FAMILY COPING AND HOSPITAL-BASED EXTENDED KINSHIP NETWORK INTERVENTIONS WITH CAREGIVERS OF CHILDREN DIAGNOSED WITH SICKLE CELL DISEASE

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Introduction

The relationship between family functioning and chronic illness has been the basis of ongoing study across academic disciplines for several decades (Gross, 1988; Knafl & Deatrick, 1987; Koch-Hattem, 1987; Shapiro, 1983). As an area of study, its roots stem from the infusion of systems theory into medicine, social work, and psychology, and the parallel emergence of the field of family medicine. Family-focused medical interventions were first approached in a systematic way in 1926 as part of a thirteen-year experimental project in a London hospital. A similar experiment followed in New York City in 1939. A third project was organized by the Montefiore Medical Group from 1950-1959. During this time, a conceptual shift occurred in psychotherapy and medicine with the inclusion of systems theory concepts borrowed from the fields of biology and sociology. This shift involved viewing the family, and not the identified patient, as the treatment unit. By the late 1960's, the practice of family medicine was well established, but the integration of the family unit into the medical care of individual patients has only recently occurred (Doherty & Baird, 1983). Turk and Kerns (1985) believe that family theory is increasingly gaining acceptance in applied fields, but also suggest that it still has not been adequately utilized within behavioral medicine and health psychology.

The physician with a social orientation is concerned with the medical aspects of an illness, the patient's and family's reaction to the illness, and the meaning of the illness within the patient's culture (Doherty & Baird, 1983). However, many physicians have traditionally tended to neglect psychological and family-related factors by focusing on the child in the role of patient and sharing with family members (usually the mother) only what is considered to be necessary information (Reiss & Kaplan De-Nour, 1989). As a result, the specific needs of family members with psychological and social problems related to another member's illness are often not well-understood (Piersma, 1985). This problem becomes particularly complex when a family member is chronically ill with a disease that has an intermittent and unpredictable course of crises and remissions (Rolland, 1987).

The interaction between families and chronic illness involves a number of complex interrelationships among the ill child and family, family members, spouses, siblings, and health-service providers. Many studies indicate that who is ill in the family determines the nature of the overall family response (Hadley, Jacob, Miliones, Caplan, & Spitz, 1974; Worby, 1971). Highlighting the need to study family coping from a social systems perspective, increasing attention has been paid to siblings (Grossman, 1972; Seligman, 1983; Wasserman, 1983), fathers (Lamb, 1983), and grandparents (Sonnek, 1986). However, many studies only explore the perceptions and experiences of the mothers, because maternal adjustment is one of the more important factors in

overall family functioning within the family system (Brown, Kaslow, Doepke, Buchanan, Eckman, Baldwin, & Goonan, 1993). The mother's emotional and psychological status has the strongest influence on the health of the chronically ill child (Brown, et al., 1993; Gil, 1991; Jessop, Reissman, & Stein, 1988) and the family system in general.

The physical and psychological dimensions of the family's environment closely relate to health and disease outcomes in children. (Shapiro, 1983). Minuchin (1974) articulates the complex nature of the interplay between the dysfunctional dynamics within the family system and the presentation of symptoms in an ill child. Chronically ill children from higher functioning families do better than those in more dysfunctional families (Pless & Satterwhite, 1973). In more severely dysfunctional families, the sick child often suffers the consequence of not getting basic or medical needs met. Healthy family functioning in the face of chronic illness is defined by McCubbin and Patterson (1982, 1983) as the family's ability to provide for the emotional and developmental needs of all family members, integrating medical management of the disease into the family's ongoing routines, and raising the ill child to be autonomous, self-accepting, and achievement-oriented. These researchers point to the importance of examining family variables related to treatment, illness outcome, and overall coping with chronic illness, and support the idea that if the family system can influence both the course and outcome of an illness, then understanding how families cope with illness and exploring ways to facilitate coping becomes an important task.

Coping tasks are determined by demands placed on the family due to the specific aspects of an illness. Thus, it is important to keep characteristics of the illness in mind when discussing family coping. Chronic illnesses, such as sickle cell disease (SCD), demand that the family is able to be both flexible and stable, which allows them to handle repeated crises over a long period of time. Another skill needed by families in this situation is that the members can easily share information with each other about the illness (Treadwell, Jackson, Antonaidis, Samuel, Holladay & Vichinsky, 1994), so how well they communicate is also important. A fourth consideration is how resourceful the family can be in terms of asking for help and social support in crisis situations, since this has been found to help buffer the stress experienced by the family (Cohen &Wills, 1985). Each of these factors (illness characteristics; family dynamics including adaptability, cohesion and communication; coping; social support; and stress), which contribute to families coping successfully with a chronically ill child will be discussed in more detail in the following sections.

Sickle Cell Disease

Until fifteen years ago, undiagnosed sickle cell disease in children under age two was associated with a 25% mortality rate in this country. Since that time, studies have shown that a reduction in the incidence of infection, as well as a dramatic reduction in death due to infection and splenic sequestration, are a direct result of early disease identification and family-based intervention (Gaston, et al., 1986; National Institute of Health, 1987). In 1987 in the state of California, cord-blood screening was practiced on a trial basis to test for sickle cell trait and disease on selected babies. In 1988, there were 22 states regularly practicing newborn screening for trait and disease. By 1990, 41 states and two territories had established regulations mandating that all infants be screened for sickle cell disease. As a part of this process, a standard newborn screening protocol was established in the state of California in 1990. Newborn screening has since resulted in a greatly reduced rate of mortality in the first five years of life for children with sickle cell disease.

Intervening with the family at the time of screening and diagnosis is hypothesized to be crucial for the development of active coping strategies that will serve the child and family in managing the physical and psychological symptoms created by the disease (Treadwell, Fortune Pinhiero & Lessing, in press). Although this has been clinically demonstrated, it has not been empirically investigated. For this reason, the types of interventions made with families who have been identified through screening are deserving of critical attention.

Sickle cell disease is a genetically transmitted blood disorder. A protein inside of the red blood cell, called hemoglobin, is altered by the disease so that it is synthesized in a manner that is different from that of healthy individuals. Persons with sickle cell disease, or sickling disease, have inherited one of several types of hemoglobin that cause red blood cells to change into a sickled shape. Because sickled-shaped red blood cells cannot flow easily through the circulatory system, they can create numerous medical problems. Characteristic symptoms may include periodic unpredictable musculo-skeletal pain, delays in physical and sexual maturation, acute and chronic pulmonary problems, aseptic necrosis of the hips and shoulders, and, in some cases, strokes (State of California Department of Health Services, 1992; Vichinsky, Johnson, & Lubin, 1982). The highest mortality rate occurs within the first five years of life and is typically due to infection. The next highest rate occurs between ages 20 - 24 and is typically due to multiple organ failures (Serjeant, 1985; Thompson, Gil, Abrams, & Phillips, 1992). The course of the illness is highly unpredictable and can involve multiple complications. Research on remedial treatments such as gene replacement procedures and bone marrow transplantation with chemotherapy is still in the early stages, but there is still no cure for the disease. Ongoing advances in palliative therapies involving the management and treatment of acute and chronic complications

have, however, resulted in more favorable long-term prognosis (Charache, Lubin, & Reid, 1985).

There are multiple variations of hemoglobin types associated with sickle cell disease. The two most common types of sickle cell disease are Sickle Cell Anemia (SS) and Sickle C disease (SC). Less common disease types are Sickle Beta Thalassemia disease (SB Thal) and Hemoglobin SD disease (SD). Each of these hemoglobin diseases has a distinct type of defect in the genetic code for hemoglobin synthesis that contributes to their clinical picture.

There is a high incidence of sickle cell disease found in populations inhabiting areas where falciparum malaria is or was endemic, such as Africa, the Middle East, the Mediterranean and Southern India. A selective advantage exists within the populations of these regions for persons having hemoglobin A, which is found in all healthy individuals, in combination with hemoglobin S, the type of hemoglobin associated with sickle cell disease. It is believed that persons with the gene for hemoglobin S were better able to to survive malarial infection, leading to a much higher number of gene carriers for the hemoglobin type that causes sickle cell disease in certain regions. Today, this is also true for anyone whose descendants come from these regions. In the United States, 1 in 10 African Americans carry the gene for sickle cell disease and 1 in 500 actually have the disease (State of California Department of Health Services, 1992).

The medical regimen that must be followed by a sickle cell patient can be very demanding and involves regular visits to a doctor, extra immunizations, prophylactic penicillin treatments, a host of blood and urine tests, kidney and liver function tests, and x-rays. Caregivers caring for a child with sickle cell disease have to learn to monitor the child's temperature, fluid intake, increased nutritional needs, and physical activity on a daily basis. When the child experiences painful episodes, the caregiver must be able to assess when it is appropriate to contact a doctor, and when home management of the illness will suffice (State of California Department of Health Services, 1992). Even under optimal circumstances, when all appropriate precautions have been taken, the child is subject to unpredictable episodes of severe pain or complications that can require extended periods of hospitalization. These caregivers need information, training and outside support to successfully manage their child's care (Lessing & Vichinsky, 1990). Helping caregivers to identify and develop a network of support is believed to be critical to the child, caregiver, and family in learning to live with a chronic illness. Although hospital-based sickle cell programs offer the most appropriate means for providing these resources to caregivers, they do so with varying degrees of success.

Three concepts from family systems theory identified by Doherty & Baird (1983) as crucial to understanding and treating families in a medical context are: (1) interactional patterns, (2) adaptability, and (3) cohesion. An effective and comprehensive model for understanding family functioning that systematically examines the factors of communication, adaptability, and cohesion, is the Circumplex model. It also provides the theoretical framework for a widely used family assessment tool. Accordingly, there are many other theoretical models that rely on concepts closely resembling these dimensions (Beavers & Voeller, 1983; Epstein, Bishop & Levin, 1978; Kantor & Lehr, 1975; Olson, Russell & Sprenkle, 1979). The model juxtaposes three dimensions of family behavior (cohesion, adaptability and communication) to identify sixteen types of family systems.

In Figure 1, the left of the center of the model's axis represents families that are separated, or, at its extreme end of the continuum, disengaged; the right of the center of the axis represents families that are connected, or, at its extreme, enmeshed. The top half of the circle represents the flexible family, and, to its extreme, disengagement; the bottom half represents the more structured family, and, to its extreme, rigidity. The communication dimension is represented by the three concentric bands within the circle which are distinguished by different linear patterns. The center circle depicts healthy (open) styles of communication within the family system, while the middle and outer bands show declining degrees of dysfunctional family communication (random or closed) styles. Combined with the adaptability dimension, the upper half illustrates random communication and the lower half illustrates closed communication.

Cohesion refers to "the degree of emotional bonding members have with one another and the degree of individual autonomy a person experiences in the family system" (Olson et al., p. 5). It is quantitatively assessed at four areas along a continuum: (1) Disengaged (low), (2) separated, (3) connected, and (4) enmeshed (high). Barbarin, Hughes, and Chesler (1985) discuss how cohesiveness serves a positive function in families with ill children. Adaptability is defined as "the ability of a marital/family system to change its power structure, role relationships, and relationship rules in response to situational and developmental stress" (Olson et al., 1983, p. 70). Adaptability is also quantitatively assessed at four areas along a continuum: (1) Rigid (low), (2) structured, (3) flexible, and (4) chaotic (high). Kazak (1989) describes how lower levels of adaptability can actually contribute to more effective functioning in families where the treatment for the illness involves a dietary regimen. Olson, et al. (1983) later added a third dimension, the communication dimension, to the Circumplex model. This dimension functions as a facilitative dimension and is an indirect indicator of how a family facilitates or restricts balanced functioning on the other two dimensions.

The model hypothesizes that the central areas on the continuum for the cohesion and adaptability dimensions indicate optimal family functioning, while the areas at the extremes of the two continuums describe dysfunctional states. On the cohesion dimension, too much closeness leads to an enmeshed system and too little closeness leads to a system that is disengaged. On the adaptability dimension, too much change will lead to a chaotic system while too little change leads to a system that is rigid and inflexible. For example, disengaged families of chronically ill children would be characterized by underinvolvement demonstrated through denial of or withdrawal from the illness, or by non-inclusion of the ill child in family activities. Enmeshed families could be characterized by overprotectiveness or overinvolvement, demonstrated by high anxiety around letting the sick child participate in activities that offer healthy challenges to their development. Rigid families could be characterized by instability demonstrated by a conflict between maintaining strict roles and the need for change in new or evolving circumstances. Chaotic families could be characterized by a lack of rules and roles demonstrated by excessive and unsuccessful negotiation around conflict. Families who have difficulty communicating would be characterized by limited emotional expression shown by members interacting with one another in non-supportive ways such as using criticism or double messages.

