RISK-RESISTANCE ADAPTATION MODEL FOR CAREGIVERS AND THEIR CHILDREN WITH SICKLE CELL SYNDROMES[1,2]

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ABSTRACT

This investigation examined the risk-resistance adaptation model for children with sickle cell disease and their primary caregivers. Participants were 55 children, ranging in age from 5 to 16 years with a mean age of 9 years 2 months, diagnosed with sickle cell disease and their primary caregivers, recruited from a university medical center. Measures included adjustment (i.e. primary caregiver and child adjustment), risk factors (i.e. disease and disability, functional independence, and psychosocial stressors), resistance factors (i.e. intrapersonal health locus of control, social-ecological), and stress processing (coping). Primary caregivers' adjustment was associated with developmental coping, $_{change}R^2 = .08$, *and child adaptation was associated with an internal health locus of control,* $_{change}R^2 = .22$. *An indirect effect of primary caregivers' coping on child adjustment was found through influence on primary caregivers' adjustment,* $_{change}R^2 = .11$. *The findings support research among other chronically ill populations that suggests an association between coping and disease adjustment. The results were interpreted to support the use of theoretically driven models in predicting the adaptation of children with chronic illness and adjustment in their caregivers.*

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INTRODUCTION

Sickle cell disease (SCD) is a chronic, hereditary disorder that affects 1 of every 400 to 500 African-Americans (1). Medical management of the disease is life-long and includes routine clinic appointments, emergency room visits, and frequent hospitalizations. The pain episodes require ongoing pain analgesia (2). Because the disease is chronic and has multiple symptoms, children and their families undergo significant stressors as they cope with normal developmental tasks and the medical regimens and sequelae associated with the illness (3,4).

Over the past several years, researchers have shifted from more descriptive accounts of chronically ill populations (5–7) to the development of more conceptually based, theoretically driven models that predict the adaptation of children and their families (1,8). These investigators have demonstrated the utility of theoretical frameworks to examine both the commonalities found across various childhood chronic illnesses and the characteristics unique to a particular illness. As Brown, Doepke, and Kaslow (1)

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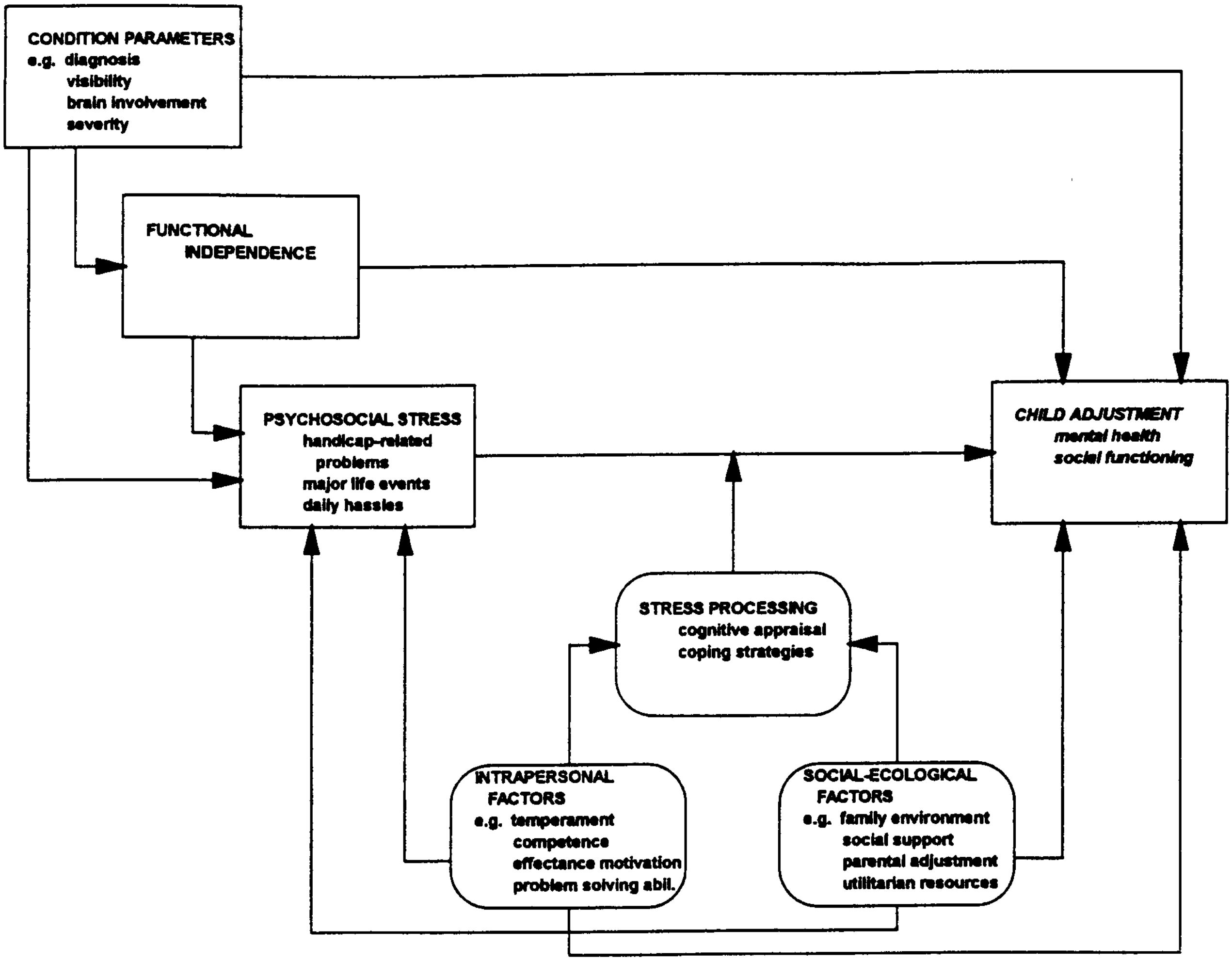


FIGURE 1: Wallander's risk-resistance adaptation model. [Reprinted with permission Guilford Press.]

concluded, the consistent use of a theory-driven model may serve as a basis for the comparison of results across different groups of children and adolescents with chronic illnesses and their families.

Wallander and colleagues (8) developed a risk-resistance adaptation model that considers risk and resistance factors in predicting children's adaptation to a chronic illness. Risk factors include disease and disability parameters, functional independence, and psychosocial stressors. Key resistance factors encompass intrapersonal dimensions (social and academic competence and self-esteem), social–ecological factors (family environment, members' adaptation, and utilitarian resources), and stress processing (locus of control, coping strategies). Wallander and colleagues' model is presented in Figure 1. For a complete review of the risk-resistance adaptation model, particularly as it pertains to SCD, see Brown, Doepke, and Kaslow (1).

