

# Comparing Parental Distress, Family Functioning, and the Role of Social Support for Caregivers With and Without a Child With Juvenile Rheumatoid Arthritis

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**Objective** To assess parental distress, family functioning, and social support among parents of children with a lifetime diagnosis of juvenile rheumatoid arthritis (JRA) and comparison families. **Methods** Parents of 64 children with JRA (64 mothers, 46 fathers) completed questionnaires and in-home interviews along with 64 matched comparison families. Average time since diagnosis for children with JRA was 70 months. **Results** Families of children with JRA generally reported levels of parental distress, family functioning, and social support similar to those for comparison families. More mothers of children with JRA exceeded the clinical cutoff on the SCL-90-R than comparison mothers. Although disease characteristics and social support did not distinguish subgroups of parents at greater risk for problems, family supportiveness and conflict were associated with caseness for mothers of children with JRA. **Conclusions** Families of children with JRA exhibited substantial resilience over the long term. Further multisite study of children recently diagnosed and with more severe forms of JRA is warranted to determine intervention needs, especially for mothers.

**Key words** parents; families; adjustment; juvenile rheumatoid arthritis; social support.

Affecting 30,000 to 50,000 children in the United States each year (Lawrence et al., 1998), juvenile rheumatoid arthritis (JRA) is the most common pediatric rheumatic disease and one of the most common chronic illnesses in childhood (Cassidy, 1997). JRA is a heterogeneous group of conditions with considerable variability in the severity and chronicity of symptoms. Although some children may have multiple remissions of their disease, JRA can cause long-term physical disability (Peterson, Mason, Nelson, O'Fallon, & Gabriel, 1997), and up to two thirds of children may have active disease in adulthood (Minden et al., 2000; Peterson et al., 1997; Zak & Pedersen, 2000). Children with JRA can experience acute and chronic pain in inflamed joints, decreased mobility due to contractures and joint deterioration, vision problems, and growth retardation. Systemic involvement can cause organ damage (e.g., heart or liver) and, on rare occasions, can result in death. Treatment is palliative, not curative. It typically in-

cludes restrictions on activities and frequent medications that can cause uncomfortable side effects.

From a systems perspective, the diagnosis of a childhood chronic illness such as JRA has the potential to disrupt parental and family functioning (Kazak, 1989). Parents must manage their own feelings of uncertainty and loss of control, as they face the added responsibilities and daily hassles associated with caring for a child with special needs (Quittner, Opiari, Regoli, Jacobsen, & Eigen, 1992; Quittner et al., 1998). It is not uncommon to see evidence of mild to moderate levels of problematic adjustment for parents of children with a chronic illness (Jessop, Reissman, & Stein, 1988; Silver, Bauman, & Ireys, 1995). Mothers of chronically ill children are particularly at risk, presumably because they assume the greatest responsibility for the day-to-day care and medical needs of their child (Noll et al., 1995; Silver et al., 1995).

Limited work has examined the specific impact of

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JRA on parental distress and family functioning, with conflicting results. A recent review concluded that families of children with JRA may experience multiple difficulties due to the social, emotional, and financial impact of the disease (Reisine, 1995). For example, Vandvik, Hoyeraal, and Fagertun (1989) found that over two thirds of parents of children with JRA reported moderate to severe family difficulties (e.g., parental health problems, family conflict, and lack of social support), and more than half reported recent stressful life events. However, this study did not have a control group. As with other chronic pediatric conditions, mothers of children with JRA are presumed to be at greater risk for psychological distress than fathers (Lustig, Ireys, Sills, & Walsh, 1996; Timko, Stovel, & Moos, 1992; Vandvik & Eckblad, 1991). Concern for the well-being of these mothers has prompted randomized clinical trials to decrease their isolation and distress (Ireys, Sills, Kolodner, & Walsh, 1996).

Other data show that JRA has no significant detrimental impact on parents and families (e.g., Daltroy et al., 1992; Myones, Williams, Billings, & Miller, 1988). For example, in several controlled studies, families of children with various rheumatic diseases were found to display levels of adjustment comparable to those of families of healthy children (Harris, Newcomb, & Gewanter, 1991; Myones et al., 1988). Although one study indicated that mothers of children with JRA experienced greater symptoms of depression and general distress than mothers of healthy children, average scores for these mothers fell within the normal range of functioning (Frank et al., 1998). Similarly, Timko et al. (1992) found that mothers of children with JRA were more distressed than fathers were, but both groups had levels of distress within the normal range.