Cohesion and adaptability are repeatedly identified in the literature on family adaptation to medical stressors as two of the most prominent family resources that positively relate to adaptation (Olsen et al., 1979, 1983; Seligman & Darling, 1989; Stinnet & Sauer, 1977; Turnbull, Summers & Brotherson, 1986). As a measure of healthy family functioning, cohesion correlates with personal adjustment, social adjustment, and positive self-esteem in the child (Kumar, Powers, & Haywood, 1976; Wells, Nash, McMillan, Hails, & Lauria, 1986). Thus, the Circumplex model can be used to effectively study family dynamics involving communication, flexibility, and closeness, in families of chronically ill children. As outlined by the model, the extent to which a family can easily share feelings increases the likelihood that it can maintain a balanced degree of flexibility and connectedness. "In the face of a severe chronic illness situation, balanced families indicate more positive health outcomes for the chronically ill child" (McCubbin, 1986, p. 6). The balanced family is more able to adjust to change that is necessitated by crises while at the same time continuing to meet the needs of its individual members.

Coping

The burden of chronic care of an ill child places numerous additional demands on the roles of all family members. More facilitation of independence and less pampering is required of mothers, more psychological and instrumental support is required of

fathers, and more caretaking is required of siblings (Seligman & Darling, 1989). Over time, the burden experienced by individual family members can become compounded as the stressors pile up. This is a critical time for the family that draws upon their ability to successfully communicate, problem-solve, and ultimately adapt to their unpredictable situation.

Defining and quantifying the elusive profile of behaviors and characteristics that constitute successful coping has been attempted with varying degrees of success (Shapiro, 1986; Thiots, 1986). The application of a family systems approach to the understanding of coping with the stress of a chronic illness is even more of a challenge. Many studies focus on the adjustment of the individual child (Gil, Williams, Thompson & Kinney, 1991) or the child and mother (Brown, et al., 1993; Crnic, Friedrich, & Greenberg, 1983). Shapiro (1986) points out that coping is not an individual response despite the fact that it is primarily studied in that way, but rather an interactive process occurring between family, extended family, friends, neighbors, colleagues, and professional helpers. Accordingly, the family's perception or interpretation of an illness crisis can impact the illness itself. "If the family interprets the illness as a threat, the crisis will produce anxiety; if it is interpreted as a loss, it will produce depression; and if it is interpreted as a challenge, both anxiety and hope will create problem-solving energy and promote motivation and growth within the family" (Shapiro, 1983, p. 915). So, the meaning a family attaches to difficult situations is another important aspect that contributes to their overall coping strategy. Since families have to maintain and manage several dimensions of family life simultaneously, assessment of multiple tasks and functions is required to fully assess family coping. Thus, identifying how family members communicate, seek help and make decisions during a crisis, and interpret the meaning of that crisis, is relevant to how well individual family members will cope with that crisis.

To better understand this complex process, it is important to examine coping theory. Thoits (1984) draws a parallel between the functions of support and coping by describing how the same methods of offering assistance to others are also used by some individuals in responding to stressors. Three commonly used categories of support also used to describe approaches to coping are: Problem-focused coping, emotion-focused coping, and perception-focused coping. Problem-focused involves changing environmental circumstances, emotion-focused involves directing thoughts and actions to control feelings, and perception-focused involves changing the meaning attributed to a stressful situation. Thoits defines effective support as support that matches the needs and values of the distressed person. Thus, Thoits adds to the stress-buffering literature by suggesting that helpers who are socioculturally or experientially similar are more likely to be perceived as empathic, and responded to by the distressed person. By virtue of being more responsive to the offered support, they are coping

more effectively. This viewpoint is particularly relevant to the effectiveness of support-seeking in African American extended families.

Lazarus (1974) distinguishes between stressful situations and stress reactions to stressful situations. He views coping as serving two functions: (1) problem solving and (2) regulation of emotional distress. Accordingly, he identifies two forms of coping: Palliative and problem-solving. Coping by problem-solving involves direct action on the stressor, and palliative forms involve working at the cognitive or intrapsychic level. Lazarus sees the appraisal of a threatening event as ultimately more powerful than the event itself. Both palliative and active problem-solving methods have been shown to be effective coping strategies on the part of families with a chronically ill child (Treadwell, et al., 1994).

Knafl and Deatrick (1987) make a distinction between active and passive family coping styles based on the underlying assumptions of each: The passive approach assumes the illness will negatively impact the family while the active approach assumes the family will continually adjust their perceptions of the illness in functional ways. Active coping styles have been identified as reducing emotional symptoms in children with chronic illness (Dracopoulos & Weatherly, 1983; Gross, 1988; Sargeant & Leibman, 1985; Treadwell, 1989). These coping behaviors, in which family members actively seek out needed information, draw on past experiences, consider multiple alternatives for handling problems, and take positive action, match coping behavior as described by Lazarus' problem-solving function.

Shapiro (1983) presents a model for understanding coping strategies that is based on control theory (she posits control to be a fundamental human drive) and attempts to incorporate concepts from several models of coping. Issues of control are particularly relevant to the experience of illness. Having a sense of mastery over the course/outcome of a disease is very difficult and raises issues of competence, vulnerability, fragility, dependence, and sacrifice. Shapiro's model has two dimensions of control: Active vs. Passive and Adaptive vs. Dysfunctional. These dimensions make up four quadrants: (1) Active-positive, (2) Passive-positive, (3) Active-negative, and (4) Passive-negative. Active-positive coping strategies would include seeking information, directly acting on the illness, choosing not to act on the illness, mastery of procedures related to managing the illness, and addressing the quality of life for the ill person and the family. Active-negative strategies would include dysfunctional denial and/or avoidance, excessive hypervigilance, excessive accumulation of information on the disease, and intellectualization. Passive-positive strategies would include acceptance, utilizing support available from others and finding meaning from the illness experience. Passive-negative strategies involve taking on the role of a helpless, hopeless victim, pervasive dependency, depression, low self-esteem, and generally giving up.

Using Shapiro's definitions, different stages in the course of an illness will necessitate either or both active and passive strategies. For example, in the early stages of adjustment to a diagnosis, more direct and active strategies would be appropriate, but later in the course of the illness, passive strategies such as using distraction as a way of tolerating a pain episode might be what is needed. Families have been observed to go through stages of coping with chronic illness that closely parallel the classic stages of death and dying (Power & Dell Orto, 1980). The beginning stage, usually when a diagnosis has been made, is where both adaptive and maladaptive coping responses become evident. These early responses will be persistent throughout the course of an illness which supports the notion of enhancing active and passive coping strategies during this time (Treadwell et al., in press).

Treadwell et al. (1994) found that caregivers who scored high on a coping measure in terms of their ability to reframe problems in more positive ways, and to actively problem-solve, also reported better family functioning. These findings are consistent with Thoits' theory of problem, emotion, and perception-focused coping functions, Lazarus' theory of palliative vs. problem solving, Shapiro's theory of active-positive vs. passive-negative coping, and McCubbin and Patterson's theory involving family resources and meaning perception. However, in contrast to Treadwell's model, Lazarus and Shapiro do not directly address the role and function of a social support network and its impact on the overall coping of the family. Thoits acknowledges the overlapping functions of coping and effective support, but does not discuss the development of a network in terms of coping behavior. This aspect of coping behavior becomes particularly relevant when the focus is on caregiver coping since the support network typically revolves around the caregiver. Treadwell et al. (1994) also found that efforts on the part of the caregiver to educate and involve extended family in the care of the child with SCD, positively affected functioning for the caregiver and family. This research helps to substantiate what are otherwise theoretical assumptions about successful coping behaviors in families of children with SCD such as actively seeking help and support from others, active problem-solving, and altering the perceived meaning of a stressor.

Stress

Efforts to integrate concepts from family systems theory with knowledge of families with chronically ill children have resulted in significant contributions in the area of how stress affects family functioning (Crnic et al., 1983; McCubbin & Patterson, 1983; Wikler, 1981). The original theoretical model of family stress was developed by Hill (1949). It was later elaborated on and designated as the ABC-X family crisis model by McCubbin and Patterson (1983). The strength of this model is

in its applicability to the study of family coping within an interactional framework by combining the effects of family stress, illness, social support, and functioning.

In Hill's model, the stressor event (A) interacts with the family's coping resources (B) which then interacts with the meaning given to the event by the family (C); and in combination, these components produce the crisis experience (X). Factor A, the stressor, is an event or a transitional period that generates change in one or more parts of the family system, such as roles, interactional patterns, boundaries, beliefs or values. Factor B, the family's coping resources, refers to the family's ability to prevent a crisis from occurring within the system. This factor involves flexibility, the ability to shift or adjust in healthy ways, and is usually determined by the family's functional status prior to the stressor event. Factor C, the meaning attributed to the stressor event by the family, comes out of the family's previous experiences with handling crisis events.

Figure 2 diagrams the Double ABC-X model (McCubbin & Patterson,1983) that is originally based on Hill's model and elaborates on their own ABC-X model with the addition of a two-phase component. In this model, the first phase of response to a crisis or catastrophe is referred to as the adjustment phase, followed by an adaptation phase. McCubbin and Patterson hypothesize that a number of factors interact to jointly influence the outcome of a family coping with an initial stressor such as the diagnosis of an hereditary disease in a child, and their subsequent adaptation to ongoing associated stressors (Koch, 1985; McCubbin & Patterson, 1983; 1982).

In support of using the ABC-X model for studying families with chronically ill children, Cherry (1989) points out that having knowledge about individual stress, family stress, and coping responses, lays the groundwork for being able to intervene with the child or parent in order to facilitate adaptive coping on the part of the family. In a similar vein, Thompson (1993) found that daily stresses, apart from those related to caring for an ill child, related to emotional distress and family dysfunction in mothers of chronically ill children. Using a model of transactional stress and coping, Thompson was able to determine that daily stress contributed 13% more variance to psychological distress in mothers of children with SCD. Figure 3 diagrams Treadwell and Gil's (1994) model of stress and adaptation which was adapted from the ABC-X model. It illustrates the interrelationships among the family's resources, disease characteristics, adaptational factors, and outcomes, and shows how, at any given stage of development, adaptational factors can be mediated by a number of variables, including stress. These mediating variables impact how the family defines the severity of the crisis and the manageability of its demands. This model, tested by Treadwell-Gad, Holladay, Lessing, Fortune Pinheiro, and Vichinsky (1991), was able to demonstrate how psychological, social and behavioral factors impact the initial phase of adjustment to chronic illness in families of infants and young children diagnosed with sickle cell disease.

Social Support

The integration of family systems theory with the literature on family coping with chronically ill children has led to significant contributions on the question of how social support affects adjustment and adaptation of individuals and families (Crnic et al., 1983; Kazak & Marvin, 1984; Kazak & Wilcox, 1984; Taylor, Chatters, & Mayo, 1988; Varni, Wilcox & Hanson, 1988). Social support is consistently identified in the literature as a significant moderating variable that impacts life stress (Cobb, 1976; Cohen & Hoberman, 1983; Cohen & Wills (1985). Cobb (1976) describes how social support facilitates coping with crisis and adaptation to change in that it serves a protective function by mediating the effects of stress over a variety of lifespan transitions. He found that women who had a confidant (close, intimate, confiding relationship) were 10 times less likely to be depressed. House (1981) provides further support for this in describing how social support provides instrumental aid, socioemotional aid, and informational aid to distressed persons.

Social support is viewed by family clinicians as a critical component of emotional well-being, especially during stressful periods that involve major life transitions and other situational and personal crises (McCubbin & Boss, 1980; Turner, 1981). Support from spouses, relatives, and friends is typically found to correlate with higher levels of psychological and social adjustment (Kazak & Marvin, 1984). Seligman and Darling (1989) discuss the importance of including an assessment of the family's social support network in any model of family functioning in order to gain a precise understanding of the types of stresses impacting the family system, to get a picture of the nature and degree of isolation they may be experiencing, and to explore alternative forms of support for families who are unable to utilize more formalized support services.

There is wide variability in definitions of social support found in the literature (Cooke, Rossmann, McCubbin, & Patterson, 1988). Attempts to operationalize social support have been as simple as a verbatim tally of the number of people in a social network, and as complex as using theoretically derived dimensions. Cooke et al. (1988) state that "the assessment of social support should include determination of as many as is possible of the aspects of the various kinds of social support as well as the many potential sources of social support" (p. 212). Support factors often used as measurement criteria are structure (the number of relationships in a support network), function (the type of resource provided by each relationship), support timing (critical point during a stressful event), informal (i.e., relatives, friends, or neighbors), formal support (i.e., medical professionals or social service workers), density (amount and intensity of contact between support group members), marital status, support group

size, and stress (degree of emotional distress experienced) (Cobb, 1976; Cohen & Hoberman, 1983; Cohen & Wills, 1985; Jacobson, 1986; Kaplan, 1975; McCubbin & Patterson, 1983; Mitchell, 1969; Schumaker & Brownell, 1984; Thoits, 1985). However, it can be misleading to use criteria identified only by the investigator. Murawski, Penman, and Schmitt (1978) suggest that a more meaningful measure should involve assessing the quality of social support as perceived by the caregiver, and employs the sources of support identified as helpful by the subject. House (1981) agrees with this idea, adding that,"...an instrument that measures the perceptions of the respondents is most useful since (social support) is likely to be effective only to the extent that it is perceived" (p. 27).