Researchers are interested in the psychological adaptation of children with chronic illnesses and adjustment in their families as they negotiate the stressors associated with managing an illness that requires continual medical attention. In their investigation of the stability and change in the psychological adaptation of mothers of children and adolescents with SCD and cystic fibrosis, Thompson and colleagues (9–11) found that current levels of daily stress and styles of coping accounted for over 40% of the variance in maternal adjustment even after controlling for disease severity. Similarly, Gil et al. (12) found that children who were active in their attempts to cope achieved higher levels of school, household, and social functioning during periods of painful episodes. Coping style accounted for up to 15% of the variance in various activities after controlling for disease severity. Finally, Kliewer and Lewis (13) examined the contribution of parenting and family variables to the coping processes of children and adolescents with SCD. After controlling for the effects of age, gender, family structure, and type of SCD, they found a cohesive family environment was associated with children who engaged in active coping strategies. This finding was consistent with other investigations. In fact, family cohesion explained nearly one-fifth of the variance in children's active coping strategies. Additionally, they found that greater parental use of active coping strategies predicted the use of avoidance coping by children. Parental coping accounted for over 40% of the variance in children's coping. Kliewer and Lewis interpreted their data to suggest the influence of parental coping and adjustment on coping processes in children with SCD. Further, Lewis and Kliewer (14) tested mediating and moderating models of hope, coping, and adjustment in children with SCD. After controlling for the effects of family type, type of SCD, frequency of SCD-related complications, and age of the child, they found that hope was negatively associated with children's anxiety when active coping, support coping, and distraction coping were high. Together, the variables accounted for nearly 50% of the variance on anxiety scores. In a recent investigation designed to investigate family

TABLE 1

Disease Severity Measures, Syndrome Types, and Demographic Characteristics for Sickle Cell Patients

	n	%	*M*	*SD*	Range
Disease severity parameter					
Days hospitalized past 12 months			8.34	14.69	0–100
Emergency room visits past 12 months			4.50	8.76	0–50
Pain episodes past 12 months			7.86	14.01	0–80
Frequency of symptoms past year			1.82	1.86	0–7
Frequency of symptoms ever			2.92	1.98	0–8
Hemoglobin mean of last 3 clinic visits			8.40	2.51	6.3–13.00
Sickle cell syndrome type					
HbSS	43	78.2			
HbSC	10	18.2			
HbS-beta thalassemia	2	3.6			
Caregivers					
Mothers	44	80.0			
Fathers	5	9.0			
Grandmothers	3	5.0			
Aunts	2	4.0			
Uncles	1	2.0			
Marital status of caregiver					
Married	16	29.0			
Never married	20	36.0			
Separated or divorced	16	29.0			
Widowed	3	5.0			
Annual income					
<$10,000	31	56.0			
$10,000–$19,999	11	20.0			
$20,000–$30,999	11	20.0			
$31,000–$40,000	1	2.0			
>$40,000	1	2.0			
Caregivers who completed high school	36	66.0			
Classroom type					
Regular	41	74.5			
Special education	14	25.0			
Sex					
Males	38	69.0			
Females	17	31.0			
Chronological age					
Caregivers			33.3	7.6	20–59
Children			9.2	2.6	5–16
Children's grade level			4.2	3.1	K–12

competence in adolescents with SCD, when demographic variables (age, gender) and medical variables (severity of disease) were controlled, greater family competence was associated with fewer internalizing and externalizing behavioral symptoms in adolescents (15).

Although the risk-resistance adaptation model has driven the research program of Wallander and Varni, the model has been used primarily with children with physical and developmental disabilities, including children with congenital orthopedic disabilities, limb amputations, and spina bifida. For a complete review of this program, see Wallander and Thompson (6). Our investigation examined the psychological adaptation of both children and adolescents with SCD and the adjustment of their primary caregivers, using all aspects of the risk-resistance adaptation model, including disease-specific factors influencing adjustment. Because components of the model have been demonstrated effective in predicting psychological adaptation in youth with physical disabilities and other chronic conditions and in their families, it was used in our investigation. This allowed a comparison of children with SCD to the already available literature of children with physical disabilities and their families.

We hypothesized that once disease and disability parameters were adjusted, the resistance factors (i.e. intrapersonal factors, social–ecological factors, and stress processing) would predict adaptation (see Figure 1). In addition, we predicted that there would be a positive association between primary caregivers' and children's psychological distress and several factors, including perceived stress from illness-related causes and recent and past stressful events, external health locus of control expectations, disengagement or palliative coping, and family enmeshment and disorganization. When child and primary caregiver resistance factors were considered separately, each was hypothesized to account for a significant amount of variance in adjustment and adaptation over and above that accounted for by disease and disability parameters (physical symptoms and functional limitations). Finally, guided by the work of Kliewer and Lewis (13) that demonstrated the relationship between child and maternal adaptation, the association of child coping and adjustment on children's adaptation was predicted to be related to primary caregiver coping, with the relationship mediated by primary caregiver adjustment.

METHOD

Participants

Participants were 55 children and adolescents with SCD and their primary caregivers. The children and adolescents received treatment at a comprehensive National Institutes of Health (NIH) sickle cell center. Participants were referred to us by the pediatric hematologists as part of a larger neuropsychological investigation designed to examine the cognitive, academic, and emotional functioning of children and adolescents with SCD (1,3,16,17). Patients and caregivers were referred if the children with SCD were medically stable and if both the caregiver and the patient were deemed by the physician to be of sufficient mental abilities to complete the requirements of the protocol. Because children were referred by their pediatric hematologists for learning and behavioral problems, more males (69%) were referred than females. This may explain why a higher frequency of children with behavior problems was likely to be referred to the study than children with fewer behavioral problems.

Our sample reflected a fairly low socioeconomic group and is characteristic of other investigations of children and adolescents with SCD (18,19). Of the 55 children and adolescents participating, 43 had HbSS (78%), the most severe form of the disease, in which the individual is homozygous for the condition; 10 had HbSC (18%), a heterozygous condition that is frequently associated with a more benign presentation; and 2 had HbS-beta thalassemia (4%), a compound heterozygous condition and the least severe form of the disease. The means, standard deviations, and ranges of indices of disease severity, SCD types, and demographic characteristics are presented in Table 1.

Procedure

After obtaining informed consent from primary caregivers, interviews were conducted at the NIH sickle cell center by research assistants. The children and their primary caregivers completed questionnaires and interviews during the interview session. Primary caregivers were paid $30 for participation.

