predictors of family stress?

Two sets of variables – demographics indicators and disease status – were examined as predictors of perceived stress (paths a and b, Figure 1). Given the large number of variables, a series of multivariate analyses of variance (MANOVAS) was

computed to control for Type 1 error. Significant univariate findings will be reported only if the multivariate F statistic is significant ($p < .05$). However, statistical trends ($p < .10$) will be reported to illustrate patterns of relationships among major variables.

Table 10 shows few relationships between demographic variables with either global or JRA-specific stress.[7] In the two cases where a demographic variable was related significantly to a stress measure, the multivariate F approached, but did not reach significance. Parents of boys with JRA reported higher levels of global stress than parents of girls ($M = 7.4$ and 5.3 respectively). Single parents reported higher global stress ($M = 7.2$) than those who were currently married ($M = 5.5$). Additionally, socioeconomic status was inversely correlated with adolescents' reports of JRA-specific stress ($r = -.31$, $p < .05$): Adolescents in families with lower socioeconomic status reported higher levels of JRA-specific stress (but not global stress).

In contrast to the paucity of associations between demographic indicators and stress, Table 11 shows a much stronger pattern of relationships between measures of disease severity and stress. The multivariate F was significant for JRA diagnosis, and approached but did not reach significance for JRA treatment and time since diagnosis. Significant univariate main effects were found for all six medical variables. Duncan post hoc comparisons ($p < .05$) indicated that adolescents with more severe arthritis symptoms (e.g., increased disease activity, multiple joint involvement) and those requiring a complex treatment regimen reported significantly more JRA-related stress than adolescents with less severe arthritis symptoms or treatments. For example, adolescents who were prescribed a treatment regimen of medication and range-of-motion exercises

[7] Analyses were repeated using age as a continuous variable, and there were no changes in findings.

reported almost double the amount of JRA-specific stress (M = 13.2, SD = 8.9) compared with adolescents who were prescribed medication only (M = 6.9, SD = 7.3). Adolescents who were either diagnosed with JRA within the past year or diagnosed between one and five years reported significantly higher stress than adolescents who had been diagnosed for more than five years (M's = 13.8, 14.0 vs. 7.4, respectively).

Overall, JRA diagnosis and functional limitations were the strongest indicators of family stress. Higher levels of family stress were reported for adolescents with polyarticular JRA compared to adolescents diagnosed with either pauciarticular or systemic JRA. Similarly, increased levels of functional limitations were associated with increased perceptions of both global and JRA-specific stress.

In sum, there were few relationships between demographic variables and stress. However, there was a clearer pattern of relationships between disease variables and stress, in particular, arthritis severity and treatment complexity were associated with adolescents' increased perceptions of JRA-specific stress.

What is the relationship among disease and demographic variables, family stress, and adjustment?

Demographic indicators or disease characteristics may affect adjustment by influencing stress levels. Thus two mediational models were tested (represented by paths a, c and e; and paths b, d and e in Figure 1). Baron and Kenny (1986) outlined four conditions that must be satisfied to demonstrate mediation: (1) The predictor must be related to the outcome; (2) The predictor must be related to the mediator; (3) The mediator must be related to the outcome after controlling for the predictor; and (4) The effect of the predictor on the outcome must not be significant after the mediator is

controlled. Satisfying all four conditions would demonstrate complete mediation, that is, the mediator (here, perceived stress) is necessary for the effect to occur. Partial mediation is demonstrated by a significant reduction in the absolute size of the path between the predictor (disease severity) and outcome (adjustment), indicating that the mediator (stress) is a potent but neither a necessary nor sufficient condition for the effect to occur (Kenny, Kashy & Bolger, 1998). These conditions were tested using a series of hierarchical multiple regression analyses (Schiaffino & Revenson, 1992).

To test the first condition of a mediational relationship between disease severity, stress, and adjustment (paths b, d and e in Figure 1), adjustment was regressed on the set of six disease variables. (The disease variables were positively correlated, average r = .32.) Separate equations were computed with the six adjustment measures as criteria. Only two adjustment measures were significantly related to disease variables: adherence to medication and quality of life. As shown in Table 12, the set of disease variables accounted for 20% of the variance in adolescents' adherence to medication and 50% of the variance in adolescents' quality of life. Significant β weights indicated that less disease activity (β = -.33) and greater joint involvement (β = .33) were associated with increased medication adherence. Fewer functional limitations (β = -.64) and longer time since diagnosis (β = .26) were associated with improved quality of life. As the first condition of mediation was satisfied only for adherence to medication and quality of life, testing of mediation can continue with only these two variables.

To test the second condition of mediation, the four stress variables were regressed on the set of medical variables. The full equation was significant for JRA-specific stress (both parents' and children's reports, R^2 = .19 and .29 respectively) and for parents'

reports of global stress (R^2 = .20). In families where the adolescents had been diagnosed more recently both parents and children reported more arthritis-related stress (β's = -.23 and -.30 respectively). Parents reported more global stress if the adolescent had more functional limitations (β = .29).

The results of the third condition testing mediation, path e, are shown in Table 12. The set of six disease variables was entered on the first step of the regression equation, and the set of four stress variables was entered on the second step. Adherence to medication and quality of life were the two criterion variables. The third condition was satisfied for both variables; that is, controlling for disease, the set of four stress variables explained 26% additional variance in adherence to medication and 24% additional variance in quality of life. Interestingly, the significant betas were for adolescents' (but not parents') reports of stress. Lower JRA-related stress (β = -.39) was related to adherence to medication and lower global stress (β = -.43) was related to quality of life.

Tests of the fourth condition required for mediation indicated different results for the two criterion variables. Disease variables remained related to medication adherence after controlling for stress. In other words, stress did <u>not</u> mediate the relationship between disease severity and adherence. And, although the association between disease variables and quality of life remained significant when stress was partialled out, the β for time since diagnosis dropped from .29 to .15 and was no longer significant, indicating partial mediation.

The same procedures were used to test a mediational relationship for demographics, stress and adjustment (paths a, c and e in Figure 1). When adjustment was regressed on the set of six demographic variables no significant relationships were found.

Thus, the first condition of mediation was not satisfied prohibiting any further testing of mediation. In sum, demographic variables were not related to adjustment, and although some of the measures of disease were, stress did not emerge as a strong mediator of this relationship.

Family Coping in Juvenile Rheumatoid Arthritis

Before examining the predictors of family coping and the role of coping in dealing with arthritis-related stress, I will first describe the nature of coping in families with adolescents diagnosed with JRA. Family interviews were coded for use of 22 possible coping strategies. Of these, the mean number of strategies used was 12, and 95% of the sample was coded as using between 7 and 17 strategies. Normalizing the illness experience, self-reliance, seeking support or information about JRA, coordinated family efforts to resolve JRA-related problems, and/or directly expressing feelings were the strategies used by 70% or more of families. The coping strategies used by the majority of families (Table 7, column 2) also were used to a greater extent (Table 7, column 3). In other words, the most common types of family coping strategies were also used more extensively.

In order to reduce these 22 strategies into a more parsimonious set, I examined whether the coping strategies could be grouped in a similar way to those used by Stuart Hauser in his original work with families of diabetic adolescents (Hauser et. al., 1993). Drawing on Lazarus' stress and coping paradigm (Lazarus & Folkman, 1984), Hauser conceptualized family coping as involving three modes: Appraisal-focused coping involves the family's cognitive interpretation of illness-related stressors; Problem-focused coping involves planning of specific actions to deal with these stressors; and

Emotion-focused coping involves efforts by the family to regulate their emotional distress. Basically, these modes can be thought of as how families think about the illness, what actions the family takes to deal with the illness, and how the family feels about the illness. One important question is whether this conceptualization is useful for understanding coping processes among families with chronic illnesses other than diabetes.

An exploratory factor analysis was computed with all 22 coping items, with no limit set regarding the number of factors. This analysis produced six factors with eigenvalues greater than 1, which explained 70% of the variance. However, factors were a combination of all three modes of coping. For example, the first factor contained three appraisal-focused, four problem-focused, and two emotion-focused coping items. In an attempt to force a solution that mimicked the tripartite structure, a second exploratory factor analysis limited the number of factors to three.[8] Again, the factors did not resemble Hauser's three modes -- each factor contained a combination of cognitive, behavioral and emotional items. Two other pieces of evidence suggested that the tripartite structure of appraisal-, problem-, and emotion-focused coping was a poor fit with these data. First, when three scales were created according to the three modes of the original FCCS study, internal consistency reliability was extremely low (all alpha coefficients < 0.29). Second, there was an inconsistent pattern of inter-item correlations among coping items within each mode, as shown in Table 13. Thus, it became necessary to search for a different way of reducing the 22 family-level coping strategies into fewer measures that would be conceptually clearer.

[8] Exploratory factor analyses requesting oblique and orthogonal rotations produced similar solutions.

Going back to the initial exploratory factor analysis, examination of the scree plot suggested that a five factor solution was both empirically acceptable (accounting for 65% of the variance) and understandable. The five-factor solution is presented in Table 14. All 22 strategies loaded on at least one of the factors, only four strategies loaded on more than one factor, and inter-factor correlations were low (see Table 15). This five-factor oblique solution was used to create scales.[9] After reversing scores for negatively loaded items, items on each factor which loaded > .40 were averaged. Internal consistency coeffiecients for the five scales had a wide range (α = .30 to .92).[10]

Summary statistics for each scale are presented in Table 16. The two larger factors are a combination of appraisal-, problem- and emotion-focused items, whereas, smaller factors depict clear appraisal-focused strategies or emotion-focused strategies. Smaller factors had fewer items and lower alpha coefficients. Caution should be used interpreting findings with these latter factors.

Team effort is a combination of all three modes of coping. It describes families working together as a consensual, coordinated unit to maintain a positive outlook and solve problems associated with the child's arthritis. Items describe family self-reliance in problem solving, instrumentality in seeking both social support and information about JRA, and shared family appraisals of arthritis-related stressors. Additional items reflect the family's ability to control impulsive or destructive outbursts of emotion among family members. The percentage of common variance explained was 34.3%. The following

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[9] A five-factor solution with orthogonal rotation yielded an essentially identical structure.

[10] Due to the low internal consistency of Factors 4 and 5, a 3-factor oblique rotation was computed. This 3-factor solution was unsatisfactory, as explained variance dropped (from 65% to 53%), two items did not load on any factor, and there was a lack of conceptual clarity. Therefore the 5-factor oblique solution was retained.

excerpt exemplifies team effort, a mother discusses decision-making processes that include all family members.

> *Literally for three weeks we beat our brains out getting as much data on this (methotrexate) and really just discussing it with everybody in the family and we made a decision. You know we made Karen [11] understand the side effects and risks of it. And we have taken a vote that we are going to hold off for a little while – mainly because she is doing better and we have seen some improvement. And we are going to hang in there and keep doing what we have been doing. We figured that physical therapy would help too.*

Status quo, explaining 10.4 % of the common variance, includes a combination of appraisal- and problem-focused strategies. Items describe families handling arthritis stressors without altering normal routines and rejecting the sick role by covering up obvious arthritis symptoms and not allowing arthritis to assume a central role in the child's life. An additional item describes a refusal to focus on negative outcomes or to allow pessimistic attitudes to dominate. One father described this normalizing attitude succinctly, "*We've tried to keep her in an active but normal lifestyle and not let her arthritis take away or detract from her growing up as a normal person.*"

Emotion processing describes families expressing emotions openly and acknowledging the feelings of other family members. For example, "*Yeah, I was crying and my mom was frustrated... She was very mad at the doctor and I was crying because I thought it [the JRA diagnosis] was like a death thing.*" An additional item described

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[11] Participant's names have been changed to preserve confidentiality.

reliance on prayer or spiritual beliefs; however, this was used infrequently in this sample. This factor explains 8.1% of the common variance.

Cognitive flexibility includes two appraisal-focused items and explains 6.5% of the variance. This strategy describes the family acknowledging multiple perspectives of the stressful situation and weighing different opinions when planning action to deal with arthritis-related problems. For example, "*When I came home with the [JRA] diagnosis [my husband] didn't believe me and he made me go somewhere else for a second [medical] opinion. He didn't accept it at all. I think I was a little more prepared.*"

The final factor, Seeking meaning (explaining 5.7% of the variance), also depicts an appraisal-focused strategy and involves family struggles to develop a personally meaningful -- albeit, not necessarily accurate -- explanation of JRA. Items include recognizing the stressful nature of JRA and its possible ramifications, and focusing on alternative rewards that may result from these struggles. For example, in the following passage, a father describes the family's search for meaning and attempts to understand why his daughter has been afflicted with crippling arthritis.

> *My wife and I have been asking "Why did this happen to Barbara? Is it genetic, or is it a viral thing?" We've been talking with Dr Z and we're really trying to accept that there is no good reason for it. I've abused my body all my life with playing sport ... playing football, and I'm OK. So why did it have to happen to my daughter?*

What are the predictors of family-level coping?

Associations between demographics, disease characteristics, and stress with family coping were analyzed using multivariate analyses of variance (MANOVA) and

correlational analyses. As shown in Table 17 there were few associations between demographic indicators and family coping. There were only two significant relationships found, both with team effort. Duncan post hoc comparisons indicated that families of early adolescents were more likely to use team effort than families of late adolescents (M's = 2.7 and 2.1 respectively). Additionally, higher socioeconomic status was related to greater use of team effort ($r = .29$, $p = .02$).

There were slightly more significant relationships between measures of disease severity and coping than demographic indicators and family coping (Table 18). However, the pattern of relationships between disease severity and coping showed fewer significant relationships and was less consistent than between disease severity and perceived stress (refer back to Table 11). Only three coping variables -- status quo, cognitive flexibility, and seeking meaning -- were related to disease characteristics and the pattern was inconsistent. Disease activity and pain were related to cognitive flexibility. Duncan post hoc comparisons indicated that families with adolescents whose arthritis symptoms were either in remission (M = 2.3) or quiescent (M = 2.8) used significantly more cognitive flexibility than families where the adolescents had mild, moderate, or severe disease activity (M's = 1.7, 1.3 and 1.7 respectively). Additionally, families whose children were in less pain were using cognitive flexibility to a greater degree. The nature of the medical treatment was related to the coping strategy of maintaining the status quo. Families in which adolescents were prescribed either no treatment or medication only (M's = 2.6, and 2.1 respectively) were more likely to use status quo compared to families whose children were prescribed a combination of medication and physical therapy (M = 1.6). Time since diagnosis was related to seeking

meaning: Families with adolescents who had been diagnosed with JRA for five or more years used significantly more seeking meaning (M = 2.2), than those who had been diagnosed less than five years (M = 0.9).

Finally, there was an interesting pattern of negative correlations between stress and coping variables (Table 19). When families perceived lower stress they used more team coping and maintaining the status quo. (Three of the four stress measures were significantly related to status quo.) Although there was a consistent pattern of negative correlations among stress and the three smaller modes of coping, only the relationship between parents' global stress and seeking meaning reached significance.

In sum, there were few relationships between demographic variables and coping. There was a stronger pattern for disease severity and coping, but the pattern of correlations was neither as strong nor as consistent as between disease severity and stress. Thus, the severity of adolescents' arthritis was more strongly associated with stress perceptions (both global and arthritis-related stress) than with family coping. Additionally, lower stress was associated with use of team effort and maintaining the status quo.

The relative contributions of stress, coping and disease severity to adjustment

The pattern of relationships described above leads to the question of whether and to what degree each of these variables contributes to the psychosocial adjustment of JRA families. The next set of analyses considers the full model (Figure 1). Separate hierarchical regression equations were computed with the six measures of adjustment as the criterion variables. In each regression equation the set of six disease characteristics was entered on the first step, the set of four family stressors was entered on the second

step, and the set of five family-level coping strategies was entered on third step. Results for the regression equations are shown in Table 20.

The total R^2's for all six equations were significant. Disease severity, stress and family coping accounted for at least 44% of the variance in adjustment, accounting for 82% of the variance in quality of life. Together, arthritis severity, perceived stress and family coping variables were excellent predictors of adjustment.