Much of the literature discusses social support in the context of two models of support: the direct-effect and the buffering-effect models (Cohen & Wills, 1985). The direct-effect (or main effect) model postulates that social support has an overall beneficial effect of support regardless of the presence of stressors. The latter model describes a complex process whereby social support positively affects well being by protecting (or buffering) a person or family from adverse stressors. This idea, referred to as the buffering hypothesis, postulates that without the presence of stressors, social support would not particularly impact well-being. Cohen and Wills (1985) conducted a comparative review of studies supporting both models and found that when the social support measure includes an assessment of perceived availability of supportive resources, evidence for the buffering model is provided. A support enhancement study on families of children with SCD (Treadwell et al., in press) incorporated a variety of support constructs mentioned above, such as support structure and function; timing; informal and formal support; density; group size; and stress. It also utilized assessments of support that were based on the caregiver's identification of support types and perceptions of support quality. This study provided information on how social supports can be enhanced for caregivers and offered a more effective means of assessing support networks.

Another extremely useful model for studying family support networks is Bronfenbrenner's Social Ecology model (1979). This model allows for the examination of families within the context of four overlapping systems, and describes how they are viewed as a system nested within a number of other societal systems. The Microsystem refers to the nuclear family; the Mesosystem refers to the extended family and community supports; the Exosystem refers to the social, legal, and educational systems; and the Macrosystem refers to social belief systems that the family function within. Thus, the main premise of the Social Ecology Model is that the family can be understood only when looking at the internal dynamics within the nuclear family unit in conjunction with the larger context of the family's social reality.

The Social Ecology model is exceptionally appropriate when applied to the study of African American families of children with SCD. The Microsystem and

Mesosystem refer to the structure, make-up and function of the extended support network which is known to significantly influence caregiver functioning. The Mesosystem refers to resources available to a family that facilitate effective interfacing with the larger society. This would include medical and other professional service providers. The Macrosystem refers to how the family attributes meaning to situations and events, such as a child being diagnosed with SCD, which shapes the experiential quality of their daily living. Each of these levels provides a categorical structure for defining extended networks and for identifying key points of intervention that could effectively impact the network of families with children who have SCD.

Extended Kinship Networks

In adjusting to the stress of caring for a chronically ill child, many families create support networks among extended family members (Travis, 1976). Contact with an outside social support system is a factor that can influence and predict the well-being of the household network members (Mitchell, 1969). Anderson (1982) found that households that have little contact with an outside social support system are more psychologically troubled. Fortunately, this is an atypical household in an African American community. African Americans characteristically tend to be part of extended families: families with ties that often extend beyond the boundaries of the nuclear unit to include other households of blood and non-blood relatives (Billingsley, 1968; Nobles, 1981).

The classical definition of extended family, borne out of the slavery period, involves biologically related kin who live geographically close to one another. During the post-slavery era, migration to the north after emancipation into larger and more industrial cities led to changes in the expression of extended family life but not in the psychological need for a kinship network (Martin & Martin, 1987). This new expression was a modified version of the classical extended family which could include persons who were blood relations, friends, neighbors, or church members. Thus, the current-day extended family could be more appropriately termed extended kinship network.

These kinship networks, often based on the formation of "fictive kinships", are an important coping strategy that continues to persist across socioeconomic classes (McAdoo, 1980). Fictive kinship refers to a type of relationship brought about by social or economic circumstances between individuals who are not related by blood or marriage (Penn et al., 1993). "Extended kinship network" is a term that has been adopted for the current study which encompasses the full range of relational possibilities found within the population of families studied. The term borrows heavily from definitions used by various historians, sociologists, anthropologists, and

psychologists over the past few decades in the literature on African American and other traditional ethnic societies (Aschenbrenner, 1973; Billingsley, 1968; Bronfenbrenner, 1979; Cantor, 1979; Castillo, Weisblat & Villareal, 1967 Dressler, 1985; Martin & Martin, 1978; Nobles, 1981; Shimkin, Shimkin, & Frate, 1978; Speck, 1973; Staples, 1976).

Nobles (1985) described extended African American family structures as being "...comprised of several individual households having authority lines that often extend beyond any single household unit, in which the extended family is most visible in periods of crisis, and at times of ceremony, and provides needed emotional support for its members" (p. 27). Billingsley (1968) conceptualized them as a network of interlocking systems, with each family unit being a part of a larger network of mutually interdependent relationships within the community. Similarly, Martin and Martin (1978) describe them as "....a multigenerational, independent kinship system which is welded together by a sense of obligation to relatives; is organized around a 'family base' household; is generally guided by a dominant family network; and has a built-in mutual aid system for the welfare of its members and the maintenance of the family as a whole" (p.1). This author employed a combination of the above definitions to describe the network of individuals connected to the caregiver: The extended kinship network is a group of people identified by the caregiver who are either a part of the household, identified as part of the caregiver's family, have learned about SCD from the caregiver and/or hospital staff, and who provide advice, emotional support, social support, and instrumental support to the caregiver.

In referring to the high levels of cooperation and sharing in black families, Aschenbrenner (1975) suggests that the extended kin network in part helps to define what makes African American families culturally unique. Since we know that African American families typically find support within the extended kinship network, it would be logical to assume that African American families with children who have SCD will look to their extended networks for support. It is also possible that the larger the network, the more it could potentially provide support to caregivers. Kazak, Reber and Carter (1988) found that larger, less dense networks were related to decreased experiences of psychological distress in parents with and without an ill child. This finding of a relationship between social network characteristics and psychological distress points to the importance of the extended kinship network as a family coping resource for African American caregivers of children with SCD.

Summary

In summary, a relationship exists between family functioning and chronic illness, and the study of this relationship is rooted in the beginnings of family medical practice. Yet, hospital-based programs and services fall short in

understanding and addressing the psychological reactions to illness in patients and their families. There is a complicated interrelationship between all members of a family when a child is ill, but the mother has the most critical role in affecting the overall well-being of the family. Since the family environment is an important factor in health and disease outcomes for children, then exploring ways to facilitate family coping, by focusing on caregiver coping, becomes an important task.

The most important coping tasks that need to be mastered by families of chronically ill children involve their: (1) ability to understand and manage the characteristics of the illness (2) degree of stability and flexibility, (3) ability to communicate, (4) ability to employ active problem-solving strategies, and (5) ability to buffer stress by seeking support and assistance from others. The following paragraphs present these tasks in more detail.

The statewide mandate for newborn screening of SCD resulted in greatly reduced mortality rates during the first five years of life. This allowed for early intervention with the caregivers and families in order to assist them in successfully coping with the disease. SCD is a genetically transmitted disease affecting 1 in 500 persons. By altering a person's hemoglobin type, SCD can cause an array of medical problems which can be life-threatening. Caregivers attempting to manage their child's disease symptoms often need assistance in the form of information about the disease, and need training, particularly in terms of learning how to utilize available resources and seek outside support. Hospital-based sickle cell programs do not consistently provide this type of assistance to caregivers.

Family characteristics important to the study of family functioning are stability (cohesion) and flexibility (adaptability). Cohesion, adaptability, and communication are factors represented in the Circumplex model that can be used to describe family dynamics on a continuum of least desirable to optimal functioning. Ideal functioning is attributed to families who can maintain a balanced degree of flexibility and connectedness. This allows them to adjust to change during crisis situations while also addressing the needs of individual members.

Coping theories present several factors that are important to understanding family coping behavior: the ability to (1) change circumstances, (2) manage negative emotions, (3) alter meaning attributed to a stressor, (4) utilize available support from others, and (5) actively problem-solve. These factors are also presented in theories of family stress and are examined in studies on social support as well. One model that is particularly applicable to the study of family stress in relation to coping, the double ABC-X model, allows us to study the combined effects of stress, illness, social support, and functioning. It also accounts for how the initial (early) phase of adjustment can impact long-term adaptation to a chronic stressor. An adapted version of this model, developed for use with families of

children with SCD, helped to demonstrate how adaptational factors can be compromised with these families in the early stages of the illness by variables such as stress. Bronfenbrenner's Social Ecology model is also exceptionally appropriate for studying this group of families, in that it provides the categorical structures to talk about potential points of intervention with family networks.

According to both family stress models, social support is identified as a significant moderating variable that mediates the effects of stress over a variety of lifespan transitions. It is therefore important to include an assessment of a family's social support network in order to clearly understand the nature of their stressors, their degree of isolation, and their specific support needs that are not being addressed. More meaningful measures of family support involve assessing quality of support and identifying sources of support as perceived by the caregivers. When support measures include an assessment of perceived availability of supportive resources, the buffering hypothesis can be effectively demonstrated. This hypothesis suggests that social support serves as a buffer by protecting the family from adverse stressors,

A final important factor in successfully adjusting to the stress of caring for a chronically ill child is the creation of support networks which help insure the emotional well-being of family members. Intact extended kinship networks, often including "fictive kin", are a unique cultural feature of African American families that, in part, was borne out of events that occurred during and after the slavery period in the U.S. These networks can serve an important coping function, particularly for families of children with SCD. Literature on the relationship between extended network characteristics (size and density) and the degree of psychological distress in parents of chronically ill children point to the usefulness of including network characteristic variables in the study of coping in African American families of children with SCD.

Current Study

The current study is based in family systems theory, and incorporates Bronfenbrenner's Social Ecology model, the circumplex model, and a model of family stress and adaptation. The infusion of family systems theory into medicine, social work and psychology has influenced research in the areas of family stress, social support and family coping. Beginning with Hill's research on family stress, followed by McCubbin and Patterson's work on the ABC-X adaptation model, and finally Treadwell's stress and adaptation model, the theoretical and empirical groundwork was laid to begin to apply these models to special populations. Brown, Doepke and Kaslow (1993); Burlew, Evans and Oler (1989); Gil, Williams, Thompson and Kinney (1991);

Thompson, Merritt, Keith, Murphy and Johndrow (1993); Treadwell, Fortune Pinhiero and Lessing (in press), among others, have focused on different variables within these models in looking at aspects of family coping in African American families of children with SCD.

The current study attempted to build on this area of research by contributing to the literature in the following ways: (1) It replicated a study of family functioning in African American families of children with SCD during the early phases of the course of the child's illness. (2) It further tested an adapted model of family stress and adaptation and its applicability to this special population. (3) It tested a hospital-based intervention designed to develop more active coping strategies in the caregiver and to enhance the involvement of the extended network. No systematic assessments of screening protocol interventions of this kind had been done. (4) It examined size and type characteristics of "extended kinship networks", a unique cultural feature of African American families, and their impact on caregiver and family functioning in families of children who have SCD.

The intervention being tested was designed to teach active coping strategies to the caregiver and to enhance the amount and quality of support received by the caregiver from her extended kinship network. The intervention was made on families of children who received a positive diagnosis for sickle cell disease from blood screening procedures between January, 1988 and December, 1991.

The replication and model testing in this study involved the relationship between caregiver resources, life-stress, support, and active coping, and their combined impact on family functioning outcomes. Caregiver resources included level of education and income; family stress was determined by the effect of a variety of life stressors on the family over the previous year, and interview items on perceived caregiver and family burden; support was assessed in terms of perceived amount and type of support available to the family and caregiver; family coping strategies used in crisis situations were considered in both cognitive and behavioral terms; and family functioning was determined by the caregiver's degree of satisfaction with family dynamics, and a profile of interview items concerning the caregiver's satisfaction with support, perception of their own level of coping, and perception of the family's level of coping. All interview items are shown in Appendix L.

This study borrows from the Circumplex model in that it incorporated the constructs of adaptability, cohesion, and communication in order to assess family functioning through the use of the Family Adaptation and Cohesion Scales III. It borrows from McCubbin and Patterson (1983) and Treadwell et al.'s (in press) models of family stress and adaptation by utilizing coping factors such as family resources, meaning perception, disease characteristics, freedom from stress, social supports and active coping. Meaning perception and active coping, identified through the use of the Family-Crisis Oriented Personal Evaluation Scale (McCubbin & Patterson, 1983) are

factors used to determine the family's cognitive and behavioral problem-solving strategies used during the first few years following the child's diagnosis. Since the more meaningful measures of support were found to be those based on the perceptions of the respondent (House, 1981; Murawski et al., 1978), social support was measured by using the caregiver's perceptions of support, based on Caregiver Interview items (Appendix M) and the Family Support Scale (Dunst et al., 1984). Family resources and disease characteristics were statistically controlled. Stress was measured by using the Family Inventory of Life Events (McCubbin & Patterson, 1980) and defined according to Cobb's (1976) and Cohen and Will's (1985) buffering-effect definition of support.

The question of what factors most impact family coping was addressed in this study. However, the main focus was on a method of intervening that best facilitated the caregiver's ability to cope. The predominance of single-parent families in the population of families with children who have sickle cell disease gives special importance to the caregiver's impact on the family (Brown et. al.; 1993; Gaston & Roose, 1982). And healthier levels of family functioning increase compliance with recommended treatments and improve medical outcomes for chronically ill children (Czajkowski & Koocher, 1987; Patterson, 1985). Because it has been found that overall functioning of the family is improved by increasing the caregiver's ability to cope (Jessop et al., 1988), the current study's definition of healthy family functioning includes, among other variables, several caregiver related variables: perception of own level of coping, caregiver perception of family coping, caregiver perception of burden on self and the family, caregiver ability to acquire support, and caregiver satisfaction with family dynamics. Additionally, the focus of the intervention examined by this study was to assist the caregiver in taking on the challenge of caring for a chronically ill child.