MEASURES

Primary Caregivers' Adjustment

The Symptom Checklist-90-Revised (SCL-90-R) (20) was used as an index of primary caregivers' adjustment. The SCL-90-R is a 90-item self-report measure of symptoms of psychological distress. The items are rated on a 5-point Likert-type scale that assesses caregiver distress by measuring psychological symptoms. Responses are classified into nine subscales of psychopathology (depression, hostility, psychoticism, somatization, obsessive–compulsive, interpersonal sensitivity, anxiety, phobic anxiety, and paranoid ideation) and a Global Severity Index (GSI), which combines information on frequency and intensity of symptoms. Internal consistency coefficients ranging from .77 to .90 and test–retest reliabilities ranging from .78 to .90 have been reported for the symptom dimensions of the SCL-90-R. The SCL-90-R has been demonstrated valid in the assessment of psychological distress for caregivers who are African–American (9,19–21). The SCL-90-R GSI was used in the analysis.

Child Adaptation

The Child Behavior Checklist (CBCL) (22) was used to measure child adaptation. The CBCL is a 113-item measure that addresses a broad range of internalizing (e.g. depression, anxiety) and externalizing (e.g. acting-out behaviors, attention problems) factors for children and adolescents ranging from 4 to 16 years of age. It was completed by each of the caregivers for their children with SCD. In our study, primary caregivers rated the severity of each symptom they observed in their children during the past 6 months. The CBCL has been used extensively in the child psychopathology and pediatric psychology literatures (17,22–24) and has good psychometric properties (22). The CBCL also has proved effective in a number of investigations with minority and disadvantaged youth (17,23,25) and the corresponding psychometric data for these instruments have been found to be adequate in these investigations (26). Both the CBCL internalizing and externalizing broad band factors (*T* scores) were used in the analyses.

Children's Disease and Disability Parameters

The measure of disease severity of the children with SCD consisted of a composite of standardized indices that were averaged and included caregivers' reports of the number of days hospitalized, emergency room contacts, and pain episodes over the past 12 months. In addition, hemoglobin levels averaged over the past three clinic visits were reverse scored and incorporated into the disease severity composite. Hemoglobin levels were obtained through a review of medical records. Recall biases are associated with reporting of past health care utilization, but this procedure has been used successfully for similar indices of disease severity in previous investigations of children and adolescents with sickle cell syndrome (11,14,17,27–29).

Information on diagnostic type of sickle cell syndrome (HbSS versus HbSC and HbS-beta thalassemia) for the children were obtained from the clinic's computer system. The frequency of symptoms (e.g. cerebral vascular accidents, meningitis, delayed growth, infections) in the past year and the lifetime frequency of symptoms were recorded.

Children's Functional Independence

The Vineland Adaptive Behavior Composite (Vineland-ABC) (30) is a semistructured interview with scales that assess several domains of functioning: communication (receptive, expressive, and written language), daily living (personal, domestic, and community living skills), socialization (interpersonal, play/leisure, and coping abilities), and motor skills (fine- and gross-motor abilities). The Vineland was administered to primary caregivers and used to assess functional independence or activities of daily living (e.g. talking, personal care skills, social skills, and motor skills) of the children with SCD. Each of the areas yields standard scores with means of 100 and standard deviations of 15. An adaptive behavior composite score derived from the sum of the domain scores provides an overall measure of adaptive functioning. Each of the adaptive behavior scores is normed for age, thus allowing interpretation of these domains across a wide age span (31). The Vineland scales were standardized on a large representative national sample and have good split-half, test–retest, and interrater reliability, as well as adequate construct, content, and criterion-related validity.

Primary Caregivers' Psychosocial Stressors

Primary caregivers' current income and caregivers' education or social class were designated as stressors in our investigation because of the relationship between poverty and stress (32). Additional information on recent and past family stressors was collected from the primary caregivers.

The Family Inventory of Life Events and Changes scale (FILE) (33) is a self-administered questionnaire with nine subscales that function as indices of past and recent family stressors. Recent stressors are those within the past year, and the frequency of those stressors within a lifetime is considered past stressors. There is little overlap between these two variables of family stress, and construct validity indicates good convergent validity between the FILE as a measure of family stress and measures of adjustment within the family. Overall reliability of the FILE has been reported to be adequate ($r = .81$), and factor analysis has provided evidence for construct validity (34). A total score is calculated for both recent (FILE-R) and past (FILE-P) life events. The measure was administered to primary caregivers.

Children's Intrapersonal Factors

Competence: The Social Competence factor of the CBCL (22) measures observed social behaviors related to contacts with peers, family, and friends. This particular measure is separate from the symptoms of internalizing and externalizing behaviors that were included in the adaptation measure. Social competence is assessed through parent responses to questions aimed at measuring the child's or adolescent's involvement in activities (e.g. sports, hobbies, organizations and clubs, chores at home, number of friends, number of activities with friends, performance in academic subjects, behavior with peers and siblings) and the quality of the child's performance in these activities. Test–retest reliability has been reported to be very high ($r = .97$) and criterion validity has been demonstrated to be high, particularly in discriminating clinical groups, with lower social competency scores being associated with higher frequencies of mental health referrals. Thus, the total *T* score of the CBCL competence score was used in the analysis. The measure was completed by caregivers regarding the social competencies of their children with SCD.

Self-Esteem: The Negative Self-Esteem factor of the Children's Depression Inventory (CDI) (35) is one of the five primary factors indicative of problems with self-esteem and self-competence. The instrument is a self-report measure completed by the children with SCD. Included in this measure are six items (e.g. "There are some bad things about my looks," "I do not like myself") that is scored from 0 to 3, with a higher score indicative of more problems with self-esteem. Internal consistency of the CDI Negative Self-Esteem factor has been reported to be .68, and moderate correlation coefficients have been reported between the Negative Self-Esteem factor and other factors of the CDI (correlations factor and total CDI scores $r = .79$). In our investigation, positive self-esteem was operationalized as fewer problems that are indicative of negative self-concept, and for this reason, the scoring on the factor was reversed to represent positive self-concept. The CDI Negative Self-Esteem factor was used in the analysis.

Health Locus of Control: The Children's Health Locus of Control (HLOC) scale (36) is a 20-item self-report measure developed to assess children's locus of control pertaining to health and illness. The children with SCD completed it. Scores may range from 20 to 40, with a total higher score indicative of greater internality. Kuder-Richardson coefficients have been reported by Parcel and Meyer (36) to range from .72 to .75 and a moderate association ($r = .40$) has been reported between the HLOC and the Nowicki-Strickland Children's Locus of Control Scale (37). Although the measure was originally developed for children aged 7 to 12 years, other investigators have used the HLOC successfully with children and adolescents up to the age of 16 years (38–40). The HLOC total score was used for analysis.