Little is known about what might account for the variability in parental and family adjustment. Several studies have suggested that disease factors and social support may influence outcomes (e.g., Lustig et al., 1996; McCormick, Stemmler, & Athreya, 1986; Timko et al., 1992). With respect to disease factors, families of children with more serious and debilitating disease appear to be at higher risk for problematic adjustment and parental strain (Jessop et al., 1988; Silver et al., 1995). Specific to JRA, greater disease severity and functional impairment have been associated with greater psychological distress among mothers (Lustig et al., 1996). Greater functional impairment among children with JRA also has been associated with increased family stress (McCormick et al., 1986), increased maternal psychological symptoms (Lustig et al., 1996), and poorer concurrent functioning among both mothers and fathers (Timko et al., 1992). In contrast, maternal distress has not been associated with disease severity in one

study (Vandvik & Eckblad, 1990), and higher levels of maternal distress have been found when children exhibited mild disease activity as compared to none or moderate/severe disease activity (Daltroy et al., 1992). Daltroy et al. speculated that mild disease may create difficulties, because it is less visible and results in less support for children and families.

With respect to social support, previous work has identified two potential mechanisms for its role in influencing distress (Cohen & Willis, 1985). First, social support may have an overall beneficial effect for any individual, regardless of the number of stressful events to which he or she is exposed (main effect). Unfortunately, a lack of social support has been reported by parents of children with JRA (Ireys et al., 1996; Vandvik et al., 1989), perhaps because heightened isolation results from their child's restricted activities and from the additional time devoted to child care. This lack of social support and increased isolation may contribute to greater parental distress (Jessop et al., 1988; Silver et al., 1995). A greater availability of social resources has been associated with lower levels of depression for parents of children with JRA (Timko et al., 1992), as well as other chronic conditions (Kronenberger & Thompson, 1992).

Second, social support may play a more predominant role when individuals are exposed to particularly stressful circumstances or numbers of stressful events. This stress by support interaction would result in a stronger association between support and distress for individuals experiencing higher levels of stress. Parents of children with JRA may worry about the health of their child (Gerhardt et al., *in press*) and experience numerous stressful life events or daily hassles (Vandvik et al., 1989, 1991). These stressors may include administering daily medications, clinic visits, and hospitalizations that would not be expected among parents of healthy children. Thus, social support and distress may be more strongly associated for parents of children with JRA than for controls.

The literature on psychosocial morbidity in parents and families of children with JRA presents a number of methodological shortcomings, including the use of unstandardized measures, inappropriate comparison groups, poor recruitment rates, ascertainment bias, and a lack of information from fathers. The purpose of this study was to evaluate parental distress, family functioning, and social support from the perspective of caregivers of children with JRA and comparison caregivers without a chronically ill child. The impact of disease factors also was examined. Based on prior research and clinical observations of the challenges associated with having a child with JRA, the following hypotheses were developed: (a) caregivers of children with JRA will report greater parental distress,

greater family difficulties, and fewer social resources than comparison caregivers, with parental distress being more pronounced for mothers of children with JRA than fathers; (b) greater disease severity will be associated with greater parental distress and family difficulties; and (c) parents who report more social support will report less distress (main effect), and this effect will be stronger for parents of children with JRA (interaction).

## Method

### *Participants and Procedures*

Following approval by the institutional review board, rosters of children with a lifetime diagnosis of JRA who were receiving treatment at a large children's hospital were used to identify potential participants. The center is the only facility with board-certified pediatric rheumatologists within a 50-mile catchment area, and a hospital tax levy ensures treatment for disadvantaged youths. The four inclusion criteria for children with JRA were (a) a diagnosis of JRA according to American College of Rheumatology classification standards (Cassidy et al., 1986), (b) age 8–14 years old, (c) no full-time special education, and (d) residence within 50 miles of the medical center. Of the 78 eligible families, 64 participated (4 were not located, 3 refused, and 7 schools declined participation in the initial phase of the study; see Noll et al., 2000).

Disease severity for the previous 6 months was rated by the child's pediatric rheumatologist to overlap with the timing of data collection. Severity was classified as mild or moderate/severe based on disease onset and course, the presence of iritis/uveitis, radiographic evaluation, pharmacotherapy, number of active joints, and Steinbrocker Functional Class rated by the physician on a scale of 1 to 4 (e.g., completely able to largely incapacitated; Hochberg et al., 1992). Any patient receiving methotrexate was considered at least moderate/severe. Radiologic evidence of moderate/severe disease was defined by the presence of joint space narrowing or subchondral bone erosion. Thirty-four (53%) had mild disease, and 30 (47%) had moderate/severe disease. In addition to disease severity, the rheumatologist rated each child's disease status as active ( $n = 32$ , 50%) or in full/partial remission ( $n = 32$ , 50%). Active disease included the presence of synovitis as defined by swelling or painful loss of motion. Partial remission was defined as the lack of objective synovitis but continued use of medication. Disease course was classified as pauciarticular ( $n = 31$ , 48%), polyarticular ( $n = 26$ , 41%), or systemic ( $n = 7$ , 11%). Mean time since diagnosis was 69.77 months ( $SD = 35.89$ , range = 10–149).