Each component and its unique contribution to the prediction of adjustment will be examined next. The set of disease severity measures explained significant variance in two of the six adjustment measures, adherence to medication (20%) and quality of life (50%). As reported previously, increased joint involvement and lower disease activity were associated with greater adherence, and adolescents with fewer functional limitations and those who had been diagnosed with arthritis for an extended period of time reported higher quality of life. Perception of stress explained significant variance in four of the six adjustment measures: adherence to medication (27%), parental depression (40%), internalizing behavior (34%), and quality of life (24%). The change in R^2 approached, but did not reach, significance for a fifth outcome, externalizing behavior. Adolescents' (but not parents') reports of less JRA-specific stress were related better adherence (β = -.39), but, increased arthritis-related stress was associated with more internalizing behaviors (β =.62). Lower levels of adolescents' global stress were related to better quality of life (β = -.43). Higher levels of both parent's and adolescents' overall stress were related to increased parental depression (β's = .33 and .27 respectively).

Once disease severity and stress were controlled, family coping added significant unique variance to the explanation of only two adjustment measures: Externalizing

behavior (24%) and quality of life (8%). Family use of team effort was associated with lower levels of adolescents' externalizing behavior (β = -.53), and use of cognitive flexibility (β = -.33) was associated with reduced quality of life.

In sum, measures of disease severity alone were not good predictors of adjustment. However, arthritis severity combined with stress and coping measures accounted for large proportions of variance in all adjustment measures. Levels of perceived stress appeared to be the strongest predictors of adjustment, and family coping appeared to have minimal influence. However, this finding is somewhat ambiguous, for in an additive model the last variable to be entered into the regression equation shares common variance with all preceding variables and may have little unique variance. For example, disease severity and stress don't contribute much of the explained variance in externalizing, moreover, their shared variance with coping is low so the unique variance explained by family coping added after these variables in the equation is substantial (24%) and significant. In contrast, disease and stress contribute significantly to adherence and shared variance with coping is high (20% and 27% respectively). In this latter instance, coping only contributes an additional 7% of unique variance. Thus, by partialling out disease severity and stress variables, coping may not have an "opportunity" to explain a lot of the variance in adjustment.

Does family-level coping help in dealing with arthritis-related stress?

Although the above analyses suggest that family coping has a minimal relationship to adjustment, they test only for direct (or main) effects. To clarify what role (if any) family-level coping plays in managing the stresses of pediatric arthritis I will next examine how stress and coping interact to influence adjustment. One possibility is that

family coping is differentially effective at different levels of stress perceived by the family – a moderational effect. The stress buffering hypothesis (e.g., Thoits, 1986) suggests that in times of high stress, families who use a specific coping strategy should show better adjustment than families who do not use that strategy. To explore if family coping serves as a stress-buffer, separate hierarchical multiple regression analyses were computed for each of the six adjustment measures. In all equations, adolescents' perception of JRA-related stress was entered on the first step.[12] The set of five family coping variables was entered on the second step, and a set of five interaction terms composed of the product of adolescents' JRA-specific stress and one of the coping variables was entered on the third step. A significant interaction term, in the predicted direction, would indicate a moderator effect (Cohen & Cohen, 1983).

To avoid the multicollinearity problems that may occur when two continuous variables form an interaction term, deviation scores were created for stress and coping variables by subtracting the sample mean from each respondent's raw score. This procedure is known as centering. The interaction term was the product of the two centered scores. In centered regression equations the interaction effect, that is, the R^2 change and associated significance tests, is the same for raw and centered scores. However, the constant (A), unstandardized regression coefficients (B), and the main effects of the variables constituting the interaction change (Finney, Mitchell, Cronkite & Moos, 1984).

The results of these regression analyses are presented in Table 21. The R^2s for

[12] Previously discussed results indicated that adolescents' (not parents') perception of stress was predominantly related to both disease status and adjustment measures; therefore, only adolescents' JRA-related stress was entered into the regression equation.

the full equation were significant for five of the six adjustment measures; moreover, the R^2 was significant for the set of stress-by-coping interaction terms for three of the six outcomes: adherence to medication, externalizing behavior, and quality of life. Although the set of interaction terms was significant for externalizing behaviors, none of the individual stress-by-coping interactions reached significance. Therefore, only significant interactions for adherence to medication and quality of life were examined in detail. Looking at each interaction term with the set, the β weights that were significant for adherence to medication were: stress-by-team effort (β = -.40) and stress-by-status quo (β = .40). The following β weights were significant for quality of life: stress-by-team effort (β = .35), and stress-by-seeking meaning (β = .31). To illustrate the nature of each interaction, regression lines were plotted following procedures recommended by Cohen and Cohen (1983) using centered scores (Finney et al., 1984). High and low stress scores were calculated as one standard deviation above and below the mean.

Three of the four plots in Figures 2 (b, c, and d) depict similar cross-over effects. Showing the direct effect of JRA-related stress, adjustment was higher when perceived stress was low and adjustment was compromised when stress was high. However, when families perceived high levels of stress, their use of team effort, seeking meaning, and maintaining the status quo ameliorated the impact of the stress. In other words, there was evidence of stress-buffering. Conversely, when perceived stress was low, use of these coping strategy was associated with somewhat lower adjustment. As depicted in Figures 2b and 2c, when adolescents' perception of stress was high use of the family coping strategies of team effort and seeking meaning were associated with higher quality of life. Similarly, at times of high stress maintaining the status quo was associated with improved

medication adherence (Figure 2d).

A somewhat different effect emerged for the interaction of stress and for adherence to medication. As shown in Figure 2a, when JRA-specific stress was high, use of team effort did not affect adherence. However, when adolescents perceived stress to be low, the use of team effort made an important difference, increasing medication adherence. In this instance, team coping did not have the predicted stress-buffering effect, instead, it's impact was strongest during times of relatively little illness-related stress.

The results outlined in the preceding section demonstrated the nature of coping in JRA families. Multiple strategies were used by families to deal with the complexities of arthritis disease progression, and five modes of family coping were identified. In general, perceived stress and disease severity, but not demographic indicators, were related to coping, and together these variables were good predictors of adjustment. Although low stress was strongly related to better adjustment, when stress was high use of family coping was associated with better medication adherence and improved quality of life.

Age Differences in the Conceptual Model

The final study aim was to examine any differences between early and late adolescents in stress, coping and adjustment processes. In this last section, I will explore the question of developmental differences, examining how the age of the child with a chronic illness may influence the family's response to illness-related stressors and affect adjustment.

To examine differences between early and late adolescents across all the variables in the conceptual model, six separate MANOVAs were calculated, with the sets of

demographics, disease characteristics, stress, family coping, and adjustment variables as the criterion variables. Age was the between-group factor. Results of the six MANOVAs are presented in Table 22. The pattern of relationships was inconsistent and sparse. The multivariate F statistic was significant only for demographic indicators and for family coping. Univariate statistics indicated that families with older adolescents were more ethnically diverse and reported lower socioeconomic status than families of younger adolescents (M's = 31.2 and 37.6 respectively). Families of younger adolescents used the coping strategy of team effort to a greater extent. Surprisingly, there were no differences between families with younger and older adolescents in measures of disease severity, perceived stress, or adjustment.[13]

Additionally, the overall multivariate F statistic for disease variables was not significant, nor was there even a (univariate) difference in the length of illness between early and late adolescents (M's = 51 and 66 months respectively). It had been expected that the younger children would have been diagnosed for a shorter time. However, the variation was extremely great on this variable for both groups. To examine this relationship more precisely, length of time since diagnosis was trichotomized into adolescents who were recently diagnosed (<12 months), diagnosed between 1 and 5 years, or diagnosed more than 5 years. The chi-square for age with this trichotomy was significant, χ^2 (2) = 15.18, $p < .01$. Approximately half the early- and late-adolescents had been diagnosed with JRA for more than five years (45.7% and 51.5% respectively). However, early adolescents were four times as likely to be recently diagnosed with JRA

[13] The univariate statistics did yield two significant differences in adjustment: Older adolescents reported more externalizing and internalizing behavior problems than younger adolescents; however the multivariate F was not significant.

(48.6%) compared with late adolescents (12%). Because of the complex relationship between age and time since diagnosis the MANOVAs were repeated with both age and time since diagnosis as independent variables. With these analyses the unique contributions of age and time since diagnosis, as well as their interaction, can be explored. If the length of illness influences how families deal with illness-related stress, then the age effects found in Table 22 may disappear, and instead, significant main effects could emerge for either time since diagnosis or the interaction between age and length of illness.

Table 23 shows few significant relationships. There were no significant interactions between age and time since diagnosis. Similar to the first sets of MANOVA results there were no significant relationships between age or time since diagnosis with stress or adjustment variables. The multivariate statistic was significant only for one of the three analyses, family coping, but the main effect for age that emerged was not the relationship between age and team effort reported in Table 22. Instead, once length of illness had been removed statistically, families with older adolescents used more status quo than families with younger adolescents (M's = 2.0 and 1.7 respectively). Those families whose child had been diagnosed for a longer time used more team effort, and this effect was independent of the child's current age.

To summarize, the paucity of significant differences between families with early- and late-adolescents on any of the variables in the model was perplexing. Although re-analyzing the data to include length of illness helped to clarify the relationship between age and family coping, surprisingly, time since diagnosis did not have a potent influence on levels of stress or psychosocial adjustment.

Summary

Overall, there were sizable differences in the amount and type of stress reported by parents and their children with arthritis. Parents reported concerns regarding their child's future, whereas, adolescents focused on more immediate concerns (e.g. sports restrictions). Adolescents' (but not parents') reports of JRA-specific stress were strongly related to concrete indicators of disease severity. Specifically, severe JRA and treatment complexity were related to increased stress, and controlling for disease severity, lower stress was associated with better adherence to medication and improved quality of life.

Families used multiple coping strategies, and lower stress was associated with family use of team effort and maintaining the status quo. Together disease severity, stress and family coping were good predictors of most adjustment outcomes, explaining between 44% (internalizing) and 82% (quality of life) of the variance. Moreover, coping moderated the relationship between disease severity and adjustment: At high levels of stress family coping acted as a stress-buffer. Specifically, family use of team effort or seeking meaning was related to improved quality of life, and efforts to maintain the status quo were related to improved adherence. Unexpectedly, there were few difference between early and late adolescents, and no interaction between the child's age and length of illness on any of the measures in the conceptual model.

CHAPTER 4: DISCUSSION

The purpose of this study was to examine the stressors experiences by families of adolescents with JRA and to explore if family-level coping helps to ameliorate the impact of chronic illness on family functioning. Moreover, I sought to examine if there were differences in stress, coping and adjustment processes for families of early adolescents compared with families of late adolescents. Briefly, I found that adolescents' cognitive appraisals of illness-related stressors were a central component in the psychosocial adjustment of JRA families, and that family-level coping strategies played an important stress-buffering role at times of high stress. Surprisingly few empirical differences were found between early and late adolescents.

The critical role of stress appraisals in adjustment to JRA

Perception of arthritis-related stress emerged as a critical variable. Unexpectedly, perceived stress was more strongly associated with both disease severity and adjustment than was use of specific modes of family coping. So, why do perceptions of stress play such a central role for adolescents with arthritis? Lazarus' early work on stress and coping (1981) suggests that cognitive appraisals, especially, the importance of the meaning of the stressor to the respondent are critical in the coping process. Lazarus emphasized the need to take into account psychological mediators such as appraisals to understand stress reactions, noting that possible cognitive appraisals of the stressor included assessments of harm or loss, threat, and challenge. The family's primary appraisal of the seriousness of the arthritis threat would include evaluations of potentially harmful impact of arthritis on both the child and family. If the probability of joint damage and functional loss is assessed to be high, family perceptions of stress are

correspondingly high. In contrast, viewing the JRA as a challenge or an opportunity for personal growth in the face of adversity (e.g., "*It's made us stronger and closer as a family.*") may lower levels of perceived stress.

Secondary appraisals of the stressors associated with JRA include assessments of the family's resources to meet illness-related demands and the available coping options. There is a dynamic interplay of appraising the threat of JRA stressors and evaluating the efficacy of family response. For JRA families, appraisals of the adequacy of family resources are important. Deficits in tangible resources, such as money, can exacerbate perceptions of stress, with families assessing their resources to be insufficient to guarantee maximal health care for their child. For example, adolescents in families with lower socioeconomic reported higher levels of JRA-specific stress. These families often experienced difficulties paying for expensive medications, health insurance and assistive devices such as paraffin baths. As exemplified by the following dialogue between mother and daughter, arthritis-related demands can exhaust limited resources.

> Daughter: *My mom she used to work, but now she doesn't work, she's a housewife now. There's a lot of things to it. She has to be protective of me, she's always protecting me, and oh yeah, we have to be careful with the food. Sometimes it's hard for her to remember appointments. Sometimes we forget and that's why we don't come, or there's a problem with the ambulance. They [social services] were threatening us ... they might put me in an institution or something like that [for missing clinic appointments]... but they should blame the social worker, it's not our fault that the ambulance didn't come.*
>
> Mom: *Me, I want my daughter to see the doctor for everything, but I have no work...*

many problems. I am so frustrated ..., the ambulance, the telephone..., the speaking [difficulty communicating in English].

It is apparent that for this family, problems of severe disease activity are compounded by having inadequate resources (e.g., the family is unable to afford a car to attend appointment, hence the need for ambulance transport) and the mother's inability to communicate with health care workers.

Interestingly, although parents reported more overall stress, parental stress was not linked systematically with either severity of their child's medical condition or psychosocial adjustment. Parents are more cognizant of the long-term consequences of disease progression, and primary appraisals of JRA include a heightened perception of potential loss of joint function and threat to their child's normal development. Secondary appraisals involving an evaluation of available resources and coping options may inflate stress appraisals. For parents, high global stress was associated with reports of increased depression. Parental ratings of global stress may in fact be an appraisal of their coping efficacy. For example, a mother with high stress expressed a feeling of being overwhelmed by the demands of her daughter's medical condition, "*I've been trying to do all that she needs, but I feel guilty that I haven't been able to take her to physical therapy. Maybe, because I'm a single parent I think it's a little harder... I'm not at home as much as I'd like to be with her, and by the time I get home there's so much to do – it's either cleaning up or dinner. You know she's tired and I'm tired.*"

Given the extensive requirements of managing their child's chronic illness, it's not surprising that parental stress was higher than that reported by a community sample (Cohen & Williamson, 1988), and higher than their children's perceived stress. In the

family interviews, parents expressed concerns about the impact of JRA on their child's career choices, and speculated about the possibility of long-term disability, joint damage, and the effect on their child's reproductive capacity. Parents need to plan and coordinate their child's treatment, negotiate with insurance companies, and act as an advocate for their child. One mother discussed the high stress related to one of these illness-related tasks: "*It's been very upsetting, I've been working on getting [insurance] approval for the past two weeks, but there's so much red tape. It's so complicated.*"

In contrast to their parents, adolescents perceived lower levels of stress, and stress was more strongly related to indicators of arthritis severity, such as degree of functional limitations and complexity of the treatment regimen. It is not surprising that adolescents with active and severe symptoms experienced more JRA-specific stress. Adolescents diagnosed with polyarticular JRA reported higher stress than those with pauciarticular JRA. The former not only have more joints affected by arthritis, but also often have more inflammation, pain and consequent disruption to regular routines, and restriction of activities. Similarly, treatment complexity was associated with greater disruptions to adolescents' routine. Prescribed range-of-motion and strengthening exercises need to be performed daily and are time-consuming. In the following excerpt a mother describes how her daughter's quality of life has been severely compromised and the distress associated with her inability to perform tasks of daily living. "*It's hard for her to shampoo her hair or get dressed... She's very achy and she gets very upset and cries a lot. She's having a hard time ... she can do it, but she can't do it well, so a lot of times she asks me to help.*"

Most adolescents with JRA dealt in the "here and now" and expressed little

concern about their future and the possible negative impact the illness could have on their adult life. The findings suggest that adolescent's perception of stress is based predominantly on concrete markers of disease severity and the restriction of sports or social peer group activities that are central to adolescents' emerging sense of self. These represent potential losses to adolescents, especially of independence and autonomy, and a current threat to their ability to lead a normal life. Reflecting some of the developmental issues typical of the struggle for autonomy and search for identity, adolescents focused more on the impact of joint pain and actual limitations to their lifestyle, especially difficulty keeping up with peer group activities. Other adolescent concerns included the interference caused by the demands of the treatment regimen, and the effect of arthritis on their body image: "*I can't rollerblade any more because my ankle starts to hurt... and I can't do cartwheels*", "*It did bug me that I couldn't walk well enough, but that's it really. I was just concerned about my looks.*" Many adolescents expressed apprehensions about being different from their healthy peers, and one male with polyarticular JRA eloquently stated his annoyance with arthritis interfering with his life, "*When I flare up, I get turned off by everything. I get angry and nothing really excites me.*"

In many family interviews it was apparent that family conflict often entailed struggles by the adolescent to assert independence and assume control of decision-making regarding the treatment regimen and reluctance of his or her parents to acquiesce. Parents cited a fear of poor adherence as a reason for not yielding to adolescents' demands for increased autonomy. "*It's just that she's a little inconsistent (with taking medication), and I always have to ask her. Lately it seems that she's a little better, but sometimes I don't actually know if she takes it (medication).*" It is likely that family

conflict over transition and responsibility for adherence issues contributed to adolescents' perception of stress.