Primary Caregivers' Social–Ecological Factors

Family functioning was examined both in terms of the overall adjustment of the family environment and the utilitarian support provided by the family. The Family Adaptability Cohesion Evaluation Scales-II (FACES-II), the Family Support Scale (FSS), and the Family Resource Scale (FRS) were used as indices of social–ecological factors. Each of these instruments was completed by the caregivers about their families.

Family Environment and Members' Adaptation: The FACES-II (41) is a 30-item self-report measure that was completed by primary caregivers in our investigation. The FACES-II is based on a circumplex model of family functioning and measures how families perceive their functioning on the dimensions of cohesion (FACES Cohesion) and adaptability (FACES Adaptability). The 16-item FACES Cohesion subscale contains statements that refer to the degree of emotional bonding in the family (e.g. "Family members share interests and hobbies with each other"). The 14-item FACES Adaptability subscale consists of statements assessing the family's ability to adjust the power balance appropriately within the system (e.g. "Each family member has input regarding major family decisions"). This instrument has been reported to differentiate adequately between well-functioning and dysfunctional families and is used to identify families that may be at risk for developing destructive patterns of family interactions (42,43). Extensive normative data have been presented for the FACES-II, and its psychometric properties have ranged from adequate to excellent (42,44). Both the FACES Adaptability and FACES Cohesion scores were considered in the analysis.

Utilitarian Resources: The Family Support Scale (45) measures primary caregivers' perceptions of the helpfulness of various sources of formal and informal family network supports. This scale was designed to model Brofenbrenner's (46) formulation of the ecology of human development, with the notion that the family unit is embedded within broader ecological systems of formal and informal kinship units (e.g. relatives and friends) that are further embedded within larger social units (e.g. church, work, social organizations). The FSS includes 18 items rated on a 5-point Likert-type scale ranging from *not at all helpful* to *extremely helpful.* The Helpfulness index was used in this investigation and reflects primary caregivers' overall satisfaction with their social support. Internal consistency of the measure has ranged from .77 to .87 with a test–retest reliability of .91 over a 1-month interval (45,47). The FSS total score was used for analysis.

The Family Resource Scale (48) is a 30-item self-report measure that assesses the adequacy of family resource support. The scale was devised to assess types of resources identified as major components of both intrafamily and extrafamily support. Family resource support, including resources provided by other persons, has been conceptualized as one component of the social support construct (49). Factors of the FRS include personal growth and support, health, physical necessities, physical shelter, intrafamily support, employment, childcare, and personal resources (48). The individual items of the FRS are ordered in a hierarchy to reflect the most to the least basic resources, and needs are rated on a 5-point Likert-type scale ranging from *not at all adequate* to *almost always adequate.* The FRS has been shown to be reliable and valid for assessing family needs. Internal consistency of the measure has ranged from .92 to .97, with a test–retest reliability of .52 over a 2- to 3-month interval. In a sample of 45 mothers of mentally retarded, handicapped, or developmentally at-risk preschoolers, total FRS scores were associated significantly with mothers' personal well being. The FRS also has been demonstrated to be a particularly valid instrument for economically disadvantaged groups (50). The FSS total score was used for analysis.

Primary Caregivers' Stress Processing

Coping Strategies: The Coping Strategies Inventory (CSI) (51) is a 72-item, self-report questionnaire that was administered to primary caregivers regarding their own coping strategies. The CSI assesses coping along the dimensions of engagement versus disengagement and problem-focused versus emotion-focused coping. Eight subfactors (problem-solving, cognitive restructuring, expression of emotion, social support, problem-avoidance, wishful thinking, self-criticism, medical coping) are included in the scale, with second-level factors of problem-focused engagement, problem-focused disengagement, emotion-focused engagement, and emotion-focused disengagement. We used two broadband factors (engagement and disengagement) for analyses. Based on the recommendation of Armstrong, Lemanek, Pegelow, Gonzalez, and Martinez (52), only 32 of the original 72 items were used because of the low education characteristic of our sample. Consistent with Armstrong et al. (52), the 32-item total was composed of the 4 items on each of the eight scales that had the strongest factor loadings, as reported by Tobin et al. (51). Items are presented as statements of coping styles and parents used a 5-point Likert-type scale, ranging from *not at all true* to *extremely true,* to rate the degree to which a statement reflected their own styles. Subjects were presented with the same description of a child experiencing the symptoms of a vaso-occlusive pain episode and were asked to respond to the CSI as if this event was actually happening.

> Your child appears tired and sluggish. He/she may be running a slight fever, but he/she is also complaining of a headache, as well as pain in his/her shoulders, chest, and abdomen. At times, it seems that the pain is quite severe, as your child cries and sometimes screams that it hurts. (52)

The CSI has been demonstrated to have an acceptable test–retest reliability of .73 and an internal consistency of .83 with college students (50). Concurrent validity with the Kidcope, a widely used child-coping measure, has been reported to range from moderate (.33) to high (.77) (53). Only the CSI Engagement and CSI Disengagement scores were used in the analysis.

Analyses

First, descriptive statistics were computed and adjustment scores were compared with the normative sample. The number of mothers and children who met criteria for poor adjustment also was calculated. In addition, to determine whether the children differed as a function of hemoglobinopathy (HBSS, HbSC, or HBS-beta-thalassemia), a series of one-way analyses of variance (ANOVAs) were computed for each of the measures.

Second, zero-order correlations obtained on all measures were examined among the variables. Because of the limited sample size, one or two variables were chosen to represent each of the domains depicted in the risk-resistance adaptation model (see Figure 1). The selection of these variables was based on: (a) the strength of the bivariate correlation between each variable and primary caregiver and child adaptation within a given domain; (b) the strength of intercorrelations among variables in each domain; (c) the findings of research using this model; and, when possible, (d) multiple sources of ratings.