Cross-cultural research on family coping with chronic illness emphasizes the importance of developing intervention approaches that incorporate cultural considerations (Shapiro, 1983). The majority of infants born with a diagnosis of Sickle Cell disease are of African descent. Since African American families typically rely on the extended family as a coping resource, the extended kinship network was considered to be a crucial component of caregiver coping in the current study.

Extended networks were examined using a variety of relevant support constructs as cited in the literature, such as support specificity and support function (Cohen & Wills, 1985; Schumaker & Brownell, 1984), the effectiveness of support timing (Jacobson, 1986; Thoits, 1985), and the interaction between stress, social support and coping (Cobb, 1976; Cohen & Hoberman, 1983; McCubbin & Patterson, 1983). Particular attention was given to the size (number of people in the group), type (nature of the relationships to the caregiver), and function (the various ways that support is

received from the network), in order to help further clarify coping outcomes in this population. Cultural considerations for families of children with sickle cell disease point to the salience, in this study, of the size and make-up of the extended network as points of intervention with African American families. Likewise, medical considerations for these families suggested the need to distinguish between structural and functional support, since specific types of medical assistance may be required due to the characteristics of the child's illness. Additionally, both formal and informal supports within the extended support network were examined using the Family Support Scale (Appendix L), since the families with children who are ill often rely regularly on other kinds of professional helpers as part of their support network.

The secondary emphasis of this study on the type, size, and function of the extended kinship network was to allow for a closer examination of the ways in which networks met the needs of its members. Network characteristics were examined across subgroups that included: the household, the identified family, an information sharing network, a support network, and a total network. Network size (structure), type (function), and density variables were included in the Caregiver Interview (Appendix L). The intention of this systematic approach was to distinguish between those factors within the extended network that were relevant to cultural considerations, such as the make-up and interactional patterns within the network, and medical considerations, such as the types and sources of formal support often needed from social service workers and medical personnel.

Cultural factors were of particular importance to the intervention in this study in that it focused on enhancing the involvement of extended kinship network members. It was incorporated into the newborn screening protocol at a hospital that served the largest group of sickle cell patients in the surrounding area, and compared their program with hospital programs that did not use a similar intervention as part of their screening procedure.

The intervention was originally developed by Treadwell et al., (in press) for primary caregivers of patients with sickle cell disease in order to encourage caregivers to involve extended family members in learning more about sickle cell disease. Active knowledge acquisition of family members was defined in their study as being a form of support to caregivers. One finding was that some caregivers who took on this responsibility had poorer adjustment ratings after participating in the education protocol. A later study (Treadwell-Gad et al., 1991) examined how to best involve extended family members so that they are maximally supportive to the caregiver. A finding from this study was that shifting the responsibility of teaching extended family members to the medical treatment team helped to maximally increase caregiver adjustment ratings. The study also demonstrated that family coping levels actually improved after the first few hospitalizations occurred. So, the period of initial hospitalizations could actually be a time where the family's worst fears are met and

allayed, increasing their confidence in its ability to successfully respond to crisis situations.

An investigation is currently underway (Treadwell et al., 1994) that is examining stage-specific adjustment needs during the early phases of illness, before medical crises have begun to pile up. The timing of the intervention occurs during the first few years following diagnosis, because the family's initial response to a stressful event is a critical determining factor of the success of subsequent adaptation efforts. This is substantiated by the Double ABC-X model which distinguishes adjustment to an initial stressor, such as a diagnosis, from long term adaptation to a stressor, such as a chronic illness. The model demonstrates how, without intervention, pile-up stress can occur that impedes successful long-term adaptation. For these reasons, the current study tested the effectiveness of a hospital-based intervention that encouraged caregivers of children with SCD to seek support from an extended network, it incorporated the stage specific focus of this model and of Treadwell and Gil's adapted version, and replicated Treadwell et al.'s 1991 study.

Description

The purpose of this study was to test the effectiveness of a hospital-based intervention protocol with families of newly diagnosed children with SCD, and to further test the validity of a family stress and adaptation model used on an African American population. The intervention was designed to teach active coping strategies to caregivers and to enhance the amount and quality of support received by them from an extended kinship network. Outcomes of family and caregiver functioning were compared between hospitals in two groups, and the impact of stress and support on caregiver and family functioning was examined across both groups.

Levels of caregiver and family functioning in two groups of families from four different hospitals were compared to determine the effect of a hospital-based intervention available to families in one group. This occurred during the adjustment phase within the first five years after the newborn received an initial diagnosis, a time that was considered crucial for the long-term development of active coping strategies in the family (Treadwell, in press; McCubbin and Patterson, 1982). The two hospital groups were called the Enhanced and Standard groups. The Enhanced group received an educational intervention by a team of medical staff that taught and encouraged the caregiver to involve extended family members in the care of the diagnosed child. This was accomplished through the use of a family genogram and an accompanying interview protocol each time a contact was made with a family. The Standard group received a similar intervention that did not emphasize training the caregiver to facilitate extended family involvement. The Standard program was different from the Enhanced

program only in that it did not utilize the family genogram and interview protocol to emphasize the involvement of the extended kinship network. The content of both programs were organized around a curriculum taken from the sickle cell counselor/educator training programs designed to grant state certification. The specific protocols followed by the Standard and Enhanced sites are outlined in appendices G and H.

The goals for both the Standard and Enhanced programs were: (1) to provide adequate, accurate knowledge about the disease and patterns of inheritance from parents, (2) to know what steps to take to help the patient remain healthy, (3) to promote aspects of normal social, emotional and physical development, and (4) to know what steps to take to prevent serious illness and respond appropriately to medical emergencies. Additional goals of the enhanced program specifically addressed during each clinic visit included: (5) to assess and further develop social supports within the nuclear family, the extended family, and the community, and (6) to actively involve extended family members who act as caregivers of the patient with the health care team.

Definitions

For the purposes of this study, *family functioning* was determined by caregiver and family functioning variables. Caregiver variables included the caregiver's perception of her own coping, caregiver ability to acquire support from extended network members, and caregiver satisfaction with available support. Family functioning variables included the caregiver's perception of family coping; caregiver satisfaction with family dynamics; the family's ability to acquire support from an extended network, reframe problems, mobilize to acquire help from community resources, employ a spiritual means of coping, and to actively problem-solve. *Active problem-solving strategies* refer to the active seeking of support, information, advice and help from network members, and community resources on the part of the caregiver and family. The level of the family's *burden and lifestress* was defined as a function of the perception of burden experienced by the caregiver and family in terms of time, convenience, emotional and overall burden; and exposure to various life stresses over the previous year. *Social support* was determined by formal and informal sources of support available to the caregiver and family. This included the caregiver's perception of the amount and types of emotional and material support received from network members, the amount of formal and informal social support available to the family, and the number of members in the total extended network, support network, and family network.

Extended kinship network is defined as the total group of persons involved with a caregiver for one of the following reasons: (1) they have a shared living arrangement,

(2) they have been identified by the caregiver as "family" (see definition for identified family), (3) they have been involved by the caregiver in acquiring knowledge about SCD and how to care for the sick child, and (4) they provide emotional and/or material support to the caregiver. The extended kinship network was characterized in terms of its type and size within the following subgroups: household, identified family, sharing network, support network, and total network. *Household* refers to people who are in the caregiver's life because they live together. *Identified family* (as reported by the caregiver) refers to persons who are important in the caregiver's life, to whom she is related by blood, marriage, adoption, or friendship, and/or to whom she has made a long-term commitment. *Sharing network* refers to people who were given information by the caregiver that is needed to care for the sick child. *Support network* refers to people who actually provide some specific type of support to the caregiver. *Total network* refers to the sum of individuals named in all of the above groups. *Type categories* were divided into blood relations, non-blood relations, nuclear relations, extended relations, and formal or legal relations (i.e., professional helpers, foster parents, etc.). The size was simply the total number of people in each of the network subgroupings.

Enhanced group refers to the group of families who were receiving services at Children's Hospital Oakland. During each clinic visit, caregivers and other network members participated in a review of their knowledge about SCD, a review of resources available to them, a review of their emergency plans, a check-in on assigned tasks from the previous visit, updating of their short and long-term goals, and updating a family genogram that included any changes in household members, dates of deaths, marriages, births, and other major life events. Also, the communication of illness information to other network members was documented at each visit. *Standard group* refers to the group of families who received services at San Francisco General Hospital, Kaiser Hospital Oakland, or Mary Bridge Hospital Tacoma. Each of their clinic visits did not involve participation in developing or updating a genogram of the family's social history, and did not include a systematic review of their knowledge, resources, and short and long term goals.

Hypotheses

The first hypothesis was based on the assumption that the two groups are, in fact, qualitatively different from one another. It tested the effectiveness of the intervention in terms of how well it could enhance the caregiver's ability to acquire support from network members and involve them in the care of the ill child. The first hypothesis was that: *Subjects in the enhanced group will score significantly higher than the Standard group on indices of family functioning.*

The second and third hypotheses were foundational in nature in that they replicated an earlier study which tested the applicability of the Family Stress and Adaptation model (based on the Double ABC-X model) when used on a population of African American families of children with SCD. The second assumption being tested was that there is a positive correlation between social support and level of family functioning. The hypothesis which tested this assumption was: *Across both groups, those families who report having more social support available to them will show higher levels of functioning.*

The third assumption tested was that there is an inverse relationship between family stress and family functioning. To the extent that pile-up stress (from previous events) is lower, the family's ability to effectively cope with the stressor of a child with a chronic illness should be increased. The hypothesis that tested this assumption is: *Across both groups, lower levels of family stress will correlate with higher levels of family functioning.*

Methods

Subjects

Subjects were 67 primary caregivers of children with sickle cell disease who were interviewed between January, 1992 and January, 1994. The children were patients who had been diagnosed with sickle cell disease at birth (through newborn screening). The interviews were conducted only with the self-identified primary caregiver of the patient. Forty caregivers received medical services at Children's Hospital Oakland (CHO), 15 caregivers received medical services at Kaiser Hospital Oakland (KH), 10 received medical services at San Francisco General Hospital (SFGH), and 2 received medical services at Mary Bridge Hospital (MBH) in Tacoma, WA. The 40 caregivers receiving services at CHO were designated the Enhanced group because of the known difference in the way the Sickle Cell program at CHO was conducted. The 27 caregivers from other hospitals were designated the Standard group because they were not receiving the same programmatic treatment in their respective Sickle Cell programs. All families involved in the study had children with sickle cell disease who had been followed by the Sickle Cell clinic at their hospital during the period between January, 1988 and December, 1991.

All caregivers (from all hospitals) interviewed were female and between the ages of 16 and 71. Most were biological mothers to the patient and, in 34% (n=23) of the households, the caregiver was the only parent. Six percent (n=4) of the caregivers were extended relatives such as a grandmother, sister, or aunt, and only 3% (n=2) of the households had a foster caregiver. Foster parents were included in the study only when

they had been caring for the child for at least 50% of the child's life and had long term plans to continue caring for the child.

Eight percent (n=5) of the total caregivers had less than a junior high school education, 12% (n=8) of the caregivers had not received a high school diploma, another 30% (n=20) had only a high school education, 39% (n=26) had attended some college, and 13% (n=8) had a college degree or above. Thirty four percent (n=18) of the fathers were high school graduates, 19% (n=10) had completed some high school, 19% had completed some college, 17% (n=9) were college graduates, and 9% (n=5) had graduate or professional degrees.

Thirty six percent (n=24) of the total caregivers worked as unskilled laborers, 21% (n=14) were semi-skilled, 18% (n=12) usually worked in clerical, sales, or technical positions, and 15% (n=10) worked as executives or lesser professionals. Sixty three percent (n=42) of the caregivers were unemployed at the time of the interview. The majority of fathers were either semi-skilled (31%, n=15) or unskilled (25%, n=12). Sixteen percent (n=8) were skilled laborers, 10% (n=5) worked in clerical, sales, or technical positions, 6% (n=3) were small business owners, administrators or semi-professionals, and 12% (n=6) worked as executives or lesser professionals. Of the families with fathers who were involved, 27% (n=13) of the fathers were unemployed.

The family income for 15% (n=10) of the total group of families was under $7200 a year, 47% (n=31) of the families had income under $15,000 annually, 20% (n=13) had an income under $30,000, and 14% (n=9) had an annual income between $30,000 and $50,000. Table 2 shows frequencies and percentages for education, income, type of work, and work status in both hospital groups.

The total group of sickle cell patients ranged in age from 4 to 65 months, with most of the children falling between 2 and 4 years (mean age = 30 months) at the time of the interview. Sixty-one percent (n=41) of the children with sickle cell disease were diagnosed with hemoglobin SS, 18% (n=12) were diagnosed with hemoglobin SC, 16% (n=11) were diagnosed with Sickle Beta Thalassemia, and 5% (n=3) had other forms of sickling disease.