The initial phase of the study involved an assessment of the social functioning of children with JRA from the

perspective of classmates (Noll et al., 2000). Subsequently, matched comparison families (COMP) were recruited from lists of classmates who were the same gender and race as the target child and closest in age. The family of the classmate whose birthday was closest to the child with JRA was contacted first and invited to participate in a home assessment. If they did not wish to participate, the parents of the child with the next closest birthday were called, and so on, until a comparison family was recruited. Comparison families were screened to ensure the absence of a chronic illness in the family. Of these families, 59 (92%) were first choices, and the remaining families were second or third choices.

Data were collected in the home by two research assistants during a time that was convenient for the family and when the child was not hospitalized (i.e., no acute medical crisis). After providing informed consent/assent, parents and children independently completed several questionnaires. Families received \$100 for their participation. Data were collected from 64 mothers in each group (JRA/COMP), 46 fathers of children with JRA (15 families were single mother, 3 fathers declined), and 40 COMP fathers (22 families were single mother, 2 fathers declined). All measures were completed by parents only.

### *Measures*

**Demographic Questionnaire.** This instrument assesses basic background characteristics of the respondent, including marital status, religion, education, occupation, income, and number and age of offspring. Family socioeconomic status (SES) was computed using the Revised Duncan (TSEI; Nakao & Treas, 1992). This occupation-based measure is a contemporary indicator of SES that is sensitive to changes in occupational attainment (Hauser, 1994).

**Symptom Checklist 90-Revised (SCL-90-R; Derogatis, 1983).** This 90-item self-report inventory measures current psychological symptoms. It yields nine dimensions of psychological distress and three global indices of functioning (i.e., Global Severity Index [GSI], Positive Symptom Distress, and Positive Symptom Total). The GSI, considered the best single summary measure, combines information on the number and intensity of the symptoms reported. Positive Symptom Distress is an indicator of the intensity of symptoms experienced, and Positive Symptom Total represents the number of symptoms endorsed. This instrument was selected because of its sensitivity to general distress (Moore, Gilliss, & Martinson, 1988), especially internalizing difficulties. The SCL-90-R demonstrates adequate internal scale consistency (.77–.90) and reasonably good test retest reliability (1 week; .78–.90) (Derogatis, 1983). Previous work has supported

the concurrent validity of the measure (Brophy, Norvell, & Kiluk, 1988) and the sensitivity of the measure to difficulties experienced by parents of children with cancer (Noll et al., 1995). Our primary analysis of this instrument uses the GSI, but scores and exploratory analyses are reported for all scales to facilitate comparisons within the scientific literature.

**Family Environment Scale (FES; Moos & Moos, 1994).** The FES is a 90-item questionnaire designed to assess the family's social climate. Initially, the FES demonstrated adequate reliability for 10 summary scales, but later work indicated lower reliabilities (Loveland-Cherry, Youngblut, & Kline Leidy, 1989; Roosa & Beals, 1990). For this reason, four higher-order factor scores (i.e., Family Relationship Index [FRI], Supportive, Conflicted, and Controlling) were used in this study (Kronenberger & Thompson, 1990; Moos & Moos, 1994). Although the FRI has some overlap with the Conflicted scale, it was included for reference due to its use in previous studies. Internal consistencies for the 10 subscales are adequate (.61–.78) and test-retest reliabilities are also reasonably good across a 2-month period (.68–.86; Moos & Moos, 1994). Considerable evidence is presented in the instrument manual (Moos & Moos, 1994) supporting the concurrent validity of this measure. The FES has been used previously in studies of families of children with JRA (Harris et al., 1991; Myones et al., 1988).

**Norbeck Social Support Interview (NSSI; Norbeck, Lindsey, & Carrieri, 1981).** In an interview format, the respondent is asked to generate a list of significant others in his or her life (network size) and then to answer a series of six structured questions that allow for measurement of the parent's satisfaction with each person nominated. Previous work has demonstrated good predictive validity, test-retest reliability ( $r = .85-.92$ ), and internal consistency ( $\alpha > .85$ ) (Norbeck & Anderson, 1989; Norbeck et al., 1981; Norbeck, Lindsey, & Carrieri, 1983). Two scores reflecting network size and perceived functional support are derived from this measure. The NSSI has been used previously with parents of chronically ill children (Florian & Krulik, 1991; Noll et al., 1994).