Based on the risk-resistance adaptation model, hierarchical regression equations were used. First, the number of physical symptoms ever was included in the analysis because of its primary theoretical importance to the disease and disability factor, to adjust for illness severity in the regression model, and its statistically zero-order correlation with primary caregiver adaptation scores (SCL-90-R GSI). Second, the measure of functional independence was not included in the model because of the absence of a statistically significant association between the Vineland-ABC and primary caregiver adaptation. Third, to control for social class, primary caregivers' income was considered, although it was not associated with caregiver adaptation scores (SCL-90-R GSI). In addition, since the FILE-P life events scores were significantly associated with caregiver adaptation scores (SCL-90-R GSI), the FILE-P also was entered into the regression model as a psychosocial stressor. Fifth, the child's social competence measure (CBCL competence) was entered as an interpersonal resistance factor, although it was not significantly associated with primary caregiver adaptation (SCL-90-R GSI). Sixth, a measure of social–ecological factors, the Dunst-FRS was entered because it was a social–ecological variable that was associated with primary caregiver adaptation scores (SCL-90-R GSI). Finally, the CSI Disengagement scale was chosen to represent the stress processing domain because it was significantly associated with primary caregiver adaptation scores (SCL-90-R GSI).

A child model also was constructed and based on the risk-resistance adaptation model; hierarchical regression equations were used. Consistent with the primary caregivers' model, selection for inclusion was based on the strength of the bivariate correlations, the strength of the intercorrelations among variables in each domain, findings of past research, and the use of multiple sources of ratings. First, the variable of physical symptoms during the past year was included in the analysis because of its primary theoretical importance to the disease and disability risk factor, to adjust for illness severity in the regression model, and its significant bivariate association with the CBCL Externalizing score of children's adjustment. Second, functional independence as measured by the Vineland-ABC was included in the model because of the significant bivariate association between the Vineland-ABC and children's CBCL Internalizing and CBCL Externalizing factors of adaptation. In addition, because primary caregivers' education was significantly associated with the CBCL Externalizing factor of children's adjustment and because primary caregivers' education was conceptualized as an index of social class, primary caregivers' education was used as a variable to represent psychosocial stressors. Further, because the CBCL Social Competence measure was derived from the CBCL, which also was used as the dependent measure, the CBCL Competence measure was not employed in the regression analyses and, thus, the CDI Negative Self-Esteem factor was used as a resistance factor. This measure was significantly associated with CBCL Externalizing scores of children's adjustment. Because the HLOC total score was significantly associated with both the CBCL Internalizing and CBCL Externalizing scores of children's adjustment, it also was used as a measure of intrapersonal resistance. Finally, since the FACES Cohesion measure was associated with the CBCL Externalizing factor of children's adjustment, this measure was entered as an index of social–ecological functioning. Separate hierarchical regression equations were constructed to predict CBCL Internalizing and CBCL Externalizing scores of adjustment where the aforementioned variables representing risk and resistance factors were entered into the equation.

To test the relationships of primary caregivers' coping (disengagement) and children's adaptation as mediated by primary caregivers' adjustment (SCL-90-R GSI), a series of correlation coefficients were computed. Relationships were examined among the SCL-90-R GSI, the CSI Disengagement, and child adaptation as measured by the CBCL. To determine the mediating role of primary caregivers' CSI Disengagement scores and the relationship between primary caregiver adjustment (SCL-90-R GSI) and child adjustment (CBCL Internalizing, CBCL Externalizing), a series of equations were used. A variable is said to mediate the influence of a predictor on a criterion when it further illustrates how a predictor is related to the criterion. It offers evidence for at least part of the mechanism by which a predictor exerts an outcome of interest (54,55). Consistent with Baron and Kenny's (56) criteria for determining mediation, the relationship between primary caregivers' disengagement coping and each of the measures of child adaptation was examined both in the bivariate cases and in the context of regression models that included the potentially mediating presence of primary caregivers' adjustment (SCL-90-R GSI).

RESULTS

Primary Caregivers' Model

Descriptive statistics are presented in Table 2. Primary caregivers' SCL-90-R GSI scores showed wide variability, ranging from the 3rd percentile to above the 99th percentile. The mean adjustment scores on the SCL-90-R GSI corresponded to the 67th percentile of the normative sample. Using the 90th percentile as clinically elevated ($T \geq 63$), 35% ($n = 19$) of primary caregivers met criteria for poor adjustment and 65% ($n = 36$) demonstrated at least adequate adjustment. No statistically significant differences were found on any of the primary caregiver measures as a function of children's gender, as indicated by t tests for independent samples. No statistically significant differences were found for any of the measures except for the measures of disease severity where children with HbSS evidenced greater symptoms of disease severity.

TABLE 2
Descriptive Statistics and Correlations for Each of the Variables

	Descriptive Statistics			Correlation Data		
				Caregiver Adaptation	Child Adaptation	
	M	*SD*	Range	SCL-90-R *r*	CBCL-Internalizing *r*	CBCL-Externalizing *r*
Child adaptation						
CBCL Internalizing[a]	56.00	11.77	31–77	.21*	—	.56****
CBCL Externalizing[a]	51.66	10.13	30–72	.36****	.56****	—
Primary caregiver adaptation						
SCL-90-R GSI[b]	56.58	14.11	30–92	—	.21*	.36****
Risk factors						
Disease and disability						
Disease severity[a]	0.01	0.75	−0.68–4.07	.02	.04	.06
Physical symptoms past year[a]	1.86	1.92	0–7	.14	−.11	.22*
Physical symptoms ever[a]	2.95	2.05	0–8	.26**	.13	.18
Functional independence						
Vineland-ABC[a]	87.29	20.21	45–130	−.17	−.25**	−.41****
Psychosocial stressors						
Caregiver's income[b,c]	1.76	.96	1–5	−.12	−.22*	−.11
Caregiver's education[b,d]	—	—	1–4	.05	−.19	−.26**
FILE Recent Stressors[c]	10.96	7.00	1–31	.16	.10	.03
FILE Past Stressors[c]	9.46	8.52	0–35	.27**	.24*	.24*
Resistance factors						
Child interpersonal factors						
Competence						
CBCL Social Competence[c]	41.11	6.86	24–55	.09	−.24*	−.20
Self-esteem						
CDI negative self-esteem	46.94	11.36	39–83	.04	−.07	−.29**
Social–ecological factors						
Family environment and members' adaptation						
FACES II Adaptability[e]	47.18	7.34	34–65	−.10	.20	−.14
FACES II Cohesion	59.22	9.69	33–78	−.26**	.05	−.23*
Utilitarian resources						
DUNST FRS[e]	104.56	21.53	58–145	−.25**	.05	−.21*
DUNST FSS[e]	35.69	13.49	6–69	.08	−.06	−.20
Stress processing						
Locus of control						
HLOC Internality[a]	6.18	1.89	1–9	−.25**	.18	.18
HLOC Externality[a]	12.72	6.80	0–34	.07	−.41****	−.38****
HLOC total[a]	18.90	7.88	3–43	.00	−.40****	−.38****
Coping strategies						
CSI Engagement[b]	13.21	2.86	4.33–18.25	.13	−.19	.05
CSI Disengagement[b]	8.95	2.34	4.80–14.90	.31****	.25**	.15

Note: The entire correlation matrix for the primary caregiver and child variables is available from the authors upon request.