In many ways treatment adherence was a "hot" issue both for families and the rheumatology treatment team. Although good adherence was a shared objective, there were many barriers to adherence. There was the common problem of forgetfulness and difficulty incorporating treatment regimens into the adolescents' busy lives. It was apparent from the interviews that families who lacked a mutually agreed upon routine for adhering to the treatment regimen were less compliant and reported frequently missing doses of medication. More successful families relied upon strategic placement of pill bottles as a visual prompt (most frequently by the bedside or on a kitchen counter), and incorporated pill-taking into mealtime or bedtime routines. Several families had devised ingenious methods that incorporated cognitive-behavioral components that specified rewards to facilitate adherence (e.g., diaries, refrigerator charts), "*She had to take so many aspirin, up to 15 each day, and we had to make sure that she had them after meals so that she wouldn't get stomach problems... We kept a chart on the refrigerator and we marked it all down, because it had to be given at different times of the day.*"

Congruent with previous findings, adherence to range-of-motion exercises was even more problematic than medication adherence (Hayford & Ross, 1988; Rapoff, 1989). Several factors could contribute to this. Families often mentioned that taking medication was easier to fit into their routines, and believed that medications were stronger and more effective than exercise in combating JRA. Adolescents reported that range-of-motion exercises often hurt, and that sets of repetitions were boring and took too long to complete. Many adolescents believed that regular sports and physical activities

were an acceptable substitute for the recommended exercises, and parents didn't counter this belief. Families appeared to have little appreciation that specific strengthening and flexibility exercises were prescribed to prevent muscle atrophy around affected joints. This lack of understanding of the benefits of exercise has been reported in other studies (Berry et. al., 1993; Hayford and Ross, 1988).

The role of family-level coping in adjustment to pediatric chronic illness

I have argued that the level of the family's perceived stress predominantly influenced adolescents' adjustment to arthritis; however, the combination of disease status, stress and family coping was an even better predictor of adjustment. At least 40% of the variance in each measure of adjustment was accounted for by a combination of these variables. Although disease status and family stress accounted for most of the explained variance in adjustment, family coping contributed significant unique variance to two adjustment measures. Use of team effort was associated with less externalizing or acting out behavior of adolescents, and unexpectedly, low family use of cognitive flexibility was related to better quality of life. Team effort involves many instrumental coping strategies such as seeking support and maintaining an optimistic outlook. These strategies have been found to be associated with better psychosocial outcome for both adults with arthritis (Felton & Revenson, 1984; Manne & Zautra, 1989; Newman & Revenson, 1993; Parker & Wright, 1997), and children with JRA (Ebata & Moos, 1991; Konkol et. al., 1989; Timko, Stovel & Moos, 1992). Therefore, it is not surprising that team effort was associated with fewer adolescent adjustment problems. The relationship between cognitive flexibility and quality of life is less apparent. Examination of the family interviews suggests that multiple perspectives in defining the problem and in

formulating a plan of action may be too confusing for adolescents, adding to perceptions of stress. Instead, an agreed upon family plan may promote better adjustment. The following dialogue between a mother and her daughter illustrates the potential stress generated by opposing views of the value of seeking information about JRA: Mom, "*For myself personally, the more I read the better I feel about the whole thing (JRA).*" *In contrast, the daughter states, "The more I read the more I know how bad it (arthritis) can get, not how better it can get....because, I heard that like 9 out of 10 kid could get rid of it, but maybe I am the 1 in 10.*"

As previously discussed, in an additive model family coping shares variance with both disease and stress variables and may have limited opportunity to explain much unique variance in adjustment. When disease severity and perceived stress were partialled out, the impact of family-level coping as a predictor of adjustment may have been minimized. So what precisely is the role of family coping in adjustment? Generally, family coping was a stress-buffer; that is, it ameliorated the negative effects of high stress. When adolescents' perception of stress was high, adherence to medication was greater for families who tried to normalize their child's illness experiences. Similarly, adolescents' reports of their satisfaction with their abilities and well-being was greater when families used team effort or tried to understand and seek meaning in the illness experience. The one exception to the stress-buffering effect of coping was that when stress was low family use of team effort was associated with better treatment adherence. Apparently, at times of extreme stress, families working together as a consensual unit to solve problems was not enough to counter the negative impact of stress.

As illustrated in the following excerpt, normalizing (or maintaining the status quo) is a useful coping strategy, related to concept of family resilience, that is, the ability of the family to regroup and bounce back to normal level of functioning after a crisis (Hawley & DeHaan, 1996).

We've tried to keep her in an active but normal lifestyle and not let the illness take away or detract from her growing up as a normal person. She's picked up on that herself and kind of taken off on it. She's always involved in all sorts of things. Now she's involved in community service.

During the interviews several families related with pride how they were able to overcome adversity associated with severe disease progression. These families were somewhat akin to the individuals described by O'Leary and Ickovics (1995) as thriving in the aftermath of a potentially life-changing event. These families were able to reframe the JRA positively as a challenge, and typically had a family schema that included a shared sense of family values, with an emphasis on "we" not "I". In an example of this enhanced family functioning, the family of a 14 year-old boy discusses dealing with the stress of hospitalization, complex treatment regimens and unpredictable arthritis flares.

He is one of the few kids who's still on liquid gold. He's responding to it, so why mess with the treatment? We went through a lot of years of pain ... physical pain, emotional pain, and a lot of stress. My wife and I and my other son, we all stuck together. We had to deal with this. We had to deal with a lot of crazy friends and relative who didn't understand the disease and would give us all kinds of crazy advice ... oh, do this or go see that doctor, go take vitamins, try acupuncture ...But I think one of the biggest problems was that we had to educate his teachers ... so we spoke

with all his teachers.... The parent really has to take the lead and be an advocate for the child.... My wife and I, we always worked together and we always encouraged him to do as much as he can. When he wanted to do it, we always tried to give him opportunities.

As well as maintaining an optimistic outlook and being pro-active in their efforts to help their son engage in normal activities, these parents also regularly attended an arthritis support group. Their son comments, "*Most people don't even realize that kids get arthritis ... You've just got to try and hopefully it will get better. You've got to deal with it... It doesn't phase me now. I don't even really think about it at all.*"

It is probably no coincidence that the family coping strategies of team effort and maintaining the status quo were found to be most strongly associated with measures of adjustment. These two strategies both had good reliability and little error variance, whereas, the other three coping modes were comprised of a small number of items and had poor reliability. Therefore, it is possible that I didn't really tap the full potential of family-level coping. More stress-buffering effects of coping may have been found if there was less error variance in the three weaker family coping modes.

Importance of studying family coping

There was really no reason to expect that modes of family-level coping would have a similar structure to modes of individual coping. The dynamics of family interaction include family members negotiating the meaning of JRA, striving to reach a consensus regarding the best course of action, resolving conflict, and managing emotional reactions. Trying to capture the family context and the flavor of family interaction with a family interview involves an additional layer of complexity. In moving

from the individual to the family level of analysis the process is seen through multidimensional lenses. However, the themes and functions of coping remain familiar (Lazarus & Folkman, 1984) – a combination of cognitive appraisals, emotional regulation and instrumental actions.

The interview data reflected the fact that families do not limit themselves to one or two types of coping. Lazarus found that "effective copers use both problem-focused and palliative coping modes" (1981, p. 198), and that "coping is not a single act but a constellation of my acts and thoughts engendered by a complex set of demands that may stretch out over time." (1981, p. 201). For a complex situation, such as dealing with a child's chronic illness, it is likely that coping strategies within a single modality -- cognitive, behavioral, or emotional -- would not be able to address the multiple issues involved. For example, to handle the common difficulty of adherence to treatment regimen, the more adaptive family response involved all three modalities, cognitive appraisal of the possible barriers to adherence, behavioral strategies to implement the agreed upon regimen, and awareness of the child's emotional reaction to the requirements of the regimen and to the parent's "nagging."

Multiple modes of coping can be assessed reliably using the quantitative family interview. The instrument can capture nuances in the coping efforts made by different families, providing richer information than questioning family members separately. Through careful reading and rereading of the transcripts, the researcher can reconstruct the dynamics of the coping processes, as opposed to simply obtaining one person's viewpoint. The interview not only provides information from multiple perspectives, but also an integrated "discussion" around a specific issue. Eliciting multiple informant's

opinions in a "public" fashion provides a built-in system of checks and balances. I found that family members would often contradict or challenge each other regarding the veracity of a situation, opening the topic for a fuller discussion. With this methodology, family members were allowed to explain their viewpoints and counter each other's statements. In fact at times the exchanges between family participants became quite heated, as illustrated in this debate between the parents regarding the tapering of their son's medication.

Mom: "He's improved a lot since he's been on the prednisone. So that's one of my fears, how's he going to do when they start reducing the medicine."

Dad: "See, why is that a fear? That's what I don't understand. You'd just as soon keep him on the medicine. Why? The doctor's already arranged a schedule to cut the medicine back. I don't want him on those levels of medicine."

By establishing rapport and striving for a nonjudgmental interview climate, families were encouraged to share both successes and difficulties they have had in dealing with the demands of JRA. Indeed, I heard many instances of both positive and negative family interactions, including admissions that one's own way of coping with the arthritis wasn't helpful. As one woman confided, "*I feel like the bad mother. I feel like I did something wrong that I just can't get her to do the exercises, and I when she doesn't take her aspirin it's my fault because I should tell her more. I just can't remember it all.*"

I had expected that some family members, particularly adolescents or fathers, might be reluctant to express emotions or particular beliefs in the presence of other family members. However, adolescents forthrightly admitted their poor adherence, their concerns with body image and peer relationships, and their displeasure with parental

over-protectiveness. Some families treated the interview as an opportunity to vent frustrations, or to explore sensitive issues related to dealing with JRA. Family members even felt comfortable admitting their exasperation with the pediatric rheumatology team:

> *You know it's a big hassle to get here [the rheumatology clinic] and I never feel like it's very productive. I don't know what I expect sometimes, I'm not looking for personal attention. I just find that it's not very informative. It used to drive us crazy because we'd make this big effort to get here and my husband would take off from work and we'd have the residents coming in... and you know there'd be more time spent teaching them. You'd have to go through the whole story... meanwhile you walk out of there, and yeah OK they [the residents] got a lot out of seeing Diana and learning about her, but what did I come away with?*

I did observe that siblings needed to be encouraged to participate in the interview. This may have been due to the young age of most siblings, or the interview's focus on the adolescent's JRA. Extra efforts may be needed to bring siblings into the conversation. In spite of the 'silent voices' of siblings, the impact of the illness on siblings was mentioned frequently. Common themes were the intensification of sibling rivalry and feelings of being neglected, similar to those reported by Madan-Swain and colleagues in a study of siblings of cancer patients (1993). One young girl described her sister's reaction, "*My sister didn't like all the attention I was getting. She wished that she had arthritis for a while! She would get mad at me, and that would make me feel worse. Because it was like she was blaming me for something that I couldn't control.*" The mother in a different family discussed her children's feelings: "*I was very protective of James and even his sisters were jealous. I was paying more attention to him than them. He comes*

first. They are all the same, but because he has arthritis I had to pay more attention to him and watch him carefully. So the others thought they were neglected."

JRA presents families with a variety of coping tasks that go beyond individual solutions. It is important for researchers to move toward studying the complete family as the unit of analysis, and studying interpersonal as well as intrapersonal processes. The quantitative family interview is a useful research tool for doing this.

Developmental differences

Although I anticipated differences between families of early adolescents as compared with late adolescents there were, in fact, few differences. Families of older adolescents were more ethnically diverse and reported lower socioeconomic status, and families of younger adolescents used more team effort. Socioeconomic status and use of team effort were associated with differences in stress and adjustment across the whole sample, but there were no age differences in any of the stress or adjustment variables. Given the well-documented differences between the developmental tasks of early and late adolescence, and the maturational changes in cognitive, emotional, social and physiological development, the paucity of empirical findings was puzzling. For example: Why didn't families of older adolescents report greater stress than the more financially secure families of younger adolescents? Any potential differences between the age groups may have been "washed out" because the younger adolescents had JRA for a shorter length of time, and recency of diagnosis was associated with increased stress.

One explanation for the paucity of developmental differences is the possible developmental delay of adolescents with severe arthritis. Medication side effects, extended periods of arthritis flare, and parental over-protectiveness may contribute to

developmental delays, and thereby dilute differences between the two age groups. Alternatively, dealing with the challenges of chronic arthritis may result in children being "wise beyond their years". This too could dilute potential age differences. It is possible that dealing with a chronic illness such as JRA is an equalizer, in that all children have to deal with stressors such as severe joint pain, activity restrictions, and the demands of a complex treatment regimen. The commonalities of these experiences may contribute to early and late adolescents reporting similar responses on measures of stress, coping and adjustment.

Additionally, major methodological problems relating to the use of discrete categorical variables for age and length of illness may contribute to the paucity of empirical findings of developmental differences. The two designated age groups (early and late adolescence) may contain too wide an age range to detect meaningful developmental differences. I had criticized previous JRA studies for ignoring developmental issues and studying adjustment in children ranging in age from 4 to 17 years, and attempted to address this problem by limiting my study to early vs. late adolescents. However, even this dichotomization may have been too broad, and did not map onto known changes in either cognitive or social development. Perhaps grouping adolescents into more homogenous groups with respect to cognitive, emotional or social developmental status would shed light on differences in stress, coping and adjustment.[14]

Similarly, illness duration is potentially a potent marker of developmental changes in adjustment to pediatric chronic illness. In JRA, there are distinct illness

[14] Such analyses were not done with these data as the groups would have been too small to have adequate statistical power. Cohen (1992) recommends a sample of 180 subjects to detect significant differences ($p < .05$) between four groups when a medium effect size is expected.

milestones that may be important for adjustment. The first milestone is the onset of symptoms and subsequent search for a plausible diagnosis. Other illness milestones are family accommodation to the demands of the treatment regimen, stabilization on medication, and the unpredictable cycle of arthritis flares and remission. Transitions at these major illness markers could herald heightened perceptions of stress and changes in patterns of family coping as the family moves from destabilization to an adjustment to the new illness status.

At best, length of illness is a crude marker of these transitions. Trichotomizing this variable into those diagnosed less than a year, 1 – 5 years, and more than five years did not directly map onto illness milestones. A better research design would be to delineate each illness milestone and sample by marker, or to group the sample according to these markers.

Given these methodological shortcomings, the question of developmental differences remains unanswered. Future research should identify and focus on these developmentally sensitive illness-related issues. The duration of JRA may be a better indicator of the family's adjustment than the child's age. A larger sample or a longitudinal design could better explore the questions of developmental changes in the adolescent and the disease, and how they affect adaptation.