### Analysis Plan

Two-tailed, independent  $t$  tests<sup>1</sup> and chi-square analyses ( $\alpha = .05$ ) were used as appropriate to compare JRA and COMP families on background variables. Two-tailed, independent  $t$  tests ( $\alpha = .05$ ) also were used to compare both types of families on parental distress (GSI), family functioning (FRI, Supportive, Conflicted, and Controlling), and social support (Network Size and Perceived Functional Support). Two-tailed, independent  $t$  tests were used

to examine the association between multiple disease factors, including severity (mild or moderate/severe), status (active or full/partial remission), type (pauciarticular or polyarticular/systemic), and measures of parental distress, family functioning, and social support. Pearson correlations were used to examine the relationship between disease duration and parental distress and family functioning.

Hierarchical multiple regressions were used to investigate the nature of the association between social support and parental distress (GSI). As suggested by Aiken and West (1991), all predictor variables were centered with respect to the mean score for each variable prior to entry into the regression equation. A main effect for group status (JRA/COMP) was first considered followed by entry of one of the two measures of support (i.e., to examine the main effect of social support). The interaction of group and support was entered last to test our final hypothesis.

Holm's corrections for multiple procedures was used to reduce Type 1 error in selected families of analyses (for details, see Holland & Copenhaver, 1988, or Holm, 1979). This procedure limits Type 1 error probability to at most  $\alpha$ , and it allows for increased power compared to traditional Bonferroni-type corrections. For this reason, corrections were completed for exploratory analyses using the SCL-90-R subscales that were not hypothesized effects. All subscale scores were treated as a family of variables separately for mothers and for fathers to facilitate corrections. Holm's correction was also applied to the family of tests involving each disease variable (i.e., severity, activity, type, duration). This was done due to the large number of tests completed and our lack of a priori hypotheses for disease activity, type, and duration.

Using GPOWER for power calculations (Faul & Erdfelder, 1992), we found that the sample of 128 mothers produced ample power (.89–.96) to detect medium effects for  $t$  tests ( $d = .5$ ) and multiple regressions ( $f^2 = .15$ ). For fathers, the sample of 86 allowed enough power (.74–.84) to detect medium effects for  $t$  tests ( $d = .5$ ) and multiple regressions ( $f^2 = .15$ ). These effect sizes are consistent with those reported in the literature.<sup>2</sup>

## Results

### Background Characteristics

Independent  $t$  tests indicated that JRA and COMP families did not differ significantly on a variety of background characteristics, including parent age, SES, parental education, number of children in the home, and child age (Table 1). The modal SES for both groups reflected occupations in clerical or sales positions (e.g., secretaries, sales clerks) or skilled blue collar jobs (e.g., craftsmen, ma-

**Table I.** Comparisons of Demographic Characteristics for Families With and Without a Child with JRA

Characteristic	JRA ( <i>n</i> = 64)	COMP ( <i>n</i> = 64)	<i>t</i> (126) <sup>a</sup>
Father age (years)	40.45 ± 5.81	41.33 ± 7.12	-.67
Mother age (years)	38.51 ± 6.70	37.66 ± 5.28	.80
SES <sup>b</sup>	49.89 ± 22.04	48.84 ± 20.67	.24
Father education	14.53 ± 2.63	14.61 ± 2.49	-.15
Mother education	13.73 ± 2.22	13.77 ± 2.42	-.08
No. of children living at home	2.69 ± 1.34	2.34 ± 1.03	1.63
Age of child at assessment (years)	11.07 ± 1.58	11.67 ± 1.59	-.18

Plus-minus values are means ± standard deviations.

<sup>a</sup>Degrees of freedom range from 93 to 126 due to missing information from nonparticipating fathers. This information was obtained from mothers whenever possible. All values are nonsignificant.

<sup>b</sup>Based on Duncan TSEI2. Higher scores represent greater occupational attainment.