$^{*}p < .10$, $^{**}p < .05$, $^{****}p < .01$.

[a] Child measure.

[b] Caregiver measure.

[c] For caregiver income: 1 < \$10,000, 2 = \$10,000–\$19,000, 3 = \$20,000–\$30,000, 4 = \$31,000–\$40,000, 5 > \$40,000.

[d] For caregiver education: 1 = completed grade school, 2 = completed high school, 3 = completed some college, 4 = completed college.

[e] Family measure as reported by caregiver.

The results of the bivariate correlation coefficients for primary caregiver and child adaptation are presented in Table 2. None of the variables (i.e. caregiver, child) were associated with chronological age of the children with SCD. Based on the risk-resistance adaptation model, the results of a hierarchical multiple regression analysis (57) revealed that disease severity did not correlate significantly with SCL-90-R GSI scores (${}_{\text{change}}R^2 = .00$, *ns*; *df* = 1, 53). Disease severity accounted for less than 1% of the variance in SCL-90-R GSI scores. The addition of primary caregivers' income and the FILE-P revealed that these variables accounted for approximately 8% of the variance in SCL-90-R GSI scores (${}_{\text{change}}R^2 = .08$, *ns*) above and beyond that of disease severity. Subsequently, the Social Competence measure of the CBCL, an index of the resistance portion of the model, was entered and accounted for less than 1% of the variance in SCL-90-R GSI scores (${}_{\text{change}}R^2 = .00$, *ns*). In addition, the Dunst-FRS was entered as a social–ecological resistance factor and accounted for 4% of the variance (${}_{\text{change}}R^2 = .04$, *ns*). Finally, the CSI Disengagement variable was entered as an index of stress processing and accounted for 8% of the variance in SCL-90-R GSI scores (${}_{\text{change}}R^2 = .08$, $p = .04$). The results of this analysis and the final standardized partial regression coefficients (betas) for each variable after each selected variable was entered into the regression equation are reported in Table 3. The results of this regression

TABLE 3

Summary of Hierarchical Analysis Predicting Primary Caregiver Adaptation

Step	Variable Added	R^2	ΔR^2	Δdf	ΔF	β^a	F^a
1	Severity	.000	.000	1, 53	<1	−.076	<1
2	FILE past					.209	2.18
	caregiver's income	.078	.078	3, 51	2.16	.052	<1
3	CBCL social competence	.079	.001	4, 50	<1	.027	<1
4	FRS	.117	.038	5, 49	2.09	−.205	2.16
5	CSI disengagement	.193	.076	6, 48	4.53***	.285	4.53***

*** $p < .04$.

[a] These are for the final model.

TABLE 4

Summary of Hierarchical Regression Analysis Predicting Child Adaptation as Measured by Externalizing and Internalizing Behavior

Step	Variable Added	R^2	ΔR^2	Δdf	ΔF	β^a	F^a
			Externalizing				
1	Disease severity	.010	.010	1, 51	<1	.003	<1
2	Vineland-ABC	.078	.067	2, 50	3.62*	−.213	3.28*
3	Caregiver's education	.085	.007	3, 49	<1	.068	<1
4	CDI negative self-esteem	.149	.064	4, 48	3.59*	−.115	<1
5	FACES II cohesion	.178	.030	5, 47	1.69	−.214	2.99
6	HLOC	.394	.216	6, 46	16.40******	−.499	16.40******
			Internalizing				
1	Disease severity	.005	.005	1, 51	<1	.126	<1
2	Vineland-ABC	.021	.016	2, 50	<1	−.118	<1
3	Caregiver's education	.021	.000	3, 49	<1	.089	<1
4	CDI negative self-esteem	.039	.019	4, 48	<1	.017	<1
5	FACES II cohesion	.042	.002	5, 47	<1	−.074	<1
6	HLOC	.215	.173	4, 46	10.15*****	−.445	10.14*****

* $p < .10$, ***** $p < .003$, ****** $p < .001$.

[a] These are for the final model.

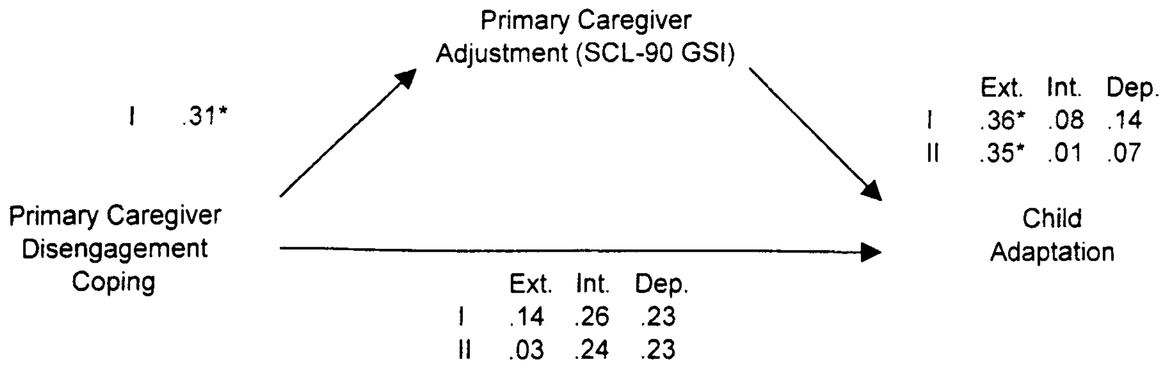
equation revealed that the CSI Disengagement scale contributed significantly to the equation.

Child Model

Descriptive statistics for each of the child variables also are presented in Table 2. Children's adjustment scores on the CBCL Internalizing domain suggested significant variability, ranging from the 4th percentile to above the 99th percentile. Scores on the CBCL Externalizing domain ranged from the 3rd percentile to above the 99th percentile. The mean adjustment score on the CBCL Internalizing score corresponded to the 69th percentile of the normative sample, and the mean adjustment score of the CBCL Externalizing domain corresponded to the 55th percentile of the normative sample. Using the 92nd percentile as clinically elevated ($T \geq 65$), 20% ($n = 11$) of the children met criteria for poor adjustment on the Internalizing domain of the CBCL, and 80% ($n = 44$) demonstrated at least adequate adjustment. Again, using the 92nd percentile as clinically elevated ($T \geq 65$), 35% ($n = 19$) of the children met criteria for poor adjustment on the Externalizing domain of the CBCL, and 66% ($n = 36$) demonstrated at least adequate adjustment. No statistically significant differences were found on any of the child measures as a function of children's gender, as indicated by t tests for independent samples.