Although few empirical differences were found, the family interviews point to developmental differences in adjustment to JRA that may not have been captured in the empirical analyses, and deserve further study. Younger adolescents were dependent on parental supervision of the treatment regimen, and concrete concerns relating to school and activities predominated. One 11 year-old described restrictions during arthritis flares:

"*There'll be certain times when my wrists or ankles will be hurting, and I'll say to my gym teacher 'I got to sit out now'... My teachers know that if my wrist is swollen that I may need help with the writing because I can't write – it'd be sloppy stuff.*" A mother describes her complete dedication to supervising her 11 year-old daughter's treatment regimen. "*It was a hard time for Kathy, because the pill was so big ... I had to crunch it and mix it with a flavor she likes. So I put chocolate syrup in it and down it goes!....I keep all the pills right there in the kitchen, because that's where I spend my time, and there they are one bottle after another. Before we leave school she takes the naprosan. There was a period of time when she had to take it three times a day, and I had to run to school with it at lunch time.*"

In contrast, many older adolescents had assumed full responsibility for adherence. They were increasingly cognizant of the long-term implications of JRA, mirroring more closely their parent's concerns. Issues of living away from home, college, career possibilities, and transfer to adult rheumatology care became salient for this group. A 19 year-old male with severe polyarticular JRA showed a mature understanding of how disabling arthritis has impacted his future plans.

> *After surgery I'll make plans (for college), I've just been putting this off until all the surgeries were taken care of... I've had three spinal fusions and two jaw surgeries ... that's why I haven't been able to start college. Perhaps I'll do a business course. I know I couldn't do one of those jobs like a carpenter or a construction worker, but I can still do writing. My writing hasn't changed much, I've pretty much adapted with my hand. I've had to learn how to do things differently. I've had to experiment.*

This young man poignantly describes his desire to find a suitable life partner.

There's only a certain type of girl that would be attracted to me. She would have to get to know me, so that she can get beyond that, and that's difficult for most. What is normal, and what is not normal, I accept that. Someone who is willing to hear what I have to say and not see what I am.

Limitations of the study

As discussed in the previous section, one of the major limitations of this study was the use of the age of the child as a surrogate for developmental status. Because the two designated age groups (early and late adolescence) did not directly map onto changes in either cognitive or social development, I was not able to detect any meaningful difference in stress, family coping or psychosocial adjustment based on the child's age. Furthermore, the distinction between the two age groups may have been further obscured by arthritis-related lags or gains in physical, or emotional maturity.

The cross-sectional design of this study precluded any conclusions regarding causality or directionality among measures of perceived stress, family coping and adjustment. Although it is acknowledged that family coping is a process that changes over time in response to the child's stage of development and disease progression, these changes were not adequately captured in this study. A longitudinal design would be necessary to understand the dynamic relationship among disease status, perceived stress, family coping and psychosocial adjustment.

Although efforts were made to include all family members the source of family informants was predominantly mothers and their child with arthritis. In only 15% of the interviews were both parents and the child present. This related to the difficulty both parents had in taking time off work for a routine clinic visit. Telephone interviews with

the absent parent would not capture the dynamics of the family interaction that I considered vital to the family interview. Evening or weekend interviews were considered, but families were already burdened by the frequency of clinic visits and were reluctant to schedule additional appointments.

The retrospective nature of the family interviews is potentially problematic. Families were asked to discuss how they coped with the initial diagnosis and sequential phases of response. Over time families may accommodate to dealing with the chronic stress and day-to-day hassles of JRA (Repetti & Wood, 1997) and may not notice that dealing with JRA requires special coping efforts.

This study relied predominantly on self-report measures. Obviously, self-report is subject to bias and the tendency to distort responses to enhance self-image. However, some families did report some negative events, such as adolescents acting out or episodes of non-compliance. The concern that treatment adherence would be overestimated was minimized by the high correlation between parent, child and physician estimates of adherence, and the different estimates for medication and exercise compliance within the same family. This was confirmed during family interviews, when families openly discussed the reasons for differential rates of adherence. As discussed previously, the family interview was an important tool in countering individual tendencies of self-enhancing bias.

Clinical implications: Applying findings to intervention

The study's focus on family context and the findings that family members are affected somewhat differently by adolescents' arthritis suggests that a family approach to intervention efforts could be beneficial. Typically pediatric psychologists are referred

children with chronic illness who have been identified by the medical team as having problematic adjustment, including high-risk behaviors such as acting out or nonadherence to treatment regimens. I envisage an expanded role for pediatric psychologists in early intervention efforts to identify 'at-risk' families, reduce family stress, and help family members develop the resources to cope effectively with JRA-related stressors. In particular, siblings need to be included as they often feel neglected or resentful of the special attention given to the ill child.

Reduction of stress should be an important component of the family intervention, as perceived stress was a critical factor in adjustment. Adolescents' reports of stress were related to disease severity and the degree of functional limitations. While medical interventions directly address disease status by prescribing treatment to reduce joint inflammation and improve functional abilities, psychological interventions could complement this by helping families change or reframe their cognitive appraisals of the illness experience. For example, instead of viewing the arthritis as a source of threat and potential loss, families could be helped to regard the arthritis as a challenge and an opportunity for both individual growth and the strengthening of family bonds. Encouragement could be given to family successes in meeting the challenges of pediatric arthritis and developing efficacious coping strategies.

The finding that the deleterious effects of high stress can be modified by use of family coping strategies such as team effort and status quo implies that clinical efforts should focus on helping families to normalize the illness experiences and adopt active problem-solving approaches. Although family therapy typically focuses on improving communication between family members, for JRA families it was not the open

expression of emotions that seemed to facilitate adjustment; rather, it was families working together as a cohesive unit to deal with stressors and solve problems. Family team effort involves a constellation of instrumental activities including strengthening social support networks and accessing accurate information about JRA. To improve adolescents' quality of life, families could encouraged to search for ways of finding meaning in the illness experience and incorporating arthritis challenges as part of their family narrative.

Whereas family encouragement of the adolescent to maintain as normal as lifestyle as possible helped to improve adherence rates, at times of high stress use of family team effort was insufficient to promote adherence behaviors. One important component of team effort is gathering information about JRA, and previous studies have found that for many chronic pediatric conditions education alone is often ineffective in promoting adherence (see Bearison 1994, 1996 for review of findings). Surprisingly, even when children with JRA had been exposed to long-term efforts to facilitate knowledge about arthritis there is a pattern of "striking inaccuracies and misconceptions" regarding disease etiology and treatment benefits (Berry et. al., 1993)

To bolster adherence, the best intervention might be to focus on reducing the family's appraisals of stress and replacing negative cognitions with more positively framed appraisals. Rapoff and colleagues (1988, 1989) found that for children with JRA, educational efforts combined with cognitive-behavioral strategies helped to improve adherence. In family interviews it was evident that families who lacked an agreed upon plan to incorporate the treatment regimen into daily activities had more problems with adherence. This suggests that compliance could be improved by working with families to

help them formulate individualized family-level coping approaches. Such approaches would incorporate the treatment regimen into regular family routines and make use of cognitive-behavioral strategies such as charting scheduled and rewarding compliance. Transition of responsibility for adherence from the parent to the adolescent is another potentially critical issue that could be addressed. Clinicians could work with families to make plans to ensure smoother transitions and circumvent the dramatic drop in adherence rates.

Clearly, although intervention to enhance family coping is important to facilitate adjustment to JRA, the most significant factor is the family's perception of stress. Thus, stress reduction techniques are essential in any intervention efforts. The focus must be on family's cognitive stress appraisals of the illness experience.

Directions for future research

This study developed a solid methodology combining quantitative and qualitative data to explore important issues in family adjustment to pediatric arthritis. One of the strengths of the family interview was that it allows the transactional nature of the family process to be examined. Given the richness of family interview it would be worthwhile to focus on expanding the family-level coping coding scheme to document aspects of family functioning in relation to illness that were not adequately captured in the FCCS. For example, codes could be developed to address issues of family dynamics, family resilience, the role of siblings, and sources of family stress.

Although the interview data was collected for a specific purpose, that is, stress and family coping in JRA, it may be possible to return to this wealth of narrative data to explore other questions. For example, the family interview could be coded to examine

the development of self in adolescents with JRA, and how chronic illness affects the self-perception and self-concept of early and late adolescents.

A second avenue of research would be to examine whether the primacy of cognitive appraisals of the illness experience is an important factor in the psychosocial adjustment of children with different chronic illnesses. The generalizability of the stress-buffering effects of family coping strategies such as team effort, maintaining the status quo, and seeking meaning could also be explored with other pediatric populations.

A final area that warrants further research efforts would be to use a longitudinal design to explore how family coping processes and perceptions of stress change as a function of both the child's developmental stage and illness stage. Adolescents' developmental status needs to be better conceptualized (based on cognitive, emotional or social developmental level) and the issue of illness transitions needs to be addressed. Only then could questions of an interaction between developmental status and disease progression, and anticipated changes in perceived stress and family coping over time be examined adequately. A recent longitudinal study of families of children with cancer (Dalquist et.al.,1996) provides a useful model for examining how family functioning changes as families accommodate to the initial shock of diagnosis and develop routines for dealing with the illness stressors.

Table 1

Disease Characteristics of the Sample

Disease characteristics	Total sample %	Stage of adolescence: Early adolescent %	Stage of adolescence: Late adolescent %	Age differences
JRA diagnosis				χ^2 (2) = 0.29, p = .86
Pauciarticular	50.0	51.4	48.5	
Polyarticular	42.6	40.0	45.4	
Systemic	7.4	8.6	6.1	
Disease activity				χ^2 (4) = 7.95, p = .09
Remission	19.1	14.3	24.2	
Quiescent	8.8	5.7	12.1	
Mild	35.3	45.7	24.2	
Moderate	28.0	20.0	36.4	
Severe	8.8	14.3	3.1	
JRA treatment				χ^2 (2) = 4.97, p = .08
No treatment	14.7	11.4	18.2	
Medication only	23.5	14.3	33.3	
Medication & exercise	61.8	74.3	48.5	
Time since diagnosis[1]	59.4 (48.3)	51.9 (50.2)	67.3 (45.5)	t (66) = -1.32, p = .19
Pain VAS [2]	17.4 (21.6)	20.9 (24.3)	13.8 (18.0)	t (63) = 1.35, p = .18
Functional limits [3]	0.35 (.59)	0.39 (.62)	0.31 (.56)	t (63) = 0.50, p = .61

Note. Total sample n = 68. early adolescents n = 35. late adolescents n = 33.

[1] Average months since diagnosis of JRA

[2] Higher scores indicate higher reported levels of pain.

[3] Higher scores indicate greater functional limitations.

Table 2

Demographic Characteristics of the Sample

Demographics	Total Sample %	Stage of Adolescence: Early adolescent %	Stage of Adolescence: Late adolescent %	Age differences
Gender				χ^2 (1) = 3.22, p = .07
Females	73.5	82.9	63.6	
Males	26.5	17.1	36.4	
Parents marital status				χ^2 (1) = 2.03, p = .15
Married	80.9	74.3	87.9	
Not married	19.1	25.7	12.1	
Children in family				χ^2 (3) = 2.61, p = .46
One child	11.8	5.7	18.2	
Two children	47.0	51.4	42.4	
Three children	35.3	37.2	33.3	
Four children	5.9	5.7	6.1	
Family ethnicity				χ^2 (3) = 4.67, p = .19
White	75.0	85.7	63.6	
Black	13.2	8.5	18.2	
Hispanic	7.4	2.9	12.1	
Asian	4.4	2.9	6.1	
Family socioeconomic status [1]	34.4	37.6	31.2	t (66) = 2.17, p = .03

Note. Total sample n = 68, early adolescents n = 35, late adolescents n = 33.

[1] Family socioeconomic is based on the Hollingshead & Redlich (1958) formula for combining occupation and educational levels. Higher score indicate higher SES.

Table 3

Interview Protocol of Topics Discussed in Semi-structured Interviews

Section A: Knowledge of JRA etiology and treatment

- description of symptom onset, the search for diagnosis, disease severity
- emotional reaction of family to diagnosis, formation of plans, action taken
- development of family theories of JRA onset
- search for JRA information -- books, Arthritis Foundation, internet, clinic staff

Section B: Treatment regimen and compliance issues

- description of treatment regimen (medicine, exercise)
- level of treatment compliance -- estimate of times medication was missed each week
- who is responsible for treatment adherence; strategies for remembering medication
- family relationship with health professionals

Section C: Effect of JRA on school/work and peers

- specific JRA difficulties at school -- absenteeism, writing, gym, carrying books
- restriction and special allowances made by teachers/friends
- exploration of peer network – reaction of friends (e.g., supportive, teasing)
- any changes in physical appearance, or social relationships due to JRA
- interference with sports or social activities

Section D: Family functioning

- impact of JRA on family functioning -- changes in routines, activities or relationships
- how family deals with arthritis flares
- explore family concerns regarding JRA , and possible over-protectiveness
- reaction of siblings -- supportive, protective, jealous of extra attention

Section E: Adaptation and adjustment

- what aspect of having JRA has caused the most problems for the family
- how well does family think they are doing in coping with arthritis
- advice to other families who have a child recently diagnosed with JRA

Table 4

Summary of Measures used in the Study

		Who completes measure		
Measure	Construct	Parent	Child	MD
Socioeconomic status (SES)	Parental education & occupational level	X		
Family information	Family demographics	X		
JRA diagnosis	JRA onset type (pauiarticular, polyarticular, systemic)			X
Disease Activity Index	JRA severity			X
Child Health Assessment Questionnaire (CHAQ)	Functional limitations in daily activities	X		
Perceived Stress Scale (PSS4)	Global perceived stress	X	X	
JRA-specific Stress	Perceived arthritis stress	X	X	
Family Coping	Family-level coping strategies (team effort, status quo, emotion processing, cognitive flexibility, seeking meaning)	X	X	
Treatment adherence	Adherence with prescribed treatment (medication & physical therapy)	X	X	X
Youth Self-Report (YSR)	Adolescent psychological adjustment (internalizing & externalizing behaviors)		X	
Satisfaction with Abilities and Well-Being (SAWS)	Adolescent quality of life		X	
Beck Depression Inventory (BDI)	Parental depression	X		

Table 5

Descriptive Statistics for Measures Included in the Study

Measures	N	Mean	SD	Range	(∝) Reliability
Functional limitations (CHAQ)	65	0.4	0.6	0 - 2.5	.92
Pain	65	17.4	21.6	0 - 76	
Global stress (PSS4 - child)	68	4.0	2.4	0 - 9	.48
Global stress (PSS4 - parent)	67	5.9	2.8	0 - 13	.68
JRA specific stress (child)	66	10.7	8.8	0 - 37	.90
JRA specific stress (parent)	67	16.9	9.2	0 - 40	.87
Adherence to medication	67	3.7	0.8	1.6 – 4.6	.84
Adherence to exercise	58	2.9	1.1	1 – 4.6	.88
Internalizing (YSR)	68	48.5	10.3	26 - 72	.81
Externalizing (YSR)	68	46.5	9.7	25 - 72	.82
Quality of life (SAWS)	65	54.4	8.2	33 - 65	.91
Parental depression (BDI)	68	5.8	5.1	0 - 21	.85

Table 6

Description of Family-level Coping Strategies

Family coping strategy	Definition
Cognitive flexibility	Family considers multiple perspectives of stressors.
Family consensual unit	Shared family appraisals of stressors and treatment plan.
Closed-minded	Disregards others opinions (e.g., MD), or blames others.
Collection of individuals	Detached relationships among family members.
Theory construction	Shared beliefs (not necessarily accurate) of disease etiology.
Avoidance	Avoidance, downplaying or denial of problems.
Positive outlook	Family is optimistic, focusing on positive outcomes.
Negative outlook	Pessimistic, focusing on negative outcomes, helplessness.
Recognition of facts	Recognize stressful nature of JRA and its ramifications.
Normalizing strategies	Rejection of sick role, keeping up appearances.
Self reliance	Family is competent and emphasize their past successes.
Inability to manage	Inability to manage demands of JRA, cite past failures.
Coordination	Coordinated family efforts to manage JRA demands.
Seeking information	Attempts to seek information about JRA.
Support seeking	Attempts to elicit support (financial, material, emotional).
Established ways	Handle JRA demands without altering daily routine.
Alternative rewards	Alternative rewards have emerged as by-products of JRA.
Direct expression	Open and direct expression of feelings to resolve problems.
Acknowledge feelings	Acknowledge feelings of family members.
Diminishing awareness	Insensitivity, or inhibiting of emotions among family.
Impulsive expression	Impulsive outbursts or self-destructive behaviors
Prayer	Prayer or reliance on spiritual beliefs.