Fifty five percent of the patients (n=37) were males and 45% (n=30) were females. They came from households that ranged in size from 2 to 10 members, from family networks that ranged from 3 to 35 members, and their caregivers reported total networks that ranged from 3 to 41 members. Forty three percent (n=29) of the extended networks included formal (professional) or legal (foster) relationships, 36% (n=24) of the families included mixed with blood and non-blood relations, and 21% (n=14) included extended blood relatives. Caregivers reported extended family networks with 50% (n=32) mixed blood and non-blood members, and 45% (n=29) extended blood members. Tables 3 reports network size means and Table 4 reports frequencies for network type characteristics.

Procedure

The caregivers of families with diagnosed patients were seen in the sickle cell clinics of three of the four hospitals between January, 1988 and December, 1991 were contacted by telephone or during a clinic visit and asked to participate in the study. At KH, the clinic nurse specialist agreed to mail out a letter to all families with children diagnosed with SCD under five years of age (see Appendix B). The letter asked that caregivers who might be interested in participating in the study respond by mail (see Appendix C). The researcher then contacted those caregivers by telephone. If they agreed, an appointment for an interview was set up to take place in their home or at the hospital. Most of the interviews occurred in the caregiver's home.

Interviews were conducted by this researcher with the help of two assistants. At the beginning of the interview, the interviewer provided a rationale for the study, described the goals of the study in detail, and informed consent was obtained (see Appendices D & E). Caregivers were asked to sign the consent form and a List of the Rights of a Participant in Medical Research (see Appendix F). Copies of each of these documents were given to the caregiver. Caregivers were assured that participation or non-participation would not affect their continued medical care. The interviews took approximately two hours to complete. The first half of the interview (see Appendix M) was conducted in a question and answer format and the second half involved having the caregiver fill out standardized questionnaires (see Appendices I to M).

After the interview, the caregiver's questions were answered to clear up any concerns raised during the interview. They were also given names and phone numbers of hospital staff who could respond to any questions or concerns that arose in the future. See Appendix N for a summary table of all measures.

Experimental Condition

The Enhanced group program is delineated by a circumscribed time period from January, 1988 to December, 1991 and was used to identify families of patients under 5 years who were diagnosed through newborn screening and introduced to the clinic program. This time interval was chosen because it is when the Enhanced program at CHO was stable, intact, and consistently followed by the same group of staffpersons. In the Fall of 1991, three out of five team members changed in the hospital's sickle cell program, and the enhanced aspect of the study was greatly weakened. Families in the Standard group were identified using the same age range as a criterion, but the hospital protocols were different. The main difference between the Enhanced and Standard programs was the development and updating of a family genogram at each visit, an

added emphasis on encouraging the development of social supports, and the active involvement of extended family members with the health care team.

Measures

The following measures, interview form, and item subscales, were used in conducting interviews. All interview sessions were conducted using the same format and instructions, and the measures were presented in a consistent order.

Family Support Scale

The Family Support Scale (FSS, Appendix M), was administered as part of the Caregiver Interview as the standardized measure of *family support*. This scale is based on Bronfenbrenner's (1979) social ecology model and was operationalized and validated with a select sample by Dunst in 1984. Similar to the qualitative information collected in the interview on family support, this scale separates out the amount and types of support available. The 19-item self-report scale asks how helpful a list of possible sources of support are to the caregiver. Responses range from 0 (not at all helpful) to 4 (extremely helpful). Scores from the FSS include a total support score that can range from 0 to 76, an adjusted informal support subscale score and an adjusted formal support subscale score, both of which range from 0 to 14. A total score is obtained by summing all items, and higher scores indicate more sources of family support. This measure has a coefficient alpha of .77.

Family Crisis Oriented Personal Evaluation Scale

The Family Crisis Oriented Personal Evaluation Scale (F-COPES) developed by McCubbin and Patterson (1982), is a standardized measure of *family coping* that gives specific information on the *meaning and perception* attached to the illness as well as the family's *resources*. It is a 30-item self-report instrument that is completed by the caregiver. This scale (Appendix I) was developed to identify effective problem solving and behavioral strategies used by families when a crisis occurs. The F-COPES was intended to draw upon and integrate two coping dimensions of McCubbin and Patterson's (1983) Double ABC-X Model of family adaptation: Family resources (B factor) and the family's ability to reframe stressful life events in order to make them more manageable (C factor).

The 30 behavioral coping items focus on two levels of family interaction--individual to family and family to social environment. Primary caregivers are asked how their family responds when faced with problems or difficulties. Possible

responses to items range from 1 (strongly disagree) through 3 (neither agree nor disagree) to 5 (strongly agree).

The F-COPES is comprised of 5 subscales which range from 0 to 45 (Acquiring Social Support); 0 to 40 (Reframing); and 0 to 20 (Seeking Spiritual Support, Mobilizing Family to Accept and Acquire Help, and Passive Appraisal). Higher subscale scores correspond with better family coping strategies. A summed score is obtained from the F-COPES on each subscale. Cronbach's alpha for the total instrument was .86 with mean alpha coefficients of .63 for Passive Appraisal, .71 for Mobilizing Family to Acquire and Accept Help, .80 for Seeking Spiritual Support, .82 for Reframing and .83 for Acquiring Social Support. Overall test-retest reliability is .81, with a range of .61 for Reframing to .95 for Seeking Spiritual Support.

The Family Inventory of Life Events and Changes

The Family Inventory of Life Events and Changes (FILE, Appendix J), a 71-item standardized self-report questionnaire developed by McCubbin, Patterson, and Wilson (1980), measures the degree of life-stress experienced by the family during the past year and provides an index of family vulnerability. Also developed from the ABC-X model, it provides information on the *pile-up stress* factor identified in the literature as a depletor of family resources. The FILE was designed to record normative (developmental) and non-normative (situational) life events and changes experienced by the family in the previous year.

Subscales comprise the following conceptual categories: intra-family strains, marital strains, pregnancy and childbearing strains, finance and business strains, work-family transitions and strains, illness and family *care*strains, losses, transitions in and out and legal strains. Although each subcategory can be computed separately, the authors recommend using only the total scale score, with a Cronbach's alpha of .81; subscale scores appeared less stable, with alphas ranging from .30 to .73. Overall test-retest reliability was .80, with subscales ranging from .64 to .84.

Weighted scores corresponding to the magnitude of the life-stressor were assigned to each item on the FILE. These scores were then summed to obtain a total family stress score which can range from 0 (no stressors occurred during the previous year) to 3,307 (all stressors occurred during the previous year), with higher scores indicating higher stress levels.

Family Adaptation and Cohesion Scales

Family Adaptation and Cohesion Scales (FACES-III, Appendix K), is a standardized self-report measure of 40 items. Based on Olson, Russell, and Sprenkle's

(1979) Circumplex Model of Family Functioning, it provides information on the *psychosocial functioning* of the family as a system along three behavioral dimensions: adaptability, cohesion, and communication.

Items on the FACES were constructed using a 5 point-Likert-type scale with values that range from 1 to 5; 1 indicating a response of *almost never* and 5 indicating a response of *almost always*. The items are answered by the caregiver in terms of how the family actually functions and how they should ideally function. The total score used is a difference score, derived from the two versions, that can range from 0 to 80 with lower scores indicating a higher degree of caregiver satisfaction with the family's functioning. Cronbach's alphas computed for each scale were found to be .77 (Cohesion) and .62 (Adaptability).

Caregiver Evaluation

The Caregiver Evaluation (CE, Appendix L) is a 5 point Likert-type scale with 10 items that are based on the specific content of the enhanced protocol. In response to being asked how much time was spent talking about each topic area during visits with the genetic counselors, social workers and psychologists, caregiver responses could range from never (0) to always (4), with the total score ranging from 0 to 40. It was developed to provide information on the *strength of the intervention* by quantifying the qualitative differences in the contacts made with families between hospital sites.

Caregiver Interview

The investigator interviewed the caregiver, using the Caregiver Interview CI, (Appendix M), a 45 item questionnaire which provides information on family *support, knowledge, burden,* and *coping* as perceived by the caregiver. Primary caregivers were asked to report on the amount and quality of support received in such areas as emotional, emergency, financial, social, problem solving, and help with other family responsibilities; describe the amount of sharing about SCD among extended family members; and rate the level of knowledge about sickle cell disease among those members. Sections of the interview were used for descriptive purposes, as a supplement to data taken from standardized tests, and as a quantitative measure of support.

Scales from Interview Items

Caregiver Support Ratings (CARESUPPORT). This group of items classify 10 types of support that make up two factors; an emotional factor, which includes emotional support, encouragement, advice about SCD, advice about raising children,

help with problem-solving and overall support; and a material factor, which includes spending time together, help with family responsibilities, financial support, and emergency support. The total score is obtained by summing the average rating for each subscale. Caregiver support ratings (along with all other interview items) were constructed using a 5 point Likert-type scale with values ranging from 0 to 5 and, in all cases, higher scores are better.

Satisfaction with Support (SATISPORT). This item asks the caregiver for a rating of satisfaction with the total amount of support received overall.

Perceived Burden. Two groups of items (8 total) assess the perception of burden in terms of time, inconvenience, emotional impact, and overall impact of having a child with sickle cell disease. They ask for an assessment of burden for both the family (FAMBURDEN) and caregiver (CAREBURDEN). Higher scores on the interview items assessing perceived burden on the family and caregiver indicate lower levels of perceived burden.

Network Size. Network sizes are simply summed totals of the number of members reported in each sub-network (i.e., household, family, sharing, and support) or in the total network.

Hospitals

Three of the four hospital sites served populations that were very similar demographically. Located in low-income communities of fairly large cities, CHO, SFGH, and MBH mainly serviced patients of a lower socioeconomic status, many of whom rely on government subsidies to pay for their medical care. KH differs in that it services a wider range of patients in terms of their socioeconomic status. Since it has the status of an HMO, most patients served at KH receive medical benefits through the employer of a family member.

Hospital sites that comprise the Standard group were chosen based on availability of subjects through a sickle cell clinic, accessibility of the hospital, and their willingness to cooperate with research procedures. At each hospital, the team of personnel responsible for newborn patient education includes pediatric hematologists, nurse specialists, psychologists, master's level genetic counselors, and social workers.

Results

Preliminary Analysis

Before making a comparison between the Standard and Enhanced groups, preliminary tests were used to clarify the nature of the relationships among the data in the two groups. Means and standard deviations of all measures for both groups were obtained and are reported in Table 1. Differences in the number of subjects for the FACES, FILE Total, FSS, and CE scores indicate that fewer subjects completed those tests.

First, it was important to insure that the two groups did not differ on variables that could confound the results of tests on outcome measures. Of particular concern was the question of whether or not factors due to differences between the hospitals could be controlled for since the comparison groups were free standing units (different hospitals). Ideally, random assignment of subjects would control for differences due to factors other than the intervention such as the socioeconomic status of the caregiver, characteristics of caregiver networks, or caregiver age. But since the subjects for the study came from different hospitals and were not randomly assigned to the Enhanced and Standard groups, the likelihood was greatly increased that differences existed between the groups that were unrelated to the intervention.

Kruskal-Wallis Chi-Square tests were performed on categorical and rank-ordered demographic variables including patient sex, caregiver employment status, caregiver education, caregiver occupation, and income. Mann-Whitney Standardized U tests were performed on continuous demographic variables including program exposure time, caregiver age, and network size. These non-parametric tests were used because the variables did not have normal distributions. Test results indicated that the Enhanced and Standard groups were significantly different in a number of ways, including network size (Z=-2.2, $p<.03$), program exposure time (Z=-3.7, $p<.002$), caregiver employment status ($H(1)=16.7$, $p<.001$), caregiver education ($H(1)=15.2$, $p<.001$), caregiver occupation ($H(1)=15.8$, $p<.001$), and income ($H(1)=5.1$, $p<.03$). They were not significantly different in terms of patient sex. Table 5 reports the Kruskal-Wallis Chi Square test results and Table 6 reports results of the Mann-Whitney tests.

These results indicate that the test for hypothesis 1 could not yield interpretable results because they would be confounded by several sources of variance not related to the intervention. Because of this, it would not be possible to rule out differences due to the groups in determining whether or not the intervention created a difference. The possibility of conducting post hoc analyses using an Ancova to control for these variables was considered but there were too many variables to be controlled (6) given the total number of subjects (n=67). Furthermore, all six variables were related to outcome variables as well as the grouping variable, and five of the variables used

categorical (not continuous, data). Since variables used in an Ancova should be for continuous data, and should only adjust for variance related to the outcome, the use of Ancova was ruled out.