**Table II.** Comparisons of SCL-90-R *T* Scores for Mothers With and Without a Child With JRA

Scales	JRA ( <i>n</i> = 64)	COMP ( <i>n</i> = 64)	<i>t</i> (126)
Global Severity Index	55.67 ± 10.57	53.27 ± 9.18	1.73
Positive Symptom Distress	54.71 ± 9.47	50.63 ± 7.99	2.43*
Positive Symptom Total	55.30 ± 9.58	53.98 ± 9.18	.84
Somatization	53.35 ± 10.98	51.67 ± 9.11	1.59
Obsessive-Compulsive	57.51 ± 9.64	55.73 ± 9.30	1.25
Interpersonal-Sensitivity	56.21 ± 9.78	54.97 ± 9.21	.99
Depression	55.54 ± 9.75	53.69 ± 8.87	1.52
Anxiety	52.00 ± 10.29	50.23 ± 8.30	1.71
Hostility	54.24 ± 9.84	51.08 ± 9.26	1.78
Phobia	49.60 ± 9.49	47.45 ± 6.76	1.81
Paranoid Ideation	53.81 ± 9.75	51.88 ± 9.76	.94
Psychoticism	54.05 ± 11.11	54.14 ± 9.21	-.98

Plus-minus values are means ± standard deviations.

\**p* < .05, two-tailed test, but nonsignificant following Holm's correction.

chinitists). Marital status did not differ between mothers of children with JRA (77% married) versus comparison mothers (66% married),  $\chi^2(4, n = 128) = 4.15, ns$ .

### Parental Distress

No significant differences were found on the GSI scores within either group of mothers or fathers. Additional independent *t* tests for the SCL-90-R subscales indicated that mothers of children with JRA reported significantly higher scores for Positive Symptom Distress than comparison mothers (Table II). When Holm's correction was applied, this was no longer significant. Mothers did not differ on the remaining SCL-90-R subscales. Fathers of children with JRA did not differ significantly from comparison fathers on the SCL-90-R subscales (Table III). In addition, mothers and fathers of children with JRA did not differ significantly from one another on SCL-90-R scores.

Further analyses were completed to compare the number of parents in each group who exceeded the clinical cutoff (i.e., a *t* score > 63 for GSI scores or for at least two subscales). Significantly more GSI mothers of children with

JRA (*n* = 29, 46%) were in the clinical range than comparison mothers (*n* = 18, 28%),  $\chi^2(1, n = 127) = 4.37, p < .05$ . For fathers of children with JRA, 22 (48%) were in the clinical range versus 13 (33%) comparison fathers,  $\chi^2(1, n = 85) = 2.08, ns$ . Disease factors, background characteristics, and social support did not significantly distinguish subgroups based on clinical status. However, clinical status for mothers of children with JRA was significantly associated with lower Supportive ( $M_{nonclin} = 33.93, SD = 5.58, M_{clin} = 27.82, SD = 7.79$ ),  $t(50) = 3.53, p = .001$ , and higher Conflicted ( $M_{nonclin} = -10.55, SD = 4.16, M_{clin} = -6.62, SD = 4.93$ ),  $t(61) = -3.44, p = .001$ , scores on the FES. FES scores were not associated with clinical status for COMP.

### Family Functioning

Four independent *t* tests were calculated for the higher-order factors on the FES to compare family functioning among those with and without a child with JRA (Tables IV and V). There were no significant differences between the two groups based on mother or father report.

**Table III.** Comparisons of SCL-90-R *T* Scores for Fathers With and Without a Child With JRA

Scales	JRA ( <i>n</i> = 46)	COMP ( <i>n</i> = 40) <sup>a</sup>	<i>t</i> (84) <sup>b</sup>
Global Severity Index	57.39 ± 11.09	55.70 ± 10.62	.89
Positive Symptom Distress	54.91 ± 8.02	52.10 ± 9.06	1.21
Positive Symptom Total	56.61 ± 10.02	54.80 ± 10.02	.93
Somatization	55.74 ± 10.70	53.23 ± 11.12	1.07
Obsessive-Compulsive	59.39 ± 10.27	56.68 ± 8.27	1.65
Interpersonal-Sensitivity	55.83 ± 11.32	56.37 ± 9.15	.32
Depression	57.72 ± 10.25	55.90 ± 11.15	.84
Anxiety	53.76 ± 11.32	51.30 ± 9.80	1.17
Hostility	53.98 ± 9.30	54.40 ± 9.64	-.12
Phobia	52.02 ± 8.13	50.50 ± 6.85	.88
Paranoid Ideation	54.91 ± 10.92	53.72 ± 11.08	.41
Psychoticism	55.24 ± 10.59	53.30 ± 9.83	.90

Plus-minus values are means ± standard deviations.

<sup>a</sup>The number of fathers (*n* = 46 targets, 40 controls) is lower due to single-parent households and nonparticipation.

<sup>b</sup>All values were nonsignificant.