Table 7

Family's Endorsement of Family-level Coping Strategies, and Age Group Differences

	Endorsement of family coping		
	Percentage [1]	Strength [2]	
Mode of family coping		Mean (SD)	Age group differences [3]
Cognitive flexibility	30.9	0.69 (1.14)	t (66) = -0.36, p = .72
Consensual family	64.7	1.76 (1.48)	t (66) = 1.88, p = .07
Close minded	35.3	1.07 (1.59)	t (66) = -1.01, p = .32
Collection of individuals	35.3	0.99 (1.44)	t (66) = -1.80, p = .08
Theory construction	55.9	1.32 (1.37)	t (66) = -0.50, p = .62
Avoidance	52.9	1.59 (1.64)	t (66) = -2.72, p = .01
Positive outlook	66.2	1.68 (1.48)	t (66) = 0.89, p = .38
Negative outlook	64.7	1.68 (1.49)	t (66) = -0.11, p = .91
Recognition of facts	70.6	1.65 (1.29)	t (66) = 0.07, p = .94
Normalizing	82.4	2.21 (1.34)	t (66) = -1.31, p = .20
Self reliance	77.9	2.13 (1.52)	t (66) = 1.67, p = .10
Inability to manage	45.6	1.46 (1.70)	t (66) = -0.85, p = .40
Coordinated family	72.1	2.00 (1.51)	t (66) = 1.30, p = .20
Seek information	77.9	2.38 (1.49)	t (66) = 0.92, p = .36
Seek support	79.4	2.26 (1.45)	t (66) = 1.30, p = .20
Established ways	47.1	1.07 (1.33)	t (66) = -1.58, p = .12
Alternative reward	23.5	0.49 (0.97)	t (66) = -2.05, p = .05
Direct express emotions	72.1	1.81 (1.35)	t (66) = 0.30, p = .76
Acknowledge feelings	63.2	1.50 (1.29)	t (66) = 1.23, p = .22
Diminished awareness	27.9	0.74 (1.29)	t (66) = -3.39, p = .01
Impulsive outbursts	38.2	1.03 (1.41)	t (66) = -1.93, p = .06
Prayer/spiritual beliefs	27.9	0.72 (1.26)	t (66) = -1.31, p = .20

[1] Percentage of families endorsing this particular family-level coping strategy to any extent (i.e., strategy was scored > 1 using the FCCS coding scheme).
[2] Mean score indicates how strongly each family-level coping strategy was endorsed overall (range: 0 = no use of strategy, to 4 = strong endorsement of strategy).
[3] Differences between early and late adolescents on each of the 22 family-level coping strategies.

Table 8

Intercorrelation Among Parent, Child, and Pediatric Rheumatologist Ratings of Adherence

	1	2	3	4	5
Adherence to medication (n = 67)					
1. Child	--				
2. Parent	**.48****	--			
3. MD	**.48****	**.66****	--		
Estimate of times medication missed/week (n = 67)					
4. Child	.72**	.45**	.43**	--	
5. Parent	.35**	.56**	.60**	**.51****	--
Adherence to range-of-motion exercises (n = 57)					
1. Child	--				
2. Parent	**.73****	--			
3. MD	**.39****	**.50****	--		
Estimate of times range-of-motion exercises missed/ week (n = 57)					
4. Child	.76**	.65**	.40**	--	
5. Parent	.65**	.74**	.52**	**.66****	--

Note. Numbers in boldface represent correlations among sources on the same measure, i.e., validity coefficients

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

Table 9

Comparison of Parent and Child Reports of Stress

	Parent (n = 67)			Child (n = 66)			
Type of stress	Mean	SD	%	Mean	SD	%	T-test
Global stress	**5.88**	**2.8**		**3.96**	**2.5**		**t (66) = 5.54 ****
Lack control	1.73	1.1	85.3	0.93	1.0	54.4	t (66) = 5.40 **
Many problems	1.12	0.9	73.5	0.94	1.0	60.3	t (66) = 1.16
Not your way	1.48	0.9	83.9	1.12	0.9	75.1	t (66) = 2.90 **
Pile difficulties	1.55	1.1	80.9	0.97	0.9	60.3	t (66) = 4.12 **
JRA specific stress	**16.89**	**9.2**		**10.71**	**8.9**		**t (64) = 6.78 ****
Future fears	2.15	1.2	88.3	0.98	1.1	55.9	t (64) = 8.37 **
Physical limitation	1.82	1.3	75.0	1.22	1.1	60.3	t (64) = 4.00 **
Arthritis pain	2.14	1.3	85.3	1.51	1.3	66.1	t (64) = 4.17 **
Family problem	0.80	0.9	48.6	0.34	0.7	19.1	t (64) = 3.78 **
Unpredictable flare	1.83	1.3	80.9	1.18	1.2	57.3	t (64) = 3.99 **
School difficulties	1.45	1.3	64.7	0.72	1.0	39.8	t (64) = 5.12 **
Frequent clinic	1.51	1.3	70.6	1.03	1.2	51.5	t (64) = 2.90 **
Financial burdens	1.20	1.4	53.0	0.60	1.2	24.9	t (64) = 3.69 **
Treatment demand	1.43	1.3	70.6	1.14	1.1	60.3	t (64) = 2.12 *
Disrupted routine	1.03	1.1	58.9	0.65	1.0	38.3	t (64) = 3.22 **
Sports restrictions	1.54	1.4	66.1	1.34	1.2	61.8	t (64) = 1.21

[1] Percentage of respondents reporting some stress (i.e., scores of 1 to 4) for each item.
* $p < .05$ ** $p < .01$

Table 10

Relationships Among Individual Demographic Variables and Stress

		Univariate F			
	Hotelling's	JRA-specific stress		Perception of global stress	
	Multivariate F	Child	Parent	Child	Parent
Child's age group	F(4,60) = 1.5, p = .22	F(1,63) = 1.2, p = .28	F(1,63) = 0.0, p = .96	F(1,63) = 1.7, p = .20	F(1,63) = 1.1, p = .29
Child's gender	F(4,60) = 2.4, p = .06 +	F(1,63) = 0.2, p = .64	F(1,63) = 0.9, p = .37	F(1,63) = 1.2, p = .28	F(1,63) = 7.4, p = .01**
Family size	F(4,60) = 0.9, p = .53	F(3,61) = 0.8, p = .49	F(3,61) = 0.2, p = .88	F(3,61) = 0.8, p = .51	F(3,61) = 1.3, p = .28
Parent's marital status	F(4,60) = 2.3, p = .07 +	F(1,63) = 0.8, p = .37	F(1,63) = 2.0, p = .17	F(1,63) = 0.3, p = .56	F(1,63) = 4.2, p = .05 *
Family SES [1]		r = -.31 *	r = -.04	r = .05	r = -.11
Family ethnicity	F(4,60) = 0.4, p = .95	F(3,61) = 0.6, p = .59	F(3,61) = 0.2, p = .88	F(3,61) = 0.6, p = .59	F(3,61) = 0.8, p = .48

[1] Correlations are reported for the relationship between measures of stress and family SES (socioeconomic status).
* p < .05 ** p < .01 + p < .10

Table 11

Relationships Among Individual Disease Characteristics and Stress

		Univariate F			
	Hotelling's	JRA-specific stress		Perception of global stress	
	Multivariate F	Child	Parent	Child	Parent
JRA diagnosis	F(4,60) = 2.5, p = .02*	F(2,62) = 6.6, p = .01**	F(2,62) = 4.2, p = .02*	F(2,62) = 3.0, p = .06	F(2,62) = 4.8, p = .01**
Disease activity	F(4,60) = 1.3, p = .23	F(4,60) = 3.0, p = .02*	F(4,60) = 2.1, p = .09	F(4,60) = 0.5, p = .77	F(4,60) = 1.2, p = .32
JRA treatment	F(4,60) = 1.7, p = .09 +	F(2,62) = 4.6, p = .01**	F(2,62) = 2.5, p = .09	F(2,62) = 0.1, p = .86	F(2,62) = 1.7, p = .19
Time since diagnosis	F(4,60) = 1.9, p = .06 +	F(2,62) = 4.9, p = .02*	F(2,62) = 0.9, p = .39	F(2,62) = 2.6, p = .08	F(2,62) = 1.6, p = .21
Pain VAS [1]		r = .32, p = .01 **	r = .20, p = .12	r = .16, p = .21	r = .13, p = .31
Functional (CHAQ)		r = .37, p = .01 **	r = .28, p = .02 *	r = .25, p = .05 *	r = .32, p = .01**

[1] Correlations are reported for the relationship between pain and stress, and functional limitations and stress.

* p < .05 ** p < .01 + p < .10

Table 12

Mediational Analyses with Adherence to Medication and Quality of Life as Criterion Variables

		Adherence Medication			Quality of Life		
Step	Variables entered	ΔR^2	F	Beta	ΔR^2	F	Beta
Step 1	Set of 6 disease variables	.20	2.37*		.50	7.42**	
	JRA diagnosis			.33 *			.19
	Disease activity			-.33 *			.06
	JRA treatment			.02			.08
	Time since diagnosis			.11			.26*
	Pain			-.02			-.11
	Functional limitations			.20			-.64**
Step 2	Set of 4 stress measures	.26	6.50**		.24	9.08**	
	JRA-specific stress (child)			-.39*			.03
	JRA-specific stress (parent)			-.24			-.15
	Global stress (child)			.04			-.43**
	Global stress (parent)			-.18			-.06

Table 13

Intercorrelations among the 22 family-level coping strategies

	AF1	AF2	AF3	AF4	AF5	AF6	AF7	AF8	AF9	AF10	PF1	PF2	PF3	PF4	PF5	PF6	PF7	EF1	EF2	EF3	EF4
AF1	--																				
AF2	-.21	--																			
AF3	-.19	-.21	--																		
AF4	.07	**-.72**	**.37**	--																	
AF5	-.01	-.16	.13	.12	--																
AF6	-.08	**-.55**	.19	**.47**	.16	--															
AF7	.06	**.56**	**-.27**	**-.47**	-.11	**-.62**	--														
AF8	.03	**-.43**	**.49**	**.51**	.07	**.33**	**-.63**	--													
AF9	.13	.17	-.08	-.02	.01	**-.30**	**.34**	-.08	--												
AF10	-.08	**.29**	-.22	**-.32**	-.16	-.14	**.48**	**-.51**	.03	--											
PF1	.08	**.60**	**-.36**	**-.58**	-.24	**-.53**	**.67**	**-.66**	.22	**.34**	--										
PF2	02	**-.43**	**.54**	**.52**	.09	**.37**	**-.67**	**.79**	**-.28**	**-.51**	**-.69**	--									
PF3	-.04	**.72**	**-.34**	**-.70**	**-.28**	**-.49**	**.69**	**-.61**	**.26**	**.41**	**.72**	**-.66**	--								
PF4	.13	**.39**	**-.31**	**-.33**	-.13	**-.42**	**.51**	**-.33**	**.38**	**.29**	**.57**	**-.47**	**.52**	--							
PF5	.09	**.51**	-.23	**-.42**	-.16	**-.38**	**.58**	**-.32**	**.44**	.21	**.48**	**-.42**	**.60**	**.44**	--						
PF6	-.09	.07	-.13	-.09	-.11	.07	.08	**-.44**	-.05	**.64**	.15	**-.33**	.10	-.01	-.09	--					
PF7	.06	.20	.04	-.09	.18	-.05	**.27**	-.22	**.33**	.21	.20	-.15	.23	.16	.17	.06	--				
EF1	.21	**.26**	.04	-.12	-.15	-.12	**.31**	.01	**.25**	.21	**.27**	-.14	**.36**	**.28**	**.35**	.05	.15	--			
EF2	-.01	**.36**	-.18	-.18	-.14	-.04	.22	-.15	**.30**	.22	.24	**-.34**	**.42**	.22	**.36**	.05	.22	**-.43**	--		
EF3	-.20	**-.45**	**.29**	**.47**	-.03	**.61**	-.51	**.25**	-.17	-.12	**-.45**	**.34**	**-.45**	**-.30**	**-.39**	**.26**	-.06	-.14	-.18	--	
EF4	-.03	**-.38**	**.37**	**.35**	.07	**.34**	**-.45**	**.55**	-.07	**-.26**	**-.49**	**.54**	**-.45**	**-.37**	**-.26**	-.18	.00	-.09	-.03	.23	--
EF5	-.02	**.26**	.09	-.20	-.14	-.13	**.27**	-.07	.23	.08	**.29**	-.02	**.28**	.22	**.26**	-.10	.19	**.39**	.13	-.13	-.11

Note. Numbers in boldface (above .26) represent significant correlations ($p < .05$)

Labels designate mode of coping: AF (appraisal-focused), PF (problem-focused), EF (emotion-focused)

Table 14 Five Family Coping Factors Derived from Oblique Factor Analysis

Items in factor	Label	1	2	3	4	5
1. Team Effort						
Collection individual	AF4	-.81	.00	.00	.22	.10
Avoidance	AF6	-.81	-.24	.00	.00	.00
Consensual family	AF2	.79	.00	.20	-.36	.00
Diminished awareness	EF3	-.76	-.35	.00	-.17	.00
Self-reliance	PF1	.72	-.21	.15	.00	.00
Coordinated family	PF3	.72	-.19	.31	.00	.00
Positive outlook	AF7	.68	-.17	.16	.00	.21
Inability manage	PF2	-.55	.52	.00	-.20	.00
Impulsive outbursts	EF4	-.53	.29	.15	-.12	.00
Seek Support	PF5	.50	.00	.41	.12	.00
Seek information	PF4	.45	.00	.26	.29	.00
2. Status Quo						
Established ways	PF6	-.23	-.89	.00	.00	.00
Normalizing	AF10	.10	-.76	.17	.00	.00
Negative outlook	AF8	-.54	.61	.19	.00	-.11
3. Emotion Processing						
Direct expression	EF1	.00	.00	.81	.12	.00
Acknowledge feelings	EF2	.00	-.18	.61	.00	.00
Prayer	EF5	.17	.15	.56	-.25	.00
4. Cognitive Flexibility						
Cognitive flexibility	AF1	-.14	.15	.13	.80	.00
Close mind	AF3	-.37	.28	.18	-.51	.19
5. Seek Meaning						
Alternative reward	PF7	.00	-.19	.18	.00	.75
Theory construction	AF5	.00	.00	-.47	.00	.71
Recognition facts	AF9	.00	.00	.39	.30	.47

Note. Labels designate: AF (appraisal-focued); PF (problem-focused); EF (emotion-focused).

Table 15

Intercorrelations of the Five Factors from the Oblique Rotation Factor Solution

	Factor				
	1	2	3	4	5
Team effort	--				
Status quo [a]	-.24	--			
Emotion processing	.25	.00	--		
Cognitive flexibility	.17	.00	.00	--	
Seek meaning	.00	.00	.15	.00	--

[a] In this matrix a high score on Factor 2 signifies low use of maintaining the status quo.

Table 16

Summary Statistics for Coping Scales

Coping scale	K	M	SD	α
Team effort	11	2.40	1.1	.92
Status quo	3	1.87	1.1	.77
Emotion processing	3	1.34	1.0	.59
Cognitive flexibility	2	1.80	1.1	.30
Seek meaning	3	1.14	0.8	.35

Note. Scale scores were computed by summing across items and then dividing by the number of items in order to permit comparability of means; thus, each scale ranges from 0 – 4.