A second consideration was the reliability of the Caregiver Support subscale items from the interview. Since the subscale scores were averaged ratings of support reported by the caregiver during the interview, there was no standardized information about what their scores represented or how they could be utilized and interpreted. A principal components factor analysis with varimax rotation was performed on the set of 10 subscale scores (the total score was not included because it was the sum of the average subscale scores). The items clustered into two factors, which together accounted for 95% of the variation among the individual subscales. Factor 1, which explained 69% of the variance, included six items representing "emotional" support; factor 2, which explained 26% of the variance, included four items representing "material" support. Factor loadings for emotional support ranged from .47 to .89, with the strongest items being "emotional support " (.70), "encouragement" (.73), and "help with problem-solving" (.73). The weakest items on Factor 1, "advice about sickle cell disease" (.44) and "advice about raising children" (.48), were problematic because they were often not clearly understood by the respondent which caused them to be inconsistently interpreted, responses to these items sometimes reflected the quality of the advice instead of the amount. Loadings ranged from .38 to .87 for material support, with "spending time together" (.63) and "helping out with family responsibilities" (.66) having the highest weights. Given the fact that the item loadings for each of the two factors were greater than .40, two new summary variables (i.e., emotional support and material support) were created by summing the subscales that clustered together for each factor. Coefficient (Cronbach) alpha was .80 for emotional support and .74 for material support.

Finally, support variables taken from the interview (Caregiver Support ratings and network sizes) were correlated with the FSS total, formal adjusted, and informal adjusted subscale scores. The size of the family network reported by the caregiver only moderately correlated with the informal support scale ($r=.37$, $p<.004$) on the FSS. Otherwise, support ratings in the interview do not correlate highly with the FSS (i.e., all were $< .25$) suggesting that they in fact, are measuring a different aspect of support (i.e. caregiver support vs. family support).

Analyses of Hypotheses

The first hypothesis tested the assumption that the intervention would result in higher scores on functioning indices for the families in the Enhanced group than those in the Standard group. Measures of caregiver functioning included the caregiver's

perception of her own coping (CARECOPE) and caregiver satisfaction with available support (SATISPORT). All of these measures were taken from selected groups of interview items; refer to Appendix M (Caregiver Interview) and N for specific items. Measures of family functioning included the caregiver's perception of family coping (FAMCOPE); caregiver satisfaction with family dynamics (FACES); and the family's ability to acquire support from an extended network (F-COPES subscale), reframe problems (F-COPES subscale), mobilize to acquire help from community resources (F-COPES subscales), employ a spiritual means of coping (F-COPES subscale), and actively problem-solve (F-COPES subscale). Interview items pertaining to the perceived levels of caregiver and family coping specifically refer to the caregiver's ratings of coping with problems associated with having a child with SCD.

In addition to comparing the groups on functioning variables, a test of the strength of the intervention was also performed. The strength of the intervention (measured by the CE Total score and Family Involvement subscale) was based on the caregiver's report of how much time was spent with treatment team staff discussing the importance of utilizing community resources and involving network members to gain support in caring for the diagnosed child.

It was originally expected that the Enhanced group would have higher functioning scores and higher intervention scores in comparison to the Standard group. Mann-Whitney Standardized U tests were used to compare the two groups on all variables. Not surprisingly, no significant differences were found between the Enhanced and Standard groups on CE scores or any of the functioning variables except the family's ability to actively problem-solve (Z=-3.70, $p<0.0002$). Results of these tests are reported in Table 7.

The second hypothesis tested the assumption that those families who reported having more social support available to them would show higher levels of functioning. Measures of social support included informal and formal sources of available family support (FSS Informal and FSS Formal subscales), caregiver perception of the amount and types of emotional and material support received from network members (CARESUPPORT interview items including the emotional and material subscales, and a total support score), and network size scores (number of network members as reported in the interview) from the household, family network, support network, and total networks. Family functioning for this (and all) hypotheses was assessed using the same measures as described for hypothesis 1. It was originally expected that higher FSS scores, higher scores on the emotional and material support subscales, and larger networks, would correspond with higher scores on the FACES, F-COPES, and interview items (CARECOPE, FAMCOPE, and SATISPORT).

Pearson r correlations between family and caregiver support variables and family functioning variables are reported in Table 8. Caregivers most satisfied with the overall support received in their lives (SATISPORT score) had the most total support (r=.41,

p<.001) and informal support (r=.38, p<.01) as measured by the FSS, and received the most material forms of support (r=.32, p<.01) based on CARESUPPORT rating subscales. Those who perceived themselves as coping best (CARECOPE items) had the most total (r=.41, p<.001), informal (r=.35, p<.01), and formal (r=.30, p<.05) support based on the FSS subscales. Caregivers who were able to reframe problems in more positive terms (based on the F-COPES Reframing subscale) received more total support (r=.36, p<.01) and material support (r=.32, p<.01) as reported in their CARESUPPORT ratings. Caregivers who were most satisfied with the overall support received in their lives based on SATISPORT scores, had the largest households (r=.30, p<.01), family networks (r=.32, p<.01), support networks (r=.29, p< .05) and total networks (r=.30, p<.01). Caregivers who had the largest total networks utilized fewer passive (avoidant) coping strategies (r=.33, p<.01) as indicated by the F-COPES Passive Appraisal subscale. Finally, caregivers with the largest support networks were also more actively engaged in acquiring support from relatives, friends, neighbors, and extended family based on the F-COPES Acquired Social Support subscale (r=.31, p<.01).

The third hypothesis tested the assumption that lower levels of family stress would correlate with higher levels of family functioning. Family stress included a measure of the total life-stress experienced by the family over the previous year (measured by the FILE total and subscale scores) along with interview items assessing the perception of burden experienced by the family and caregiver in terms of time, inconvenience, emotional impact, and overall impact of having a child with sickle cell disease. It was originally expected that higher FILE scores and lower scores on the family and caregiver burden items would correspond with higher scores on the FACES, F-COPES, and interview items (CARECOPE, FAMCOPE, and SATISPORT). Pearson r correlations for the family stress and family functioning variables are reported in Table 9.

Caregivers who were least satisfied with the overall support they received (SATISPORT score) reported their families as having the highest level of total life-stress over the previous year (r=-.32, p<.01) based on the FILE Total score, the most intra-family strains (r=-.36, p<.01) based on the FILE Intra-family subscale, and problems related to illness (r=.31, p<.01) based on the FILE Illness subscale. Families with the most financial stresses (based on the FILE Financial subscale) were the most likely to employ a spiritual means of coping (r=.30, p<.01). Families with higher levels of marital stress (based on the FILE Marital subscale) rated their families as coping least well (r=-.38, p<.01). Those with the most intra-family strains (measured by the FILE Intra-Family subscale) were most dissatisfied with the way their families were functioning (r=.30, p<.05) based on the FACES score. Caregivers who reported being the most burdened financially rated their families as coping the best with having a

child with SCD ($r = .31$, $p < .001$).

Post Hoc Analyses: Testing of the Model

The previous use of this model with African American families of children with SCD had been limited to an earlier study by Treadwell-Gad et. al. (1991). Because this study is a partial replication of Treadwell-Gad's work, it seemed important to repeat a test of the theoretical basis for the Family Stress and Adaptation Model. A series of multiple regression analyses were conducted to identify individual and family characteristics in this population of subjects which predicted family functioning. This was also a way to gain more information about contributing factors to functioning outcomes, given the lack of interpretable results on hypothesis 1.

The dependent measures used in the model were measures of family functioning based on caregiver ratings of their family's level of coping with a child who has SCD (FAMCOPE), self-ratings of their own level of coping with a child who has SCD (CARECOPE), and caregiver satisfaction with their family's level of functioning (FACES). Independent variable-sets (or blocks) included resources, life-stress, support, active coping, and intervention strength. Dependent and independent variable blocks were taken from Treadwell-Gad's study with the addition of FACES as a dependent measure, and the addition of intervention strength as an independent variable block (see Table 10).

Variable selection was performed using a combination of hierarchical (theory-based, researcher-determined) and stepwise (exploratory, data-determined) multiple regression analyses (Cohen and Cohen, 1983; Montgomery and Peck, 1982). Sets (blocks) of variables (e.g., resources) were forcibly entered into the analysis in a predetermined, hierarchical order, whereas variables within sets (e.g., diagnosis, income, education) were allowed to enter in a stepwise fashion according to the amount of variance each explained in the dependent variable. The order of inclusion for the sets of variables was based upon several criteria, including their substantive role in the study (theoretical significance) and knowledge about their importance as predictors in prior research (e.g., Treadwell-Gad et al., 1991, 1994). In general, simpler, less controllable, fixed variables (e.g. resources) were entered before predictors that were less well-defined, more complex, and/or possibly open to intervention (e.g., active coping, support, or intervention strength). For these reasons, blocks of variables were entered in the following order:

Block 1: Resources
Block 2: life-stress
Block 3: Support
Block 4: Active Coping
Block 5: Intervention

Table 10 shows all variables included in each of the five blocks. Variables included within block 1, Resources, were diagnosis, income, education, and patient age; block 2, Life-stress, included total family stress (FILE Total), perceived caregiver burden (CAREBURDEN), and perceived family burden (FAMBURDEN); block 3, Support, included total family support (FSS Total), total caregiver support (CARESUPPORT), network size, and household size; block 4, Active Coping, included all F-COPES subscales; and block 5, Intervention Strength, included the CE total scale. Results of the multiple-regression analyses for the three family functioning variables (FAMCOPE, CARECOPE, and FACES) are summarized in Tables 11 through 13, including the variables entered at each step (within blocks), step change statistics, total equation statistics, and zero order correlations. The numbers in the *Step* column to the left of the variable indicates the order in which that variable entered the equation. All other variables within each block without a corresponding *step* number did not contribute enough variance to enter into the model after controlling for all variables entered previous to that step. The probability of F to enter at each step was set at 5% (.05).

When the variables for Block 1 were entered, 8% of the score variation in family coping with the disease (FAMCOPE) was explained by the caregiver's level of education. The negative standardized beta weight indicated that the higher the level of education (which is associated with lower scores due to the rating process of the Hollingshead Social Position Scale), the higher the rating given to the family's level of coping. When variables for Block 2 (life-stress) were allowed to enter, no predictors were significantly related to FAMCOPE. None of the life-stress variables predicted how well the family was coping with the disease. After controlling for resource variables, the variables for Block 3 (support) were allowed to enter. An additional 6% of the variance in FAMCOPE scores was accounted for by the scores on the FSS Total subscale. The positive beta weight indicated that higher scores on family support corresponded with higher ratings of how well the family was coping with the disease. The variables in Block 4 (active coping) were entered and an additional 16% of the variance in FAMCOPE scores was accounted for by the Mobilizing Family to Acquire and Accept Help subscale on the F-COPES. The negative beta weight showed that low scores on the family's ability to mobilize to acquire and accept help from others were associated with higher scores on family coping. Finally, the intervention strength variable in Block 5 did not enter as a predictor of FAMCOPE (or, for that matter, for

the other two family functioning variables which are discussed next). Total R-squared for the three variables was .31 (i.e., 31% explained variance in family coping). Table 11 reports results of the regression analysis for FAMCOPE predictors.

None of the variables in Block 1 (resources) entered as predictors of how well the caregiver was coping with the disease. However, when Block 2 (life-stress) variables were entered, 7% of the variance in CARECOPE scores were accounted for by the CAREBURDEN ratings. The positive beta weight indicates that lower ratings of caregiver burden due to caring for a child with SCD were associated with higher ratings of how well the caregiver was coping with the disease. After controlling for life-stress, variables in Block 3 (support) were allowed to enter, and an additional 14% of the score variation in CARECOPE was explained by the FSS Total subscale scores. The positive beta weight indicated that higher family support scores were associated with higher ratings of how well the caregiver was coping with the disease. When variables for Block 4 (active coping) were allowed to enter, an additional 9% of the variation in CARECOPE scores was explained by scores on the F-COPES Mobilizing subscale. The positive beta weight indicates that families who could seek out community resources and accept help from others were associated with higher ratings of how well the caregiver was coping with the disease. Total R-squared for the three variables was .29 (i.e., 29% explained variance in caregiver coping). Table 12 reports results of the regression analysis for CARECOPE predictors.

Approximately 18% of the score variation in the degree of caregiver satisfaction with family functioning (FACES) was shared by two variables. In Block 1 (resources), income entered as a predictor with 8% of the variance. The associated negative beta weight indicated that families with higher incomes had lower scores on the FACES, which indicated more satisfaction with family functioning. None of the life-stress variables in Block 2 came in as predictors of satisfaction with family functioning, although the rankings of total caregiver support in Block 3 (support) contributed an additional 10% of the variance related to FACES scores. Again, the negative beta weight refers to the fact that higher ratings of total caregiver support are related to higher levels of caregiver satisfaction with family functioning. Table 13 reports results of the regression analysis for predictors of caregiver satisfaction with family functioning (FACES).

In summary, caregiver education and family income each predicted approximately 8% of the variance when entered first into the equations for caregiver coping, family coping and caregiver satisfaction with family functioning; none of the resource variables came in as predictors of variance associated with how well the caregiver was coping with the disease. Support variables were consistent predictors of variance (i.e., 6% to 14%) for all three dependent variables. In addition, active coping variables were predictors for family coping and caregiver coping, but not for caregiver satisfaction

with functioning. Finally, the intervention strength was not related to caregiver coping, family coping, or caregiver satisfaction with family functioning after controlling for all other variables in the model. This finding was consistent with the univariate results for hypothesis 1 reported earlier.