Based on the risk-resistance adaptation model using the CBCL Internalizing factor as a measure of adaptation, the results of a hierarchical multiple regression analysis revealed that disease severity did not significantly predict children's CBCL Internalizing factor scores (${}_{\text{change}}R^2 = .01$, *ns;* $df = 1, 51$). Thus, consistent with the primary caregivers' model, disease severity accounted for less than 1% of the variance in children's CBCL Internalizing factor scores. The addition of Vineland-ABC scores accounted for approximately 2% of the variance in children's internalizing adjustment (${}_{\text{change}}R^2 = .02$, *ns*) above and beyond that of disease severity. Subsequently, primary caregivers' education, an index of psychosocial stressors, was entered and accounted for less than 1% of the variance in children's CBCL internalizing scores (${}_{\text{change}}R^2 = .00$, *ns*). The addition of the CDI self-esteem interpersonal–resistance factor accounted for less than 2% of the variance in CBCL Internalizing factor scores (${}_{\text{change}}R^2 = .02$, *ns*). The social–ecological factor of the FACES Cohesion measure of family environment accounted for less than 1% of the variance in internalizing factor scores (${}_{\text{change}}R^2 = .00$, *ns*). Finally, the HLOC total score was entered as an index of intrapersonal resistance and accounted for 17% of the variance in children's CBCL scores (${}_{\text{change}}R^2 = .17$; $F_{\text{change}} = 10.2$, $p < .003$). The results of these analyses and the final standardized partial regression coefficients (betas) for each variable after each selected variable was entered into the equation are presented in Table 4.

Using the CBCL Externalizing factor scores as a measure of adaptation, the results of a hierarchical multiple regression analysis also revealed that disease severity did not predict children's CBCL Externalizing factor scores (${}_{\text{change}}R^2 = .01$, *ns;* $df = 1, 51$). Again, consistent with the primary caregivers' model, disease severity accounted for less than 1% of the variance in children's CBCL



Note. - * indicates p<.05. I indicates the bivariate correlation. II indicates the standardized beta weight from the model containing both coping and adjustment. Ext.=CBCL Externalizing, Int.=CBCL Internalizing, Dep.=CDI Depression

FIGURE 2: **Testing the mediation model.**

Externalizing symptoms of behavioral adaptation. The addition of the Vineland-ABC accounted for 7% of the variance in children's CBCL Externalizing scores beyond that of disease severity ($_{\text{change}}R^2 = .07$, $F_{\text{change}} = 3.63$, $p = .07$). Subsequently, primary caregivers' education was entered and accounted for less than 1% of the variance in children's CBCL Externalizing scores ($_{\text{change}}R^2 = .01$, *ns*). The addition of the CDI self-esteem interpersonal resistance factor accounted for 6% of the variance in CBCL Externalizing scores ($_{\text{change}}R^2 = .06$, $F_{\text{change}} = 3.59, p = .07$). The social–ecological factor of the FACES Cohesion measure of family environment accounted for 3% of the variance in CBCL Externalizing symptoms ($_{\text{change}}R^2 = .03$, *ns*). Finally, the HLOC total score was entered as an index of interpersonal resistance and accounted for 22% of CBCL Externalizing symptoms ($_{\text{change}}R^2 = .22$, $F_{\text{change}} = 16.4$, $p < .001$) (see Table 4 for the results of the regression analyses).

Tests of Relationship Between Primary Caregivers' Coping and Children's Adaptation as Mediated by Primary Caregivers' Adjustment

The results of these analyses are presented in Figure 2. Primary caregiver disengagement coping was associated with CBCL Internalizing ($r = .26, p < .06$). Primary caregivers' disengagement coping was also found to be associated with primary caregiver adjustment (SCL-90-R GSI) ($r = .31$, $p < .02$). When controlling for CSI Disengagement, the SCL-90-R GSI accounted for a statistically significant additional portion of the variance in child adjustment as measured by the CBCL Externalizing scale ($R^2_{\text{change}} = .11, p < .02$). Three of the four criteria for a mediational model were met for the CBCL Externalizing outcome measure (53): (a) primary caregiver coping was not associated with child adaptation at a statistically significant level; (b) primary caregiver coping was associated with primary caregiver adjustment; (c) primary caregiver adjustment was associated with child adaptation; and (d) when primary caregiver adjustment was entered into the regression equation along with caregiver coping, the standardized beta weight for coping decreased (from .14 to .03). While the criteria for mediation were partially met, the direction of the mediating effect and the exact mechanisms by which it operates cannot be demonstrated with certainty due to the observational and correlational nature of this investigation. The mediational model was not supported for the CBCL Internalizing or CDI depression child adaptation outcome measures.

DISCUSSION

Our investigation examined factors predictive of psychological adaptation in children with SCD and adjustment in their primary caregivers. Findings supported previous research indicating that primary caregiver adjustment is associated with primary caregiver coping strategies (9–11,21,58). Internal health locus of control, which is characterized by children's beliefs that they are able to exert control over their health-related concerns, best predicted children's adaptation, as rated by their primary caregivers. The finding that approximately 35% of the primary caregivers met criteria for poor adjustment is consistent with the findings of Thompson and associates (11,21), both for mothers of children with cystic fibrosis and mothers of youth with SCD. No statistically significant differences were found in primary caregivers or children's adjustment as a function of children's gender or children's hemoglobinopathy (i.e. HbSS versus HbSC).

Some support was provided for the hypothesized effect of stress processing as a resistance factor depicted in the risk-resistance adaptation model. Specifically, when disease risk factors (i.e. disease severity) and psychosocial stressors (i.e. life events, primary caregivers' income) were adjusted, less use of disengagement coping (characterized by avoiding emotions, wishful thinking, problem-avoidance, self-criticism, and social withdrawal) by primary caretakers predicted better adjustment (i.e. fewer adjustment difficulties). Nearly one-fifth of the variance in primary caregiver adjustment was accounted for by risk and resistance factors, with nearly 8% of the variance being accounted for by stress processing (i.e. CSI Disengagement).