Table 17

Relationships Among the Individual Demographic Variables and Family Coping

	Hotelling's	Univariate F				
	Multivariate F	Team effort	Status quo	Emotion processing	Cognitive flexibility	Seeking meaning
Child's age group	F (5,62) = 3.2, p = .01	F(1,66) = 5.9, p = .03	F(1,66) = 1.1, p = .29	F(1,66) = 0.1, p = .81	F(1,66) = 0.3, p = .58	F(1,66) = 1.1, p = .30
Child's gender	F (5,62) = 1.0, p = .43	F(1,66) = 0.3, p = .87	F(1,66) = 0.6, p = .44	F(1,66) = 2.9, p = .10	F(1,66) = 0.9, p = .36	F(1,66) = 0.2, p = .70
Family size	F (5,62) = 0.8, p = .71	F(3,64) = 0.3, p = .87	F(3,64) = 0.1, p = .94	F(3,64) = 0.3, p = .82	F(3,64) = 0.7, p = .59	F(3,64) = 1.9, p = .13
Parent's marital status	F (5,62) = 1.3, p = .19	F(4,63) = 1.3, p = .26	F(4,63) = 2.0, p = .11	F(4,63) = 0.6, p = .65	F(4,63) = 0.5, p = .70	F(4,63) = 0.7, p = .61
Family SES [1]		r = .29, p = .02	r = .13, p = .28	r = .15, p = .22	r = -.02, p = .89	r = -.05, p = .70
Family ethnicity	F (5,62) = 0.5 p = .94	F(3,64) = 0.4, p = .73	F(3,64) = 1.5, p = .22	F(3,64) = 0.9, p = .43	F(3,64) = 0.1, p = .97	F(3,64) = 0.2, p = .91

[1] Correlations are reported for the relationship between family coping and family SES (socioeconomic status).

Table 18

Relationships Among Individual Disease Characteristics and Family Coping

	Hotelling's	Univariate F				
	multivariate F	Team effort	Status quo	Emotion processing	Cognitive flexibility	Seeking meaning
JRA diagnosis	F (5,62) = 0.6, p = .82	F(2,65) = 1.1, p = .35	F(2,65) = 0.1, p = .94	F(2,65) = 0.6, p = .57	F(2,65) = 0.6, p = .56	F(2,65) = 0.6, p = .54
Disease activity	F (5,62) = 1.9, p = .02	F(4,63) = 1.0, p = .39	F(4,63) = 1.9, p = .13	F(4,63) = 1.5, p = .21	F(4,63) = 3.3, p = .02	F(4,63) = 1.7, p = .17
JRA treatment	F (5,62) = 2.2, p = .02	F(2,65) = 0.6, p = .56	F(2,65) = 3.1, p = .05	F(2,65) = 2.0, p = .14	F(2,65) = 2.4, p = .10	F(2,65) = 1.0, p = .36
Time since diagnosis	F (5,62) = 1.6, p = .05	F(4,63) = 1.9, p = .11	F(4,63) = 1.1, p = .35	F(4,63) = 0.8, p = .53	F(4,63) = 0.5, p = .75	F(4,63) = 4.6, p = .01
Pain		r = -.14, p = .27	r = -.22, p = .08	r = .21, p = .10	r = -.28, p = .03	r = -.07, p = .96
Functional limitations		r = -.07, p = .57	r = -.24, p = .06	r = .09, p = .43	r = -.02, p = .89	r = .03, p = .79

Note. Correlations are reported for pain and functional limitations,

Table 19

Intercorrelations Among Stress and Family Coping Variables

	Team effort	Status quo	Emotion processing	Cognitive flexibility	Seek meaning
JRA-specific - child	-.37**	-.36**	-.08	-.20	-.10
JRA-specific - parent	-.31*	-.46**	-.03	-.08	-.04
Global stress - child	-.27*	-.13	-.19	-.02	-.18
Global stress - parent	-.28*	-.41**	-.13	-.04	-.36**

Note. * $p < .05$ ** $p < .01$

Table 20

Hierarchical Multiple Regression Equations: Regressing Psychosocial Adjustment on Disease Characteristics, Stress and Coping

	Adherence medication			Adherence exercise			Parental depression		
Variables entered on each step	ΔR^2	F	Beta	ΔR^2	F	Beta	ΔR^2	F	Beta
Total R^2 for the equation	.54	3.73 **		.44	2.08 *		.51	3.19 **	
Step 1 – Disease characteristics	.20	2.37 *		.19	1.86		.06	0.54	
JRA diagnosis			.33 **						
Disease activity			-.33 *						
Time since diagnosis									
Functional limitations									
Step 2 – Family stress	.27	6.66 **		.12	1.87		.40	9.47 **	
JRA-specific stress (child report)			-.39 *						
Global stress (child report)									.27*
Global stress (parent report)									.33*
Step 3 – Family-level coping	.07	1.45		.14	1.94		.05	0.98	
Team effort			.33 *						

Table 20 continued.

Variables entered on each step	Internalizing behavior			Externalizing behavior			Quality of life		
	ΔR^2	F	Beta	ΔR^2	F	Beta	ΔR^2	F	Beta
Total R^2 for the equation	.44	2.41 **		.45	2.59**		.82	10.60**	
Step 1 – Disease characteristics [1]	.06	0.59		.06	0.61		.50	7.42 **	
JRA diagnosis									
Time since diagnosis									.26*
Functional limitations									-.64**
Step 2 – Family stress	.34	7.30 **		.15	2.42 +		.24	9.08 **	
JRA-specific stress (child report)			.62**						
Global stress (child report)						.36			-.43**
Global stress (parent report)									
Step 3 – Family-level coping	.04	0.92		.24	4.19 **		.08	3.20 *	
Team effort						-.53**			
Cognitive flexibility									-.33**

Note. [1] Although the set of 6 medical characteristics was entered on Step 1, the set of 4 family stress measures on Step 2, and the set of 5 family coping measures on Step 3, only the significant beta weights are shown in the table.
* $p < .05$ ** $p < .01$ + $p < .10$

Table 21

Hierarchical Multiple Regression Equations Testing the Moderational Model

	Adherence medication			Adherence exercise			Parental depression		
Variables entered on each step	ΔR^2	F	Beta	ΔR^2	F	Beta	ΔR^2	F	Beta
Total R^2 for the equation	.45	3.99**		.24	1.23		.36	2.76 **	
Step 1 – JRA-specific stress (child)	.13	8.98**	-.25*	.05	2.72		.21	16.83**	.45**
Step 2 – Set of family coping	.19	3.26**		.15	1.78		.11	1.95	
Team effort			.49**			.64*			
Cognitive flexibility									
Set 3 – Stress by coping interaction	.14	2.64*		.04	0.48		.04	0.67	
Stress x team effort			-.40**						
Stress x status quo			.41**						
Stress x emotion processing									
Stress x cognitive flexibility									
Stress x seeking meaning									

Table 21 continued

	Internalizing behavior			Externalizing behavior			Quality of life		
Variables entered on each step	ΔR^2	F	Beta	ΔR^2	F	Beta	ΔR^2	F	Beta
Total R^2 for the equation	.28	1.94*		.30	1.71*		.55	4.50**	
Step 1 – JRA-specific stress (child)	.26	23.13**	.38*	.03			.24	16.37**	-.29*
Step 2 – Set of family coping	.01	0.21		.21	3.27**		.12	1.67	
Team effort						-.48**			
Cognitive flexibility									
Set 3 – Stress by coping interaction	.01	0.07		.07	2.12*		.19	3.41**	
Stress x team effort									.35*
Stress x status quo									
Stress x emotion processing									
Stress x cognitive flexibility									
Stress x seeking meaning									.31*

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

Table 22

Comparison of Early and Late Adolescents on Demographics, Disease, Stress, Coping and Adjustment

	Early adolescents (n = 33)		Late adolescents (n= 35)		
	M	SD	M	SD	Age group differences
Demographics				**Multivariate**	**F (5,62) = 3.75, p = .01**
Gender	1.17	0.4	1.36	0.5	F (1,66) = 3.29, p = .07
Family size	2.43	0.7	2.27	0.8	F (1,66) = 0.70, p = .41
Marital status	0.74	0.4	0.88	0.3	F (1,66) = 2.03, p = .16
Family SES	37.63	12.2	31.18	12.3	F (1,66) = 4.70, p = .03
Ethnicity	0.14	0.4	0.36	0.5	F (1,66) = 4.58, p = .04
Disease				**Multivariate**	**F (6,58) = 1.22, p = .31**
JRA diagnosis	1.61	0.7	1.59	0.6	F (1,63) = 0.01, p = .94
JRA treatment	1.64	0.6	1.31	0.8	F (1,63) = 3.30, p = .07
Time since diagnosis	51.36	49.6	66.03	45.7	F (1,63) = 1.54, p = .22
Pain	20.94	24.3	13.75	18.0	F (1,63) = 1.83, p = .18
Functional limits	0.39	0.6	0.31	0.6	F (1,63) = 0.25, p = .62
Stress				**Multivariate**	**F (4,60) = 1.47, p = .22**
JRA stress - child	9.52	8.7	11.94	9.1	F (1,63) = 1.20, p = .28
JRA stress-parent	16.76	8.4	17.03	10.2	F (1,63) = 0.14, p = .91
Global – child	3.67	2.4	4.44	2.4	F (1,63) = 1.67, p = .20
Global – parent	6.18	2.7	5.44	3.0	F (1,63) = 1.11, p = .29

Table 22 continued

Comparison of Early and Late Adolescents on Demographics, Disease, Stress, Coping and Adjustment

	Early adolescents (n = 33)		Late adolescents (n= 35)		
	M	SD	M	SD	Age group differences
Family coping				**Multivariate**	**F (5,62) = 3.20, p = .01**
Team effort	2.68	0.9	2.10	1.1	F (1,66) = 5.18, p = .03
Status quo	1.72	1.1	2.02	1.2	F (1,66) = 1.13, p = .29
Emotion processing	1.37	1.0	1.31	0.9	F (1,66) = 0.61, p = .81
Cognitive flexibility	1.87	0.8	1.72	1.2	F (1,66) = 0.31, p = .58
Seek meaning	1.03	0.6	1.24	0.9	F (1,66) = 1.11, p = .29
Adjustment				**Multivariate**	**F (6,38) = 1.21, p = .33**
Adherence medication	4.02	0.5	3.93	0.7	F (1,43) = 0.22, p = .64
Adherence exercise	3.06	0.9	3.11	0.9	F (1,43) = 0.03, p = .87
Parental depression	5.52	4.3	5.75	6.2	F (1,43) = 0.02, p = .88
Internalizing	44.92	8.8	51.55	9.3	F (1,43) = 6.03, p = .02
Externalizing	42.36	7.8	48.10	11.2	F (1,43) = 4.12, p = .05
Quality of life	54.52	8.5	53.20	8.6	F (1,43) = 0.27, p = .61

Table 23

Multivariate F Statistics Showing Developmental Differences in Perceived Stress, Family Coping and Adjustment

	Multivariate Hotelling's Trace F statistic		
	Age	Time since diagnosis	Age x Time since diagnosis
Stress	F (4,56) = 1.01, p = .41	F (8,56) = 1.32, p = .24	F (8,56) = 1.01, p = .43
Family coping	F (5,58) = 2.62, p = .03*	F (10,58) = 1.79, p = .07	F (10,59) = 1.19, p = .30
Status quo	F (1,62) = 4.43, p = .04*		
Team effort		F (2,62) = 2.71, p = .08	
Adjustment	F (6,34) = 0.92, p = .47	F (12,66) = 0.47, p = .24	F (12,66) = 0.15, p = .95
Externalizing	F (1,39) = 4.19, p = .05*		

* $p < .05$

Figure 1.

Conceptual model of stress, family coping and adjustment in Juvenile Rheumatoid Arthritis (JRA).