Discussion

Interpretation of Results

This study attempted to evaluate the effectiveness of a hospital intervention protocol with families of newly diagnosed children with SCD, and to further test the validity of a family stress and adaptation model that accounts for the relationships between social support, stress, coping and family functioning in an African-American population. Hypothesis 1 was a test of the intervention, hypotheses 2 and 3 were testing for correlations between factors in the model, and the post-hoc multiple-regression analyses further tested the model by identifying which of those factors were predictors of caregiver and family functioning.

Hypothesis 1

First, no difference was found between the groups on measures of family functioning and intervention strength. No differences were found across all measures of functioning except on the Passive Appraisal subscale of the F-COPES. It is interesting that the Passive Appraisal subscale also entered as a predictor of family and caregiver coping in the post-hoc multiple regression analysis, but given the fact that the groups were demonstrated to be demographically different in the preliminary analyses, a valid or meaningful interpretation of these findings is not possible.

Hypothesis 2

Secondly, a relationship was found to exist between the availability of social support and the family's level of functioning. Higher levels of support available to families related to increased caregiver coping with the disease and greater caregiver satisfaction with available support. The more total support and material support available to caregivers, the more families were able to reframe problems in positive ways. Network size was also found to be related to support and coping. The larger the extended total, support, and family network, the more satisfied caregivers were with the support they received. The larger the extended total network, the fewer avoidant coping strategies were used by families in crisis situations. The larger the extended support network, the more families actively engaged in acquiring support from relatives,

friends, neighbors and extended family.

Clearly, size of network played an important role in family and caregiver coping in this study. Kazak's (1988) work on network size and density in families with chronically ill children is supported by this finding. One problem with the interpretation of this correlation is in determining the direction of the effect. It is unclear whether higher levels of support and coping for both caregivers and families is related to larger networks because the support is automatically built-in since the sheer size of the network provides more supportive people, or if the caregiver's resourcefulness and organizing abilities maximize what might only be a potential for support. In the regression analysis, after controlling for SES and life-stress variables (education, income, and perceived caregiver burden), family and caregiver support variables were the most consistent predictors of functioning. This supports findings from earlier studies (Cobb, 1976; Mitchell, 1969; & Treadwell et al., 1991) and further confirms the theoretical basis of the family stress and adaptation model (Treadwell & Gil, 1994) as it applies to African American families of children with SCD. Tables 11 and 12 show family support (FSS Total score) as entering the model in the second step as predictors of both family coping (FAMCOPE) and caregiver coping (CARECOPE). Table 13 shows caregiver support (CARESUPPORT) entering the model in the second step as a predictor of caregiver satisfaction with family functioning (FACES). Although the ability to use active coping strategies (or the decreased use of passive/avoidant coping strategies) predicted family coping in the third step (following family support), it seems that the presence of support is a more critical predictor than coping ability or network size. However, the fact that a relationship was established in this study between quality of support and size of network offers additional credence to the focus of the extended kinship network as a point of intervention, and to the intervention goal of encouraging the caregiver to involve as many network members as possible by using active coping strategies.

The finding that decreased reliance on passive (avoidant) coping strategies predicted caregiver coping also suggests the possibility that the intervention may have positively impacted caregiver coping since the focus of the intervention is on the development of active coping strategies. However, the limitations of this study in not being able to effectively test the intervention strength, or make valid group comparisons, leaves getting a definitive answer to this question for future studies.

Hypothesis 3

Finally, an inverse relationship was found to exist between family stress and family coping across a range of specific types of stressors (based on FILE subscales). Higher levels of stress due to intra-family strains, illness, and total life stresses

correlated with caregiver dissatisfaction with their family's functioning and with the support they received. Families who were coping the least well reported having the most marital stress. There was one area of stress, however, that did not seem to affect family coping in the same manner. There was a significant relationship between increased levels of financial burden for the caregivers and their reports that their families were coping the best with having a child with SCD. One interpretation for this finding is related to the difference in medical insurance coverage for low-income families in comparison to middle income families. Most of the low-income families serviced in the sickle cell clinics across hospitals are fully subsidized by the state for their medical expenses through MediCal. Middle-income families typically have private insurance plans that require them to make co-payments for services and procedures. In some cases, special procedures required for their child's care may not be covered at all. Although they might enjoy a higher income than families receiving MediCal, a certain proportion of that income has to go toward unpredictable medical costs. This could clearly create real and perceived financial stresses for caregivers of middle income families that would impact how well they are coping with having a child with sickle cell disease. Although the low-income caregivers do talk about having financial problems, these stresses are more likely created by a history of poverty and not due to their child's SCD. When these families come in to the clinic, they often have requests for help with busfare, food, and telephone calls; one caregiver had to ask for $1.00 in order to take the bus home. An interesting related finding was that families who experienced the most financial stress utilized a spiritual means of coping over all other strategies. This came up in a variety of ways during the interviews with caregivers. A belief in and reliance on a higher power was the most commonly verbalized coping mechanism, and typically corresponded with caregiver reports of feeling less burdened. Although this relationship did not show statistical significance in this study (i.e., on the F-COPES Spiritual subscale), the frequency of its presentation during the interviews suggests that it is worthy of consideration in future studies.

Post hoc Analyses: Testing of the Model

It was expected that families on the enhanced patient education protocol at CHO would have improved outcomes, but this was not supported. The fact that the intervention did not significantly affect family functioning between the two groups suggests several possible explanations: (1) there were problems in the selection of a comparison group, (2) that there were problems with the execution of the intervention, and/or (3) there were problems with the measure used to assess the strength of the intervention (CE).

The fact that the comparison group came from different hospitals made it difficult to control for differences related to programmatic procedures. Factors such as the size of the patient load and treatment teams, the relationship between staff and patients, and general hospital procedures could easily have contributed to differences between the groups. There was a difference in the number of patients served by each of the hospital clinics, with KH having the smallest group. Additionally, the treatment team at KH was smaller than the teams at the other two hospitals. This could allow for more personable relationships between staff and caregivers. Data was not collected on the quality of the relationship between staff and patients and their families which might have proven helpful in clarifying these results. Patient retention was a particular problem at SFGH which, among other things, directly affected their compliance with the interview process. An attempt was made to determine the protocol followed at each hospital clinic, although, behavioral observations of staff and patients did not occur.

Shortly after this study was underway, there were changes in the treatment team staff at the Enhanced hospital site that affected the consistency of the intervention and ultimately led to a decision to revise the design of this study. Three staffpersons (two genetic counselors and a hematologist) involved with the sickle cell program intervention left the hospital. Once hired, the new staff did not continue the intervention with the same commitment or shared vision of the purpose of the intervention and it became difficult to determine to what extent the intervention protocol was being followed. This set of events created a situation where the subjects chosen for interviews had to be limited to those caregivers that were involved with the clinic during a two-year period of time prior to the staff change. In addition to limiting the number of available subjects, it also precluded an original plan to establish a baseline of family functioning by repeating interviews over timed intervals, starting at the time the newborn had been diagnosed. Having only a single contact with families who have been exposed to the clinic for different periods of time created serious problems with the control of the study.

The items on the CE were developed from several content areas taken from the enhanced intervention protocol and were Likert-scaled to correspond with items in other parts of the interview. The items were chosen based on the topic areas identified by the treatment team for discussion with caregivers during initial contacts and subsequent clinic visits. The CE asks the caregiver to recollect and report the amount of time spent discussing a range of topics with staff. A number of factors could affect their responses, including their ability to recall, their compliance with clinic visits, the health status of their child at the time of the visits, and their relationship with the different team members with whom they talked.

Limitations

Sample Selection

A natural context for the random selection of subject types is created by the fact that sickle cell disease is caused by a genetic mutation. Also, the fact that genetic screening of newborns with sickle cell disease is now mandatory greatly increases the potential for identifying and gaining access to subjects who range a wide spectrum of family types that represent the general African-American population. Although the potential for access exists, in reality, getting subjects for studies such as this one can be quite difficult for several reasons. Because the incidence of the disease in the United States is 1:500 births, the time and cost involved in getting a large enough probability sample can be a major disadvantage. Multiple hospital sites (which limits the control for differences between the subjects at different hospital sites) and/or long periods of time available for data collection is needed. Most studies on sickle cell disease have very small samples (n=15 or 20) for this reason.

Recruitment of subjects turned out to be difficult due to the location of two of the four hospital sites, the political considerations involved with each site, time constraints, and the need to depend on the consistent motivation and willingness of caregivers who chose to participate. This was particularly true for SFGH, where caregivers were often extremely stressed and struggling with multiple problems such as high crime, financial difficulties, a lack of transportation and substance abuse. Even when these caregivers were willing to set up an interview, they often were unable to follow through with their appointments. An administrative problem developed at KH that resulted in a lack of direct access to sickle cell patients through the hospital clinic (as was the case at CHO, SFGH, and MBH). For this reason, subjects at KH had to be solicited in a different manner that those at the other hospitals. This could have created differences in the degree of motivation attributed to KH subjects (since they were first asked to respond by mail before being contacted by phone to set up an interview) which in turn could create differences between the groups due to factors unrelated to outcome measures. These types of difficulties greatly reduced the control of subject variability and the expected sample size, which in turn reduced the statistical power and further compromised the possibility of getting significant results.

Measures

There were two areas in the interview that seemed to be problematic for the caregivers and which could have compromised the accuracy of the results. Two items on the Caregiver Support Type scale often elicited variable responses, indicating a problem in how they were being interpreted. These items ask the caregiver how

helpful or supportive others are in terms of offering "advice or information about SCD" and "advice or information about raising children". Caregivers often struggled with how much advice they received vs. the quality of the advice given. It seemed clear that their feelings about what they were told influenced and sometimes overrode whether or not advice was offered at all. Although this issue in interpretation could apply to the questions about other types of support as well (e.g., "helps with problem-solving"), it didn't seem to create as much confusion for the caregivers. Most likely, these two items were interpreted both ways by this group of subjects. This could explain the results of the factor analysis, which showed weaker factor loadings only for these two items in comparison to the others.

In another part of the interview, items on family and caregiver burden often elicited a resistant response from caregivers. Some caregivers found it distasteful to talk about their child in those terms and would often respond with remarks about the "special" qualities of the diagnosed child, or of "all children being a blessing". Others, when asked for feedback at the end of the interview, asked that more of an emphasis be placed on their child's strengths instead of the disease. Many of these same caregivers volunteered that their spiritual faith was their only means of coping, which suggested a strong possibility that what could have been seen as a burdensome situation was somehow transformed into a life challenge for them. In this way, some caregivers are aware of their experience of burden, but choose not to focus on it. This type of caregiver is the least responsive to the goals of the intervention due to their resistance to looking at the problems related to the disease.

These comments and reactions may have a cultural as well as functional interpretation. Children are highly valued in African-American culture. Support for this is found in the family literature which describes a correlation between having children and increased status in the community (Martin & Martin, 1978). Parenting is seen as a shared group responsibility even though there is always one or two main caretakers in a family network (Nobles, 1985). It is quite possible that if there is a perception that the burden of responsibility to care for an ill child is shared by a network of family members, it would not feel as great in comparison to a more isolated or nuclear family structure. Additionally, religious values are deeply rooted in African-American culture, making it highly likely that many caregivers rely on their spiritual beliefs to manage problems and crises that occur. Their propensity and ability to do this could serve as a buffer to the daily stresses they experience. In any case, the findings on burden are consistent with Treadwell's (1994) earlier study and may not have been affected by these feelings expressed by caregivers (other than a decrease in the correlation strength). However, it would have been useful to have included other ways of assessing the relationship between spiritual orientation and perceptions of stress in the caregiver.

Design of Study

The lack of a pre-functioning status is a clear limitation of this study. The original design for this study had to be revised very early along the way because of the staffing changes and difficulties in getting access to subjects as mentioned earlier. There was also a "dry" period in the incidence of newborns diagnosed for SCD between the fall of 1991 and summer of 1992. The original design involved two contacts per family, with the first being in the first few months and the second six months later. This would have offered a baseline of functioning for caregivers and families, and helped to determine the impact, if any, the intervention was to have. The revised design involved one contact with each family that occurred sometime during the first five years of the diagnosed child's life. This change extended the initial adjustment period being studied from the first year of life to the first five years. Although some control was lost, the integrity of the purpose of the study was preserved since treatment for all forms of sickling disease is the same during the first five years.

Contributions

Two of the three hypotheses were supported in this study and it was able to contribute to the literature in the following ways: (1) through replication, it was able to support Treadwell et al.'s (1994) earlier findings, and others, that caregiver support, and active coping on the part of the caregiver served as a buffer to stress and contributed to better family functioning outcomes, (2) it provides meaningful pilot data for evaluating a method of intervention designed to enhance active coping skills, (3) it further supported the use of the family stress and adaptation model with an African-American population of families with children diagnosed with SCD, (4) it provides further validation of findings on the relationship between size of the support network and coping outcomes which highlights the usefulness of the extended kinship network in research on coping in African American families, and (5) it contributed to the refinement of a method of assessing caregiver support. The preliminary analyses clarified the relationship between different categories of support on a caregiver-support-type scale, and identified emotional and material support as the relevant factors assessed by the scale.