**Table IV.** Comparisons of FES and NSSI Scores for Mothers With and Without a Child With JRA

Scales	JRA ( <i>n</i> = 64)	COMP ( <i>n</i> = 64)	<i>t</i> (125) <sup>a</sup>
FES			
Family Relationship Index	9.90 ± 4.80	9.91 ± 4.46	-.01
Supportive	31.20 ± 7.28	31.13 ± 6.54	.06
Conflicted	-8.86 ± 4.95	-9.32 ± 4.45	.55
Controlling	10.78 ± 4.17	10.96 ± 4.16	-.24
NSSI			
Network Size	7.48 ± 3.81	7.56 ± 4.19	-.10
Perceived Functional Support	190.27 ± 95.30	187.95 ± 108.04	.13

Plus-minus values are means ± standard deviations.

<sup>a</sup>All values were nonsignificant.

**Table V.** Comparisons of FES and NSSI Scores for Fathers With and Without a Child With JRA

Scales	JRA ( <i>n</i> = 46)	COMP ( <i>n</i> = 40) <sup>a</sup>	<i>t</i> (84) <sup>b</sup>
FES			
Family Relationship Index	9.15 ± 4.09	9.04 ± 5.30	.11
Supportive	30.61 ± 6.05	29.94 ± 7.94	.44
Conflicted	-8.18 ± 4.52	-8.78 ± 6.43	.50
Controlling	10.51 ± 4.24	10.56 ± 3.85	-.05
NSSI			
Network Size	6.42 ± 4.10	7.30 ± 5.14	-.88
Perceived Functional Support	153.87 ± 89.13	178.63 ± 124.03	-1.07

Plus-minus values are means ± standard deviations.

<sup>a</sup>The number of fathers (*n* = 46 targets, 40 controls) is lower due to single-parent households and nonparticipation.

<sup>b</sup>All values were nonsignificant.

### Social Support

Independent *t* tests compared the size of social support networks and perceived functional support for JRA and COMP families. There were no significant differences between the two groups based on mother or father report (Tables IV and V). Hierarchical multiple regressions showed no significant main effects of group status and social support on parental distress and family function-

ing. In addition, no significant interactions between group status and social support were found in predicting parental distress and family functioning

### Disease Factors

Pearson correlations indicated that time since diagnosis was not associated significantly with parental distress, family functioning, or social support. Other disease factors

were previously collapsed into dichotomous variables due to the small numbers of children with more severe forms of JRA or inactive disease. Independent *t* tests indicated that parental distress, family functioning, and social support did not differ as a result of mild versus moderate/severe disease or active versus full/partial remission. Having a child with polyarticular or systemic disease was associated significantly with mother and father report of higher GSI scores and with mother report of higher Positive Symptom Total compared to having a child with pauciarticular disease. When Holm's correction was applied, these results were no longer significant.

## Discussion

Previous work has indicated that families of children with JRA may be at risk for psychosocial difficulties (Reisine, 1995), but data have been limited and subject to numerous methodological problems. Although a few controlled studies have painted a brighter picture (Harris et al., 1991; Myones et al., 1988), these have been influenced by small samples and limited power. Poor recruitment and the use of unstandardized measures have been frequent problems. Also, there is a lack of information regarding the adjustment of fathers. The culmination of these methodological shortcomings has resulted in uncertainty regarding the degree to which these families are at risk for psychosocial difficulties and in need of intervention.

Seeking to ameliorate these problems by using a controlled design, this study assessed the impact on families of having a child with a lifetime diagnosis of JRA. Families of children with JRA were found to exhibit levels of adjustment quite similar to those for comparison parents, with the exception that a greater proportion of mothers of children with JRA met criteria for "caseness" on the SCL-90-R. Disease factors, background characteristics, and social support were not associated with clinical status. However, for mothers of children with JRA, clinical status was associated with Supportive and Conflicted scores on the FES. Contrary to predictions, mothers of children with JRA were not at greater risk for difficulties compared to fathers of children with JRA. Similar levels of social support were reported by parents of children with JRA and COMP parents. No main effects for social support or interactions with group status were identified. Disease factors such as severity, activity, type, and time since diagnosis were not associated significantly with psychosocial variables.

Studies that have found significant problems for families of children with JRA have tended to be those with the weakest designs and no controls. Often family dis-

ruption has been assessed using unstandardized measures that have included a variety of problems, such as parental health concerns and a lack of social support (e.g., Vandvik et al., 1989). Our finding that families of children with JRA exhibited functioning similar to that of comparison families is consistent with other controlled studies using the FES (Harris et al., 1991; Myones et al., 1988). However, these studies used small samples. Our sample yielded enough power to detect medium effects, but a multisite investigation with a sample of 500–600 would be necessary to detect small effects and further reduce Type II error.