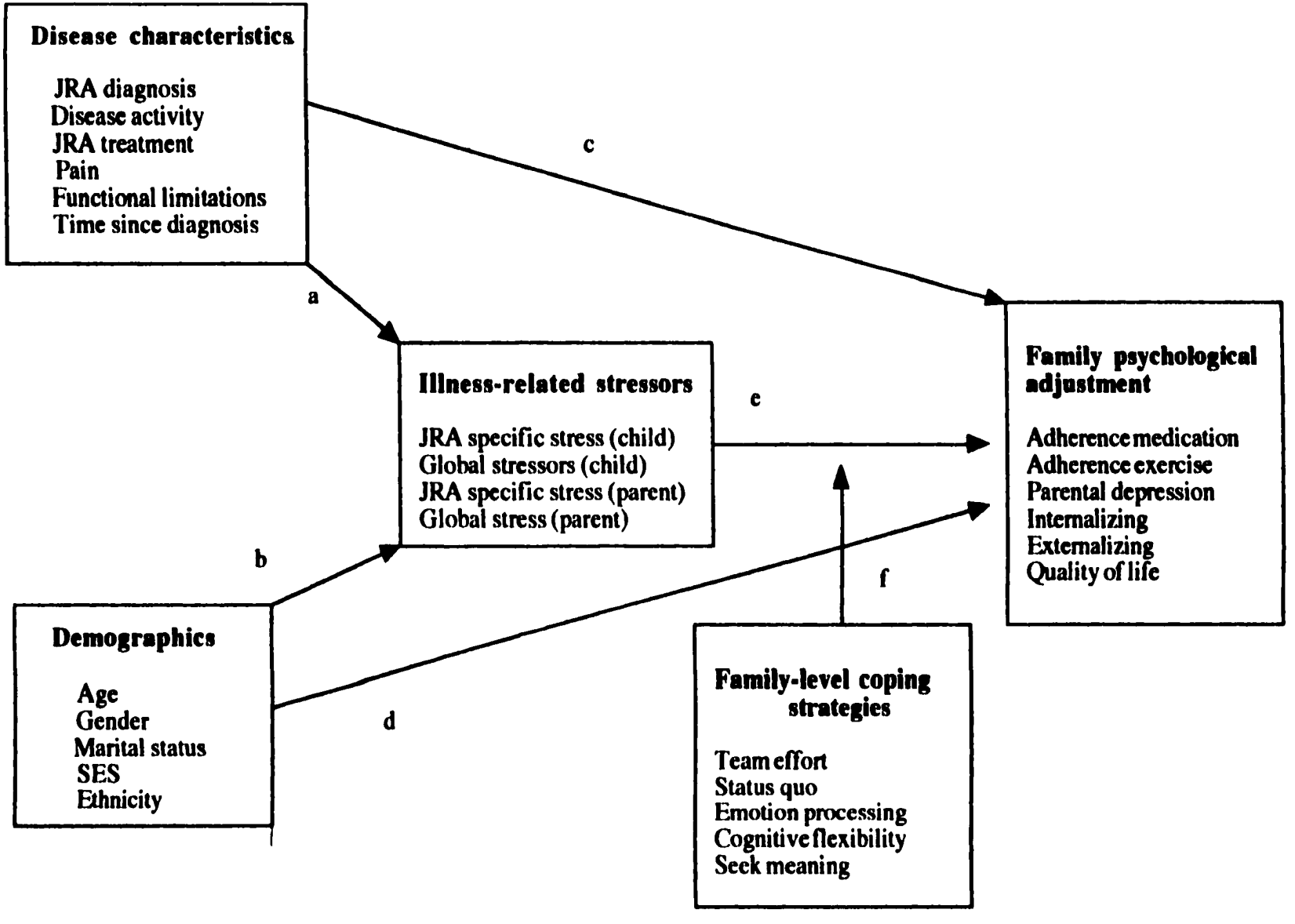


Figure 2a. Adherence to Medication

Stress x Team Effort

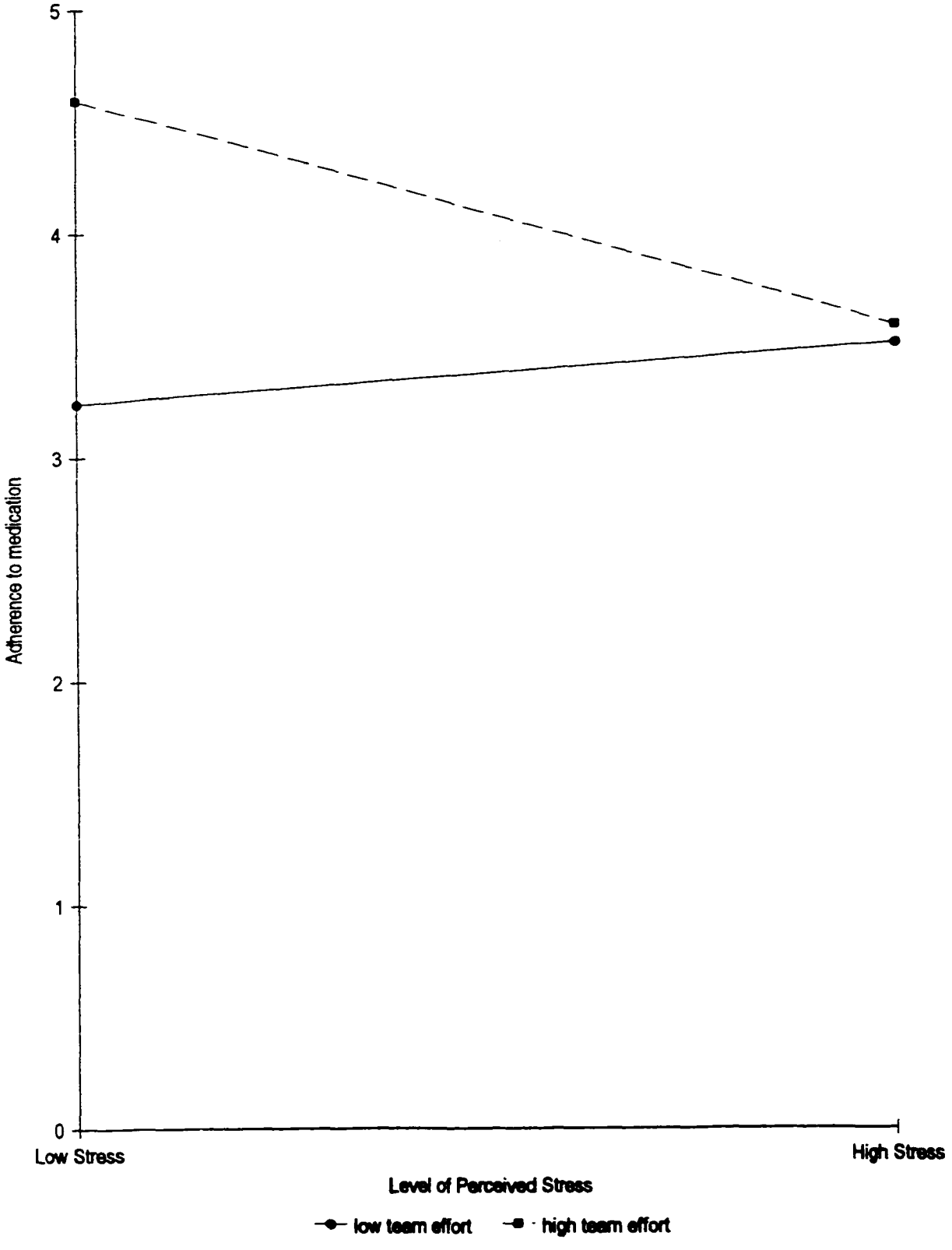


Figure 2b. Quality of Life

Stress x Team Effort

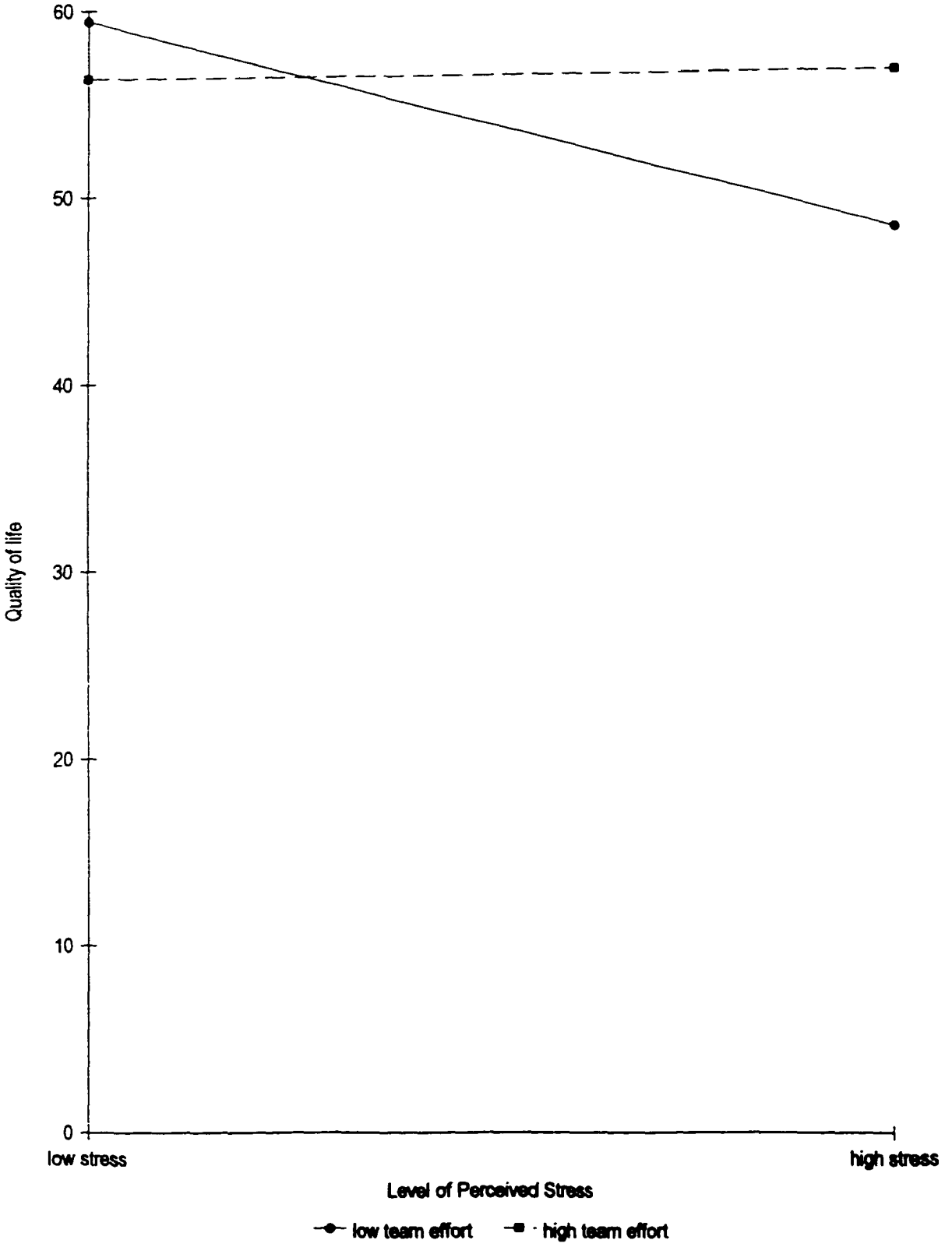


Figure 2c. Quality of Life

Stress x Seek Meaning

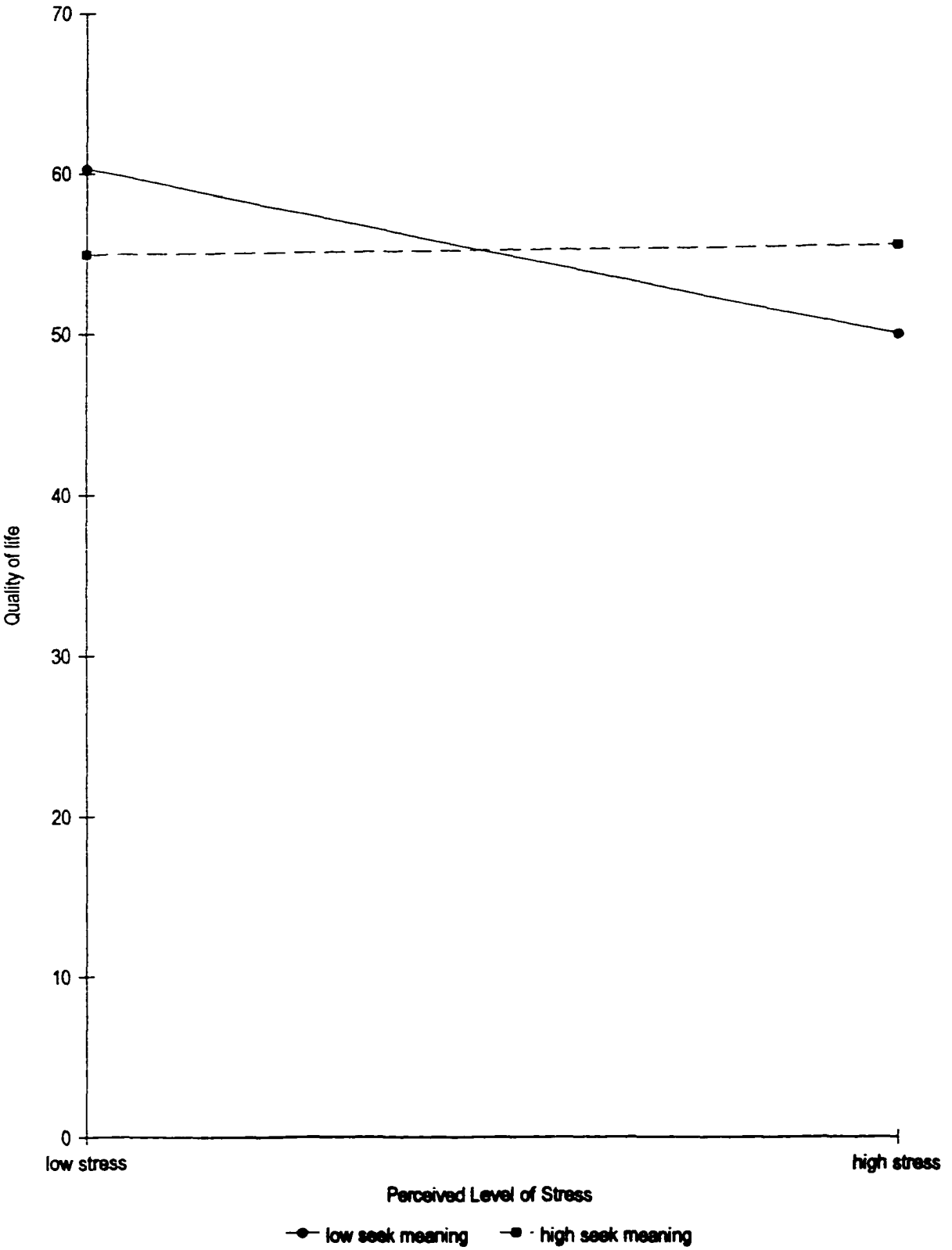
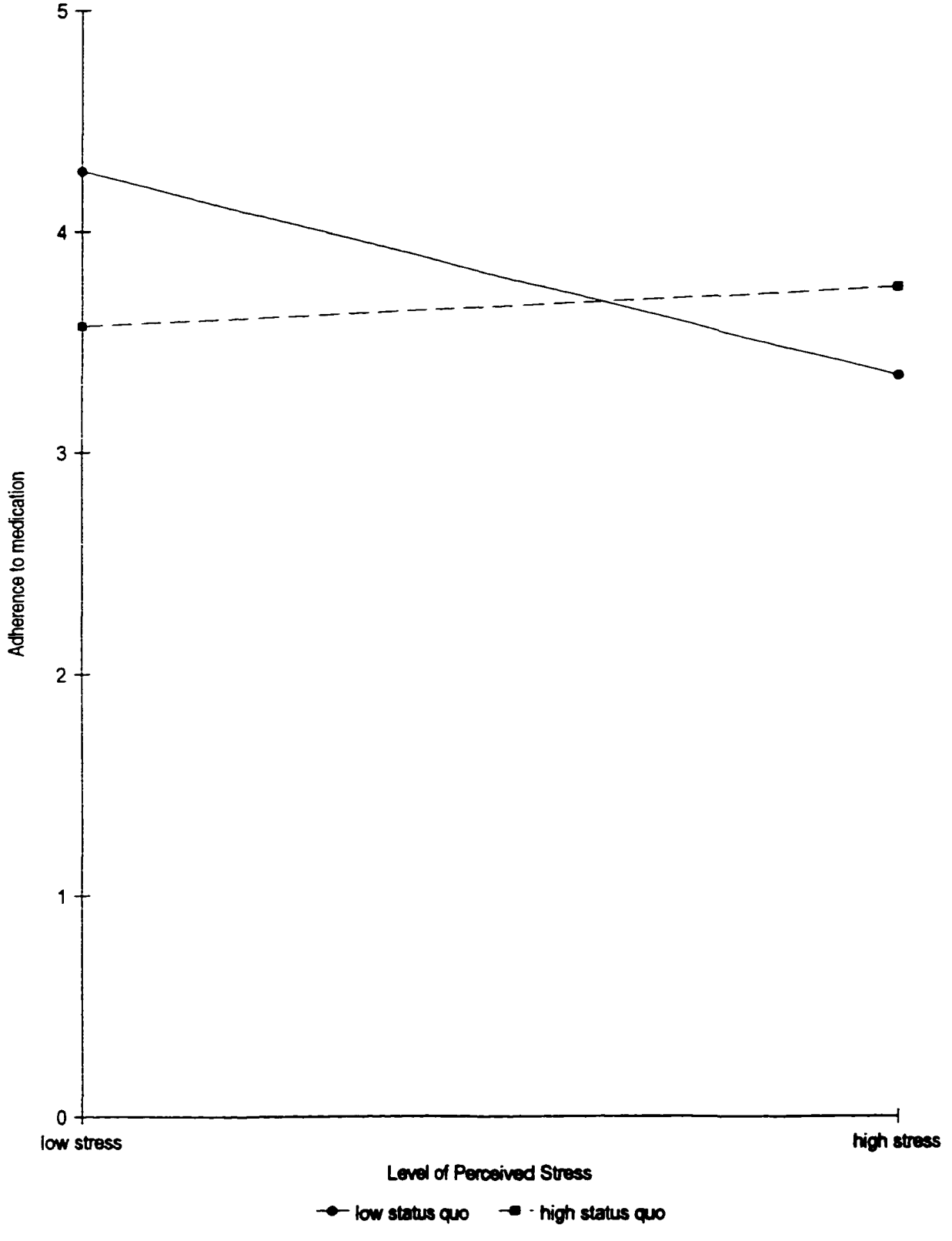


Figure 2d. Adherence to Medication

Stress x Status Quo



Appendix A

- Recruitment letter
- Human Subject's Committee approval
- Child assent
- Parent consent
- Outline of JRA family interview

SCHNEIDER CHILDREN'S HOSPITAL

Division of Rheumatology
New Hyde Park, NY 11042
Telephone 718 470-3530

Norman Ilowite, MD, Chief
Associate Professor of Pediatrics
Albert Einstein College of Medicine
Peter LoGalbo, MD

Arthritis Center
Norman Ilowite, MD
John Handelsman, MD
Angeles Badell, MD

Lyme Disease Center
Lorry G. Rubin, MD
Norman Ilowite, MD

Physical Medicine & Rehabilitation
Angeles Badell, MD, Chief

Clinical Nurse Specialist
Eileen Pagano, RN, MS

Nurse Clinician
Trudy Leicht, RN, BS

Dear parent.

This letter is to invite you and your family to participate in a research project that is being undertaken in cooperation with researchers from The Graduate School and University Center of the City University of New York. This study will focus on how families are affected by the experience of having a child with juvenile rheumatoid arthritis (JRA). The findings of this study are expected to aid family intervention efforts by health professionals.

Your participation would involve filling out a short questionnaire and participating in a family interview about your family's experience in dealing with a chronic illness. If possible, the researchers would like to include the viewpoints of parents and siblings as well as the child with JRA. In addition, the researchers would also request information from me about the type and severity of your child's illness. All information would remain strictly confidential. Your name would not appear on the questionnaire, nor on the transcription of the interview, and any details that might identify specific families would not be used in published research reports.

This study does not involve any medical tests or procedures. Furthermore, the decision to participate or not, will in no way affect the medical treatment or services your family receives.

Your family's participation would be greatly appreciated, but is entirely voluntary. If you choose to participate you can withdraw at any time without penalty, and you can omit any questions you choose not to answer. However, it is anticipated that your family will find the interview an interesting and enjoyable experience. Also, participants often feel good about contributing to the welfare of others with similar medical conditions through cooperating in research projects.