Recommendations

Systematic studies of hospital-based interventions for this population can be extremely fruitful in gaining information about what is most helpful for enhancing caregiver and family coping in families of children with sickle cell disease. This study

was the first attempt of this kind and offers some practical guidelines for repeating similar studies.

The results from the test of intervention strength strongly suggest that replication would be helpful. Teaching a model of coping behavior is a very complicated process, particularly when the treatment team members are creating a dependent type of relationship while at the same time actively promoting self-advocacy. There is a delicate balance that needs to be flexibly maintained by the caregiver and network members which integrates a "coming together" or cohesiveness with acting independently outside of the network. Minuchin (1974) talks about this in terms of the dynamic movement of families that is needed in handling crises. The type of intervention attempted at CHO was complex and multidimensional and required considerable training and coordination of staff members. It is important that the planning of future interventions involve close supervision of a core group of staff who have been trained to work toward the common goal defined by the intervention. Periodic assessments of inter-staff reliability should be done on how information is being transferred to caregivers, as well as evaluations of caregiver learning (receptivity), and staff presentation. It is a clear advantage to be able to link this type of intervention with the genetic screening protocol at a hospital by utilizing the built-in random selectivity of a statewide screening program. This allows access to a broad spectrum of family types that could come close to representing the larger population.

Some of the most meaningful information can come out of repetitive studies that allow for an evolution of ideas which systematically build over a period of time. Very little research has been conducted that applies a family-stress-and-adaptation model to a special population such as this one. This study has created another link in this research and laid the groundwork for further replication. Recommendations for the replication of this study would include: (1) further refinement of the caregiver interview items on family and caregiver support and burden; (2) assessing caregiver coping outcomes with the use of behavioral data such as parent-child interactions and compliance with hospital visits; (3) inclusion of demographic data in the statistical model on the number of crises or hospitalizations at the time of the interview and the type of insurance coverage; (4) assessment of the quality of the relationship between the treatment team and family; and (5) further assessment of spirituality as a factor that can interact with coping, stress, and support variables to impact caregiver functioning.

The literature on adaptation to medical crises and characteristics of African-American family dynamics should continue to be explored in combination. This area of research can offer important models for studies of family adaptation to caring for children with genetically transmitted diseases (e.g. Thalassemia) that need to consider culturally relevant interventions. Extended families are common in most traditional ethnic communities with some differences in the ways in which they interrelate and function (Castillo et al., 1967). The refinement of models appropriate for studying the

function of extended kinship systems in terms of health and illness, including their interaction with systems of more formalized care, addresses a clearly apparent gap in our knowledge (Litman, 1989). Additionally, recent research on health and SES variables suggest that SES may operate differently within racial groups and may interact with race to affect health. This is especially true among low SES African Americans (Adler, Boyce, Chesney, Cohen, Folkman, Kahn, and Syme, 1994). These researchers suggest that SES variables may function most powerfully in the form of combinations of variables that are chosen because of their relevance to a specific profile. For example, instead of just classifying families according to the standard SES indicators, sets of variables inclusive of SES variables as well as other domains, such as the physical and social environment (e.g., crowding, pollution, etc.), health conditions and behaviors (e.g., substance abuse), and beliefs about illness, as well as family demographic variables (network characteristics, type of health insurance, etc.) would help to tease out more subtle psychological outcome differences otherwise lost. This directly applies to the needed direction for the current research. By closely examining the co-occurence and interaction of combinations of variables related to several profiles of family types, we can more clearly understand how families with very different profiles can take different paths (in terms of intervention needs) to reach the same outcomes. Although this goes far beyond the scope of the current study, it is hoped that it has been a step along the way to beginning to identify relevant factors (i.e., active coping, stress, support, income, spirituality, network size) in relationship to each other as they pertain to family and caregiver coping. Once the patterns of relationships between the factors is clearly understood across a range of family situations, specific family profiles can emerge (e.g., flexible/active copers, dependent copers, multi-problem family, etc.). These profiles will inform future efforts to design interventions appropriately matched to family profiles.

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Appendix A

Letter to Caregivers of Children Diagnosed With SCD Under 5 Years of Age at San Francisco General Hospital

April 25, 1992

(Caregiver's Name)
(Caregiver's Address)
San Francisco, CA

Dear (Caregiver's Name),

The staff of the Sickle Cell Clinics at San Francisco General Hospital and Children's Hospital Oakland are currently in the process of evaluating the program of support and education provided for parents and guardians caring for infants and young children who are currently enrolled in the Northern California Comprehensive Sickle Cell Center. We are doing this so that we can continue to develop our program in ways that will best fit your needs. We are also interested in finding out what families find helpful as they adjust to caring for children with sickle cell disease.

We are asking that you, as (Patient's Name)'s caregiver, participate in an interview about what family life has been like since (Patient's Name) was diagnosed with sickle cell disease, what you have found helpful so far, and what more you feel you might need to help you manage. In addition to the interview, you would also be asked to complete some brief questionnaires that contain questions related to those in the interview. The interview and questionnaires together will take about 45 minutes to one hour to complete. You will be interviewed by Ms. Jackson wherever you find it most convenient to meet; at your home, in the clinic, or at another place of your choice.

All of the information gathered will be held confidential, that is, your answers will be grouped together without names. Whether or not you choose to participate will not affect your present or future medical care at San Francisco General Hospital. Your participation is strictly voluntary.

If any questions come up for you about this, please feel free to contact Sara Heller at (415) 206-3770, or Genée Jackson or Dr. Treadwell at (510) 428-3570. If you would like, you can request a copy of the results of the survey when the project is completed. Thank you for your help.

Sincerely yours,

Sara Heller, R.N.

Appendix B

Letter To Caregivers of Children Diagnosed With SCD Under 5 Years of Age at Kaiser Hospital

April 30, 1992

(Parent's Name)
(Address)
Oakland, CA

Dear (Parent's Name),

The Northern California Comprehensive Sickle Cell Center is currently in the process of evaluating programs of support and education provided for parents and guardians caring for infants and young children who are currently enrolled in sickle cell clinics in this area. They are doing this so that they can continue to develop sickle cell programs in ways that will best fit the needs of families like yours, and to find out what your family has found to be helpful in adjusting to the diagnosis and management of your child's illness.

They have asked that you, as (Patient's Name)'s caregiver, participate in an interview about what family life has been like since (Patient's Name) was diagnosed with sickle cell disease, what you have found helpful so far, and what more you feel you might need to help you manage. In addition to the interview, you would also be asked to complete some brief questionnaires that contain questions related to those in the interview. The interview and questionnaires together will take about 45 minutes to one hour to complete. You would be interviewed by Ms. Genée Jackson or Dr. Marsha Treadwell (from Children's Hospital Oakland) whenever and wherever you find it most convenient to meet. If you agree to participate, you will be contacted by one of them in the next few weeks.

All of the information gathered will be held confidential, that is, your answers will be grouped together without names. Whether or not you choose to participate will not affect your present or future medical care at Kaiser Hospital. Your participation is strictly voluntary and if you would like to do so, you can request a copy of the results of the survey when the project is completed.

Please take a minute to confirm whether or not you agree to participate in this survey, by checking the appropriate box, signing, and returning the enclosed form in the postage paid envelope I have provided.

If you have any questions or simply want more information about this, please feel free to contact me at (510) 596-6592, or Genée Jackson or Dr. Marsha Treadwell at (510) 428-3570. Ms. Jackson and Dr. Treadwell thank you for your help in this effort.

Sincerely yours,

Sharon Johnson, R.N.

Appendix C

Northern California Comprehensive Sickle Cell Center

THANK YOU FOR YOUR INTEREST IN INCLUDING ME IN YOUR SURVEY

☐ I am willing to help out by participating in an interview

☐ I would not like to be included

_____

Signature

_____

Date

Please return this form immediately in the enclosed postage paid envelope.

Appendix D

Children's Hospital Oakland/Kaiser Hospital Oakland/SFGH
CONSENT TO PARTICIPATE IN RESEARCH

Name of This Study:
Extended Family Education and Coping In Families Of Patients With Sickle Cell Disease

Purpose of This Study:
This study is intended to provide a better understanding of the role of social supports in their effect on the family's ability to cope with a child who has sickle cell disease. Your family has been invited to participate in this study because you have a child with sickle cell disease who receives medical services from the Sickle Cell Program at Children's Hospital Oakland, Kaiser Hospital Oakland, San Francisco General Hospital, or Mary Bridge Hospital in Tacoma.

Sponsorship:
This study is being conducted by Genée Jackson, M.S. of Children's Hospital Oakland in collaboration with Marsha Treadwell, Ph.D., Elliott Vichinsky, M.D., and Herbert Schreier, M.D. of Children's Hospital Oakland. Information for contacting any of the project sponsors is available on the following pages.

Procedures:
If you agree to participate in this study, the following will be scheduled:

(1) You will be contacted by phone or letter by Genée Jackson, M.S. and asked for your permission to participate during your regular clinic visit or at another agreed upon time.
(2) You will be given several questionnaires to complete in writing at one time. The questionnaires will be about recent family life events and changes; what behaviors help the family cope with stress; and how flexible and close to one another you view your family members.
(3) You will be interviewed by Genée Jackson, M.S. about who in the family gives you support, and what types of support they provide.
(4) The total time needed to complete the questionnaires and interview will be approximately one hour.
(5) You may be asked to complete the interview and questionnaires in four months, and then once yearly for five years.

Risks/Discomforts:
It is believed that there is minimal psychological or social risk involved in participation in this study. It is possible, however, that recalling stressful events and describing current stress will make you feel uncomfortable. Although the time commitment of one hour may be inconvenient for you, arrangements can and will be made to best suit your needs.

Families of newly diagnosed infants tend to feel particularly stressed at this time so every effort will be made to make this a smooth process. You may stop the interview at any time or refuse to answer any question. There are no foreseeable adverse consequences for voluntarily withdrawing from the project.

Benefits:
The information obtained by this study may help families of children with sickle cell disease who receive treatment and education. If you wish to recieve a copy of the results of this study let the investigator know. These results may be helpful in understanding how your family copes with stress.

Alternatives:
If you choose not to have your family included in this study, care routinely provided will continue to be made available to your child and any appropriate information will be provided to your family.

Confidentiality:
Your child's medical records and the records of this study will be handled as confidentially as other medical records, except that representatives of the sponsor and the FDA may need to see them. Your family will be assigned a code number (instead of using your name) at the onset which will be matched to a single master list which will be kept in a locked file and known only to the investigator. Only the code number will appear on the answer sheets. The information gathered in this study will be kept three years after the study ends, and then destroyed.

Treatment and Compensation for Injury:
In the unlikely event that you or any other family member is feeling anxious as a result of participating in this study, contact Genée Jackson, M.S. or Dr. Marsha Treadwell at the address below and a consultation with a licensed therapist will be made available to you at no cost:

Genée Jackson, M.S.
or
Marsha Treadwell, Ph.D.
Department of Psychiatry
Children's Hospital Oakland
770 53rd Street
Oakland, CA 94609
(510) 428-3356

Questions:
Before you agree to participate, you should talk with Genée Jackson, M.S. or Dr. Treadwell, who will answer your questions. If you have other questions during the course of this study, you may call:

Elliot Vichinsky, M.D.
Division Chief and Director
Dept. of Hematology/Oncology
Children's Hospital Oakland
747 52nd Street
Oakland, CA 94609
(510) 428-3651

Herbert Schreier, M.D., Chief
Dept. Of Psychiatry
Children's Hospital, Oakland
770 53rd Street
Oakland, CA 94609
(510) 428-3570

Additionally, if your wish to speak to a physician who is not involved with this research project and is available for reference, you may contact:

Robert Gerdsen, M.D.
Director of Medical Affairs
Children's Hospital Oakland
747 52nd Street
Oakland, CA 94609
(510) 428-3331

Participation in Research is Voluntary:
You have the right to refuse to take part in this study.

Consent To Be A Research Participant And List of Rights:
Your signature on the next page indicates that you consent to participate in this study. You will be given a copy of this form and a copy of the "Lists of Rights of a Participant in a Medical Experiment" to keep.

Signature of Consent:
If this project has been explained to you and you have had the chance to ask all the questions you want and you agree to take part, please sign below.

_____
Participant's Name (Please Print)

_____ _____
Participant's Signature Date

_____ _____
Participant's Address Phone

_____ _____
Witness Date

Appendix E

Mary Bridge Hospital, Tacoma, WA
CONSENT TO PARTICIPATE IN RESEARCH

Name of This Study:
Extended Family Education and Coping In Families Of Patients With Sickle Cell Disease

Purpose of This Study:
This study is intended to provide a better understanding of the role of social supports in their effect on the family's ability to cope with a child who has sickle cell disease. Your family has been invited to participate in this study because you have a child with sickle cell disease who receives medical services from the Sickle Cell Program at Children's Hospital Oakland, Kaiser Hospital Oakland, San Francisco General Hospital, or Mary Bridge Hospital in Tacoma.

Sponsorship:