Although mothers of children with JRA have been presumed to be at greater risk for difficulties than fathers, both groups often have fallen within the normal range of functioning (e.g., Timko et al., 1992). Of studies using adequate controls, only one found significant differences between mothers of children with JRA and comparison mothers (Frank et al., 1998). Specifically, mothers of children with JRA reported greater symptoms of depression and general distress than controls, but again, these scores fell well within the normal range of functioning. Furthermore, the average SCL-90-R depression score ( $M = 58.08$ ,  $SD = 10.11$ ,  $n = 98$ ) was nearly identical to the sample of mothers in this study ( $M = 57.72$ ,  $SD = 10.25$ ). Although we found that mothers of children with JRA were more likely than comparison mothers to meet criteria for caseness, we did not find significant differences on any other indices of distress.

The finding regarding caseness on the SCL-90-R was perplexing, because mothers did not differ significantly on the individual subscales. In addition, disease, background, and social support variables failed to demonstrate significant associations with clinical status. Intuitively, it fits that mothers of children with JRA who also were in the clinical range may perceive less family support and more conflict on the FES, but the same was not true for COMP mothers. Furthermore, information about the causal or temporal associations between these variables is unknown, limiting our ability to predict which subgroups of mothers will be at risk for difficulties. Future research can potentially replicate these findings and provide longitudinal data to clarify this issue.

Researchers have emphasized the importance of evaluating the well-being of fathers and the unique contribution they offer to understanding family functioning (Phares & Compas, 1992). Unfortunately, psychological research has been notorious for the exclusion of fathers and for their misuse as comparisons for mothers. Contrary to our expectations, fathers of children with JRA did not report greater difficulties for themselves or their families than fathers of healthy children. In the one study that has re-

ported data from fathers, scores fell within the normal range of functioning (Timko et al., 1992). Our findings further support the notion that having a child with JRA does not significantly increase risk for paternal distress.

Had we chosen to make comparisons to normative data in lieu of a control group, we may have concluded that JRA was associated with excessive parental distress. Some *t* scores for fathers on the SCL-90-R were nearly one standard deviation above an expected score of 50, but comparison fathers reported equally elevated scores. It is striking that approximately half of the JRA sample and a third of the COMP sample met criteria for caseness. The rate of caseness in our COMP sample is over three times that expected based on standardized scoring and available normative information (i.e., *t* score > 63, 90th percentile). One might argue that our comparison sample may have been unusual, but our matching procedure ensured that JRA and COMP families were nearly identical on multiple demographic and social risk factors. Comparisons to normative data have been criticized because they are dependent on the quality of the normative sample and can be susceptible to regional and cohort effects in spite of considerable effort to obtain a contemporary and representative normative sample (Achenbach & Howell, 1993; Sandberg, Meyer-Bahlburg, & Yager, 1991). Recent national events highlight the caution needed when using norms for measures of distress.

Our previous work using the SCL-90-R with parents of children with cancer (Noll et al., 1995) or sickle cell diseases (Noll et al., 1994) has shown a similar pattern of elevated scores for both parents of children with a chronic illness and controls. Despite previous efforts to obtain more information (sampling procedures, demographics, recruitment rates, etc.) about the “normative” sample from the publisher of the SCL-90-R (Noll et al., 1994), we have not been successful. The SCL-90-R manual was first published in 1977, and 85% of the nonpatient normal sample was single, despite an average age of 46 years. Thus, we strongly suggest exercising caution when using these norms. These findings underscore the importance of including demographically well-matched controls in future research and displaying thoughtfulness with the use of normative data. Alternative approaches to identifying controls for research in pediatric psychology might include conducting neighborhood searches, random digit dialing, or using a “snowball” technique (see Noll, Ris, Bukowski, Davies, & Koontz, 1992).

Contrary to expectations, parent and family functioning did not vary as a result of disease factors despite the use of a range of objective physician ratings for disease severity, activity, type, and time since diagnosis. Although

there is conflicting evidence, most research has indicated that more severe disease and functional impairment may increase risk for psychosocial difficulties (Lustig et al., 1996; Silver et al., 1995). A comprehensive rating of disease severity was developed for this study by incorporating multiple indicators, including functional status. However, mixing indicators of both severity and functional status may have confounded the problem. Different strategies for assessing disease factors have been employed previously (e.g., Lustig et al., 1996; McCormick et al., 1986), which has made direct comparison across studies difficult. In addition, studies often have been confounded by the use of the same source for measures of psychosocial outcomes and disease factors (i.e., mothers; e.g., Canning, Heller, Kelleher, 1996; Vandvik et al., 1991). These source variance and measurement issues (e.g., the lack of a valid, operational measure of severity) have been difficult to disentangle and may account for our lack of findings. Last, despite our rigorous recruitment strategies, we had a limited number of children with more severe diagnoses. Collaboration across multiple sites may be necessary to include children with a wider variety of disease characteristics.

Limited data have suggested that some families of children with JRA may lack social resources and support (Ireys et al., 1996; Vandvik et al., 1989), but our study found no differences in social support network size or perceived functional support for JRA and COMP families. As with disease severity, investigators have used multiple methods of assessing social support (e.g., Timko et al., 1992; Vandvik et al., 1989), many of which have included single item assessments (e.g., “Do you have a confidant?” or “Do you have sufficient support?”). In addition, no one has evaluated differences in levels of social support and its impact on distress among parents of children with JRA and controls. Using an established measure of social support and a controlled design, we failed to support our hypothesis that families of children with JRA would have fewer social resources than comparisons or that social support would be more strongly associated with parental distress in families of children with JRA. These results are consistent with previous studies using the same methodology with families of children with cancer (Noll et al., 1995) and sickle cell diseases (Noll et al., 1994).

This research has several methodological weaknesses. One should note that the sample had a limited number of children with severe disease, which may have affected the ability to detect problems for some groups of families at higher risk for difficulties. In addition, data were obtained at only one point in time at an average of 6 years postdiagnosis, reflecting long-term adjustment rather than



acute reactions. Earlier work has suggested that parents of children with chronic illness (Wallander & Varni, 1998), including JRA (Vandvik et al., 1991), may have more distress around the time of diagnosis. However, JRA is an unpredictable, chronic disease that can cause long-term disability (Peterson et al., 1997). Although many children experience long-term remission, up to two thirds of children may have recurrent disease in adulthood (Minden et al., 2000; Peterson et al., 1997; Zak & Pedersen, 2000). Because there are no established criteria for remission and cure for JRA, adult criteria have been used (Pinals, Masi, & Larsen, 1981), and a consensus group is currently studying this issue. Two thirds of our sample had active disease or only partial remission, which meant that these children were receiving ongoing treatment at the time of the assessment. Furthermore, our rheumatologists were reluctant to classify the remaining children as “cured” due to the chronic, episodic nature of JRA, the relatively young age of the children, and the significant risk of relapse.

Other methodological points include the fact that data were obtained from only one center and may not accurately reflect the status of parents and families from other locations. In addition, we used one source of information and questionnaires to measure our variables of interest. Observations, multiple reporting sources, Q-sorts, or interviews need to be included. Finally, our sample size limited our ability to detect variables that may moderate the relationship between caring for a child with JRA and parental distress. A longitudinal study across multiple sites that begins shortly after diagnosis is clearly needed.

Overall, this study supports Masten’s (2001) assertion that resilience is more common than traditionally believed. These families demonstrated a remarkable ability to adapt to the demands of having a child with a diagnosis of JRA. Alternatively, it is feasible that parents do not find having a child with JRA very demanding. This seems less likely, as limited data available near diagnosis (Vandvik et al., 1991) suggest considerable distress. The process of how parents adapt and what promotes these positive outcomes is less clear. Currently, data do not support broad-based interventions for all families of children with JRA, but some mothers may warrant additional attention.

## Acknowledgments

This project was supported by a grant to Robert B. Noll from the National Arthritis Foundation.

Received July 16, 2001; revisions received December 20, 2001, and March 16, 2002; accepted March 20, 2002

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## Notes

1 Because each child with JRA was matched to a classmate of the same gender, race, and age, it could be argued that a matched pairs *t* test would have been more appropriate. This would have been more powerful than a between-groups test if the reduction in the error term (a function of *r*) outweighed the effect of a loss in degrees of freedom ( $df = N - 1$  rather than  $n_1 = n_2 - 2$ ). Historically, we have found modest correlations on psychosocial variables between target and comparison families. Rather than reducing error, this matching procedure appears to equate groups on key demographic factors that otherwise may have contributed to spurious effects.

2 An astute reviewer suggested that a better test of the buffering hypothesis might assess the impact of social support on the association between disease severity and parental distress. Although we calculated adequate power to detect a large overall effect for the regression equation, power would have been extremely limited to adequately test the interaction effect with only a portion of the sample (i.e., 64 mothers or 45 fathers in the JRA group). Not surprisingly, when we ran these tests, none of the regressions was significant.