In the week or two prior to your child's next scheduled appointment, you will be contacted by phone by the principal investigator, Pamela Degotardi MA who will fully explain the project, and answer any questions you may have. If your family agrees to participate, Ms. Degotardi will arrange a time that is convenient for you to be interviewed. It is hoped that most interviews can be conducted either immediately before or after your child's clinic visit. The entire interview with all available members of your family should take approximately an hour. Again, you can change your mind at any time, and you should not feel obligated to participate in the interview. If you have any questions, I can be reached at (718) 470-3530, or you can directly contact Pamela Degotardi at (718) 268-6046.

Please consider having your family participate in this important study. It is anticipated that research of this type will help in the planning of family intervention and treatment of JRA.

Sincerely,

Dr. Norman Ilowite

THE
GRADUATE SCHOOL
AND
UNIVERSITY CENTER
THE CITY UNIVERSITY OF NEW YORK

OFFICE OF RESEARCH AND UNIVERSITY PROGRAMS
SPONSORED RESEARCH
33 WEST 42 STREET, NEW YORK, NY 10036-8099
212 642-2059 FAX 212 642-2546

July 2, 1997

Ms. Pamela Degotardi
97-12 72 Drive
Forest Hills, NY 11375

Re: Your Human Subjects Proposal, "Stress, Family Coping and Adaptation in Adolescents with Juvenile Rheumatoid Arthritis (JRA)

Dear Ms. Degotardi:

The Committee on the Protection of Human Subjects reviewed and approved your continuation proposal at its June 17 meeting. This approval is effective for one year, and your proposal must be reviewed annually should it extend beyond one year. Should any changes be made in the interim, the proposal must be resubmitted to the Committee for review.

The Office of Protection from Research Risks of the Department of Health and Human Services requires the consent forms to bear an approval and expiration date. Please refer to the enclosed forms which must be used in obtaining consent.

Sincerely,

Hilry Fisher
Director

encs.
c: Tracey Revenson

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GRADUATE SCHOOL
AND
UNIVERSITY CENTER
THE CITY UNIVERSITY OF NEW YORK

PH.D. PROGRAM IN PSYCHOLOGY:
HEALTH PSYCHOLOGY CONCENTRATION
33 WEST 42 STREET, NEW YORK, NY 10036-8099
FAX 212 642-2580

Approved: 6/17/97
Expires: 5/18/98

Child Assent Form for Participation in JRA Family Study

Pamela Degotardi MA; Tracey A. Revenson PhD; & Norman Ilowite MD

We would like to learn more about the positive and negatives family experiences of having a family member with juvenile rheumatoid arthritis (JRA). We would like to interview you and other family members about such things as your knowledge of JRA and it's treatment; how JRA has affected your schooling, sports activities, hobbies and friendships; and the effect of JRA on the entire family. Finally we would like to discuss the factors that have helped your family understand and adjust to JRA.

This study will not involve medical tests, and it will in no way influence the medical treatment and service that you currently receive.

I understand that if I agree to participate in this study that I can change my mind, or stop at any time. I don't have to answer any questions that I don't want to. I know that I can ask the researcher questions to clarify anything that I don't understand.

I understand that everything I discuss during the interview will be kept strictly confidential. I also understand that I am entitled to listen to the audiotape of my family interview if I wish, and I may request that my responses not be included in the study.

Name :_____

Signature : _____

Date : _____

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Parent Consent Form for Participation in JRA Family Study

Pamela Degotardi MA, Tracey A. Revenson PhD, & Norman Ilowite MD

1. Purpose of Study : This research aims to : (1) document how families are affected both positively and negatively by the experience of having a child with juvenile rheumatoid arthritis (JRA), and (2) examine how the life tasks and cognitive abilities of children influence treatment compliance and psychosocial functioning. Your participation involves :

a. Completing this consent form.

b. Completing a questionnaire concerning your family experiences.

c. Participating in a family interview that will cover topics such as your knowledge about JRA and treatment, how JRA has impacted on your child's social and academic functioning, the family experiences and concerns regarding JRA, and factors that have helped your family understand and adjust to JRA. This interview will be audio-taped so that the researcher can accurately code responses, and use this data in a group analysis.

d. Allowing, but not coercing, your children to be interviewed.

e. Allowing Dr. Ilowite to give the researchers information about the severity of your child's JRA.

2. Potential Risks : No risks are anticipated. This study will involve no medical tests and will have no effect on your child's medical treatment and services.

3. Potential Benefits : Participation will result in increased knowledge about how families experience and adapt to having a child with JRA. The interview may be thought-provoking and encourage shared family communication about JRA. Participants in this type of research often feel good about contributing to the welfare of others with similar medical conditions by cooperating in research.

I am fully aware that non-participation will not in any way affect the medical care that my family will continue to receive. I understand that I am free to withdraw my consent and to discontinue participation in this study at any time without prejudice to me. In addition, I need not answer questions that I feel do not apply to me, or that I chose to omit for any reason. I understand that I am entitled to make enquiries about any aspect of the study that I do not understand, and to

review the audiotape of my interview and request that it not be included in the study. I understand that I will be informed of any changes that might affect my willingness to continue participation in this study. If I have any questions about this study I can contact either Dr. Ilowite (718) 470-3530 or Pamela Degotardi (718) 268-6046. If I have any questions about my role as a research participant I can contact the Office of Sponsored Research at CUNY at (212) 642-2059.

I understand that my involvement in this study will be completely confidential; neither my name nor any identifying information will appear on the questionnaire, interview transcripts, or any other report; only group data will be presented. I understand that this consent form will be detached and stored apart from my responses on the interview to ensure confidentiality. I also understand that my child's physician will not have access to the completed questionnaire or interview transcripts.

I hereby agree to participate in this research study, and I give permission for my child/children _____ **to participate as well.**

Name : _____ Date : _____

Signature : _____

Please check you if you would like us to send you a brief report of our research findings after all the data has been collected and analyzed.

Yes, I would like a research report _____

Approved: 6/17/97
Expires: 5/18/98

JRA Family Study -- Interview outline

Pamela Degotardi MA, Tracey A. Revenson PhD, & Norman Ilowite MD

Section A -- knowledge of JRA etiology and treatment

* initial symptoms -- age -- family action
* diagnosis -- severity
* theories JRA ?
* read about arthritis -- AF

Section B -- treatment and compliance.

* how did check-up go --- any changes in medication/ JRA activity
* treatment regimen (medicine, exercise)
* compliance
* treatment responsibility
* tips for remembering medication
* family's relationship with health professionals -- any problems

Section C --- school/ work and peers

* school/ grade --- type of work/ college
* absenteeism -- writing, gym
* teachers -- special allowances or restrictions (eg. physical therapy, extra time to complete work).
* friends -- support, teasing, embarrassed, try to hide signs of JRA
* interfere with sports or activities

Section D -- family

* effect on family -- changes in routines or activities or relationships.
* over protectiveness
* siblings -- supportive, protective, jealous of extra attention
* future concerns -- disease progression, career possibilities

Section E -- adaption and adjustment

* What aspect of having JRA has caused the most problems for your family -- financial, clinic visits?
* What has helped your family most in coping with arthritis ? -- family support, doctors, faith, sense of humor
* What advice would you give other families who have a child who has just been diagnosed with JRA ?

Appendix B

- Medical form to be completed by pediatric rheumatologist
- Parent questionnaire (non-copyright material)
    - Background/demographic information
    - Treatment adherence
    - JRA-specific stressors
- Child questionnaire (non-copyright material)
    - Treatment adherence
    - JRA-specific stressors

Subject # _____

MEDICAL INFORMATION

SUBJECT NAME _____

DATE OF INTERVIEW _____

JRA DIAGNOSIS : pauciarticular polyarticular systemic onset

AGE AT DIAGNOSIS : _____

DISEASE ACTIVITY : remission > 2 mths quiescent
mild moderate
severe

FUNCTIONAL LEVEL : fully functional minimum assistance needed
moderate assistance fully incapacitated

TREATMENT : medication only physical therapy only
no treatment medication and physical therapy

MEDICATION :

DRUG MONITORING :

SPLINTS : _____ ORTHOTICS : _____

TREATMENT COMPLIANCE :

" This child takes medication as often as prescribed"
always usually sometimes seldom never

"This child does the physical therapy exercises as often as prescribed"
always usually sometimes seldom never

PHYSICIAN COMMENTS :

Subject # _____

Parent Questionnaire
JRA Family Study 1997

Pamela Degotardi MA, Tracey A. Revenson PhD, & Norman Ilowite MD

Funded in part by a grant from the New York Chapter of the Arthritis Foundation.

Subject # _____

Background Information for JRA Family Study

Family name : _____

Names and ages of children :

Father's occupation : _____

Highest education level of father:

Elementary school graduate High school graduate Some college
College graduate Professional / Graduate School

Mother's occupation : _____

Highest education level of mother :

Elementary school graduate High school graduate Some college
College graduate Professional / Graduate School

Marital status :

Married Separated Divorced Widowed Single

If widowed or divorced from child's father have you remarried : Yes No

Ethnicity :

White Black Hispanic Asian-American Other

Subject # _____

Medication and exercise regimens

1. Please check the statement that is most true of you and your child :

My child always remembers to take medication and do exercises on her/his own. _____

My child mostly remembers to take medication and do exercises on her/his own. _____

I sometimes have to remind my child to take medication and do exercises. _____

I always have to remind my child to take medication and do exercises. _____

I have to constantly nag my child to take medication and do exercises. _____

My child refuses to take medication or do exercises. _____

2. Please estimate how often your child forgot to take medication in the past week :

never once 2 or 3 times 4 or more times

3. Please estimate how often he/she missed doing physical exercises in the past week :

never once 2 or 3 times 4 or more times

4. Please circle the best response : " My child takes medication as often as prescribed"

always usually sometimes seldom never

5. What things DOESN'T your child like about taking medication :

6. Please circle the best response : " My child does physical therapy exercises as often as recommended."

always usually sometimes seldom never

7. What things DOESN'T your child like about doing the exercises :

Subject #

Parent JRA stressors for JRA Family Study

Pamela Degotardi MA, Tracey A. Revenson PhD, & Norman Ilowite MD

In past studies families listed the problems below as things that were stressful. Please indicate which of these items you personally find stressful and rate the amount of stress of each problem using the scale : ---

0 = not at all stressful
1 = a little stressful
2 = somewhat stressful
3 = very stressful
4 = extremely stressful

(e.g., 0 = not stressful -----> 4 = extremely stressful)

0 1 2 3 4 Fear or uncertainty about your child's future due to the arthritis.

0 1 2 3 4 Limitations in your child's physical ability, appearance or life style due to arthritis.

0 1 2 3 4 Pain or discomfort your child experiences due to arthritis.

0 1 2 3 4 Problems with family or friends related to arthritis.

0 1 2 3 4 Unpredictability of arthritis flares.

0 1 2 3 4 School difficulties related to arthritis.

0 1 2 3 4 Frequency of clinic visits.

0 1 2 3 4 Financial burden (eg. cost of medication, insurance etc).

0 1 2 3 4 The demands of the treatment regimen (eg. medication and physical therapy).

0 1 2 3 4 Disruption to family routines due to arthritis.

0 1 2 3 4 Restrictions of sports or activities due to arthritis.

Subject # _____

Youth Questionnaire
JRA Family Study 1997

Pamela Degotardi MA, Tracey A. Revenson PhD, & Norman Ilowite MD

Funded in part by a grant from the New York Chapter of the Arthritis Foundation.

Subject # _____

Medication and exercise regimens

1. Please check the statement that is most true of you :

I always remember to take my medication and do exercises on my own. _____
I mostly remember to take my medication and do exercises on my own. _____
Mom sometimes has to remind me to take my medication and do exercises. _____
Mom always has to remind me to take my medication and do exercises. _____
Mom has to constantly nag me to take my medication and do exercises. _____
I don't take my medication or do my exercises. _____

2. Please estimate how often you forgot to take medication in the past week :

never once 2 or 3 times 4 or more times

3. Please estimate how often you missed doing your physical therapy exercises in the past week :

never once 2 or 3 times 4 or more times

4. Please circle the best response : " I take my medication as often as prescribed"

always usually sometimes seldom never

5. What things DON'T you like about taking medication :

6. Please circle the best response : " I do my physical therapy exercises as often as recommended."

always usually sometimes seldom never

7. What things DON'T you like about doing the exercises :

Subject #

Child JRA stressors for JRA Family Study

Pamela Degotardi MA, Tracey A. Revenson PhD, & Norman Ilowite MD

In past studies families listed the problems below as things that were stressful. Please indicate which of these items you personally find stressful and rate the amount of stress of each problem using the scale : —

0 = not at all stressful
1 = a little stressful
2 = somewhat stressful
3 = very stressful
4 = extremely stressful

(e.g., 0 = not stressful -----> 4 = extremely stressful)

0 1 2 3 4 Fear or uncertainty about your future due to the arthritis.

0 1 2 3 4 Limitations in your physical ability, appearance or life style due to arthritis.

0 1 2 3 4 Pain or discomfort due to arthritis.

0 1 2 3 4 Problems with family or friends related to arthritis.

0 1 2 3 4 Unpredictability of arthritis flares.

0 1 2 3 4 School difficulties related to arthritis.

0 1 2 3 4 Frequency of clinic visits.

0 1 2 3 4 Financial burden (eg, cost of medication, insurance etc).

0 1 2 3 4 The demands of the treatment regimen (eg, medication and physical therapy).

0 1 2 3 4 Disruption to family routines due to arthritis.

0 1 2 3 4 Restrictions of sports or activities due to arthritis.

Appendix C

- Coding worksheet for FCCS (Family # 55) – examples of key phrases and exemplars of family coping strategies from the family interview. These worksheets were used to note contributions of family members, and to assist in assigning scores for each of the 22 family-level coping strategies as described in the FCCS coding manual developed by Stuart Hauser and colleagues (1993).

Family # 55 Date interview 4·25·97 Date coded 6·23·97 Coder PJD

COPING STRATEGY	MOTHER	FATHER	ADOLESCENT	EXAMPLES - COMMENTS
	Page # in transcript	Page #	Page #	
1. Perspective taking/cog flex				
2. Family consensual unit	4, 6 (4)	7 ~ 8 (4)		"We" and "us" eg "We're all in this together"
score for coping strategy →				
3. Close minded -- blaming				
4. Family as collection indivs				
5. Theory construction	5 (2)	5 (2)		"... typically a genetic disorder"
6. Avoidance --denial				
7. Positive outlook	3, 6 (3)	4, 9 (4)	6, 3 (4)	"I can still dance, but I do jazz rather than classical"
8. Negative outlook				
9. Recognition of facts	7 (2)	9 (2)		"time consuming coming to the doctor" "cost factor"
10. Self reliance -- competence	2, 3, 5, 6, 7, 8 ~ 9 (4)	1, 8 ~ 9 (4)	4 (4)	"We had to do physiotherapy, we told the doctor we'd do it ourselves... We knew we could do it"
11. Inability to manage stressor		5 (1)		"a lot of tension at meal times"
12. Coordinated family efforts	3, 6, 9 (4)	1, 2, 3, 7 ~ 8 (4)		"We really made a concerted effort to battle this head on"

COPING STRATEGY	MOTHER	FATHER	ADOLESCENT	EXAMPLES - COMMENTS
13. Seeking information JRA	5, 8-9 (4)	9 (4)		"We read everything - asked a thousand questions"
14. Support seeking	2, 7, 9 (3)	2, 9 (2)	6 (2)	"my friends know" "joined Arthritis Foundation"
15. Established ways		4 (2)	6, 9 (2)	"Tried to keep her in an active but normal lifestyle"
16. Alternative reward		7, 8 (3)	4, 8 (3)	"I think its made her a better person"
17. Direct expression				
18. Acknowledge feelings				
19. Diminishing awareness				
20. Impulsive outbursts				
21. Prayer / faith				
22. Normalizing	6 (3)	6 (3)	6, 9 (4)	"Be cool - try to do normal things... don't make a big deal out of it"

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