The data from this investigation lend some support to the findings of Thompson and associates (9,10,21), who have predicted adjustment in mothers of children with cystic fibrosis and SCD. Specifically, our findings indicate that increased use of disengagement coping techniques were negatively associated with primary caregiver adjustment. Stress processing factors, or coping style, appeared to be more strongly associated with primary caregiver adjustment than the other variables examined. In contrast to previous studies, the contribution of psychosocial stressors (i.e. primary caregiver income and life events) did not reach significance. One possibility for this inconsistency may be that previous studies have examined only daily stressors, while our investigation focused on life events that may have been less specific, thus producing little variance within the sample.

Our findings suggest that a cognitive processing style characterized by children's endorsements of internal beliefs about expectations of control over health were associated with children's adjustment as rated by primary caregivers. Although internal beliefs about expectations of control over health were conceptualized as an intrapersonal resistance factor, it is likely that the belief that children may exert control over their health is a variant of adaptive (i.e. problem-focused) coping. Thus, it may be that primary caregivers' coping and children's locus of control both serve as resistance factors in one's adaptation in the context of SCD.

In the child portion of the model, several interesting trends were revealed that suggest the role of risk factors, including functional risk factors (i.e. adaptive behavior and self-esteem), in predicting child adjustment as rated by caregivers. The small sample size may have precluded statistical significance by limiting the power to detect relationships. Future collaborative studies will need to focus on the role of risk factors, including functional impairments and competence, in predicting adjustment and adaptation to disease to determine the veracity of this association.

Similar to the findings of Thompson et al. (9,10), our data do not provide support for an association between family functioning and caregiver adjustment. In fact, family cohesion accounted for less than 4% of the variance in primary caregiver and child adjustment, respectively, a finding identical to that of Thompson et al. (9). In contrast, Kell et al. (14) provided data that greater family competence was associated with few adjustment difficulties in adolescents with SCD. One possibility for the discrepancy in findings may be due to the fact that Kell et al. administered only the Health Competence scale, and the family measures in our investigation assessed general dimensions of family functioning, including adaptability and cohesion, perceptions of formal and informal network supports, and adequacy of family resource support. Another explanation for these findings may be the cultural issues that are unique to African–American families. Family functioning may be conceptualized differently in lower socioeconomic African–American samples than in middle-class, White families. Primary caregivers in this investigation may have had a different notion of family than would be expected in a sample comprised of middle-class, White respondents. For example, Wilson and Tolson (59) suggested that African–American families seek support beyond their nuclear families, to include cousins, grandparents, and friends. The measures used in this investigation that assessed family functioning may not have captured the true notion of family as experienced by these primary caregivers. Future research should use family measures that are culturally sensitive and that address environments characterized by extended families.

Consistent with the majority of the pediatric chronic illness literature, disease risk factors (i.e. disease severity) in this particular investigation predicted neither primary caregivers' nor children's adjustment. These findings parallel our data of children with congenital heart defects, where children's cardiac defects accounted for less than 2% of the variance in primary caregiver adjustment (58). Similarly, Thompson et al. (10,21) found that adjustment of mothers of children with SCD and cystic fibrosis was not related to disease severity.

In addition to the direct relationships independently predicting primary caregiver and child adjustment, we also were interested in examining the association between primary caregiver coping and child adaptation. Primary caregiver adjustment was associated with children's adjustment. It must be noted that caregivers provided proxy responses for children's ratings of adjustment. This introduces some bias, since caregivers rated both their own adjustment as well as their children's adjustment, thereby failing to control for interrater bias. To examine this hypothesis of the role of primary caregiver coping in predicting children's adjustment, we tested Baron and Kenny's (56) mediational model. An indirect effect of primary caregiver coping on child adjustment was found through influence on primary caregiver adjustment. These data must be interpreted judiciously because the conceptual nature of this investigation precludes a definitive statement regarding the direction of mediating effects. Our results suggest that primary caregiver coping is not only involved in primary caregiver adjustment but also has a modest direct influence on children's adaptation to the disease, at least as rated by caregivers to the disease process. Notwithstanding the limitations of caregivers completing ratings of their children's adjustment as well as their own coping, one possibility is that primary caregivers exert a significant role in assisting their children to cope with and adjust to the arduous aspects of the disease process. These data are provocative and warrant replication with larger samples across chronic illness groups, with other raters of children's adjustment, and with longitudinal designs that can determine the specific direction of the relationship.

The results of this investigation must be interpreted in light of the methodological limitations inherent in a clinical study. First, the small number of subjects may have limited statistical power, obscuring significant differences. In addition, the ratio of variables to participants is less than optimal. We recommend that multicenter studies be conducted where data can be accrued across sites. In addition, because of the correlational nature of the design, the specific direction of any possible causal relationships cannot be determined. Moreover, because primary caregivers that rated their own adjustment also rated their own coping styles, the obtained results may, in part, be from measurement error variance. We recommend that, in future studies, data be obtained from multiple sources, including other informants and direct observations of behavior. Further, we were unable to address whether the findings are unique to children with SCD or whether they are generalizable to other chronic illnesses. Research will need to compare the model across various illness groups. Although it is recognized that the CBCL has been criticized for use with chronically ill populations (54), the analyses using the CBCL Internalizing domain were repeated when the somatic subscale was removed and the results did not change.

Our sample of caregivers was quite heterogeneous. Although most were mothers, fathers, grandmothers, aunts, and uncles also were included in the sample. Adjustment in these other caregivers may be very different from adjustment in mothers. The investigation of models associated with adjustment and adaptation must consider all issues of ethnicity and race as they relate to predictor variables. Obviously, investigators must be cognizant of and sensitive to cultural norms, values, and views of ethnic and racial groups. Clearly, the extent to which the many concepts developed from a majority cultural perspective are applicable to diverse ethnic groups is an area that needs further investigation.

The cross-sectional nature of our investigation also limits the conclusions, particularly the causal mechanisms of adjustment to chronic illness in children and their families. Future studies will need to focus on longitudinal designs that elucidate causal mechanisms that predict adjustment to a chronic illness. In addition, there is potential bias of having caregivers complete measures that relate to themselves and their children and subsequently assess the associations among the instruments. Although a corpus of research indicates that parents provide more accurate ratings of their children's behavior than do the children themselves, future research will need to examine associations among caregivers and their children when measures of adjustment are obtained from multiple informants. Finally, the post-hoc selection of scales for entry into the model also may have introduced bias, warranting judicious interpretation.

Despite these limitations, our findings have some implications for delineating important components in intervention packages. For primary caregivers and their children who exhibit problems with adjustment, the use of active, problem-focused strategies in coping with their children's illness may enhance adjustment for the primary caregivers and their children. Further, in therapeutic efforts for children with SCD, it will be important to foster mechanisms for developing internal beliefs about health behaviors and the effect of their behavior on their illness.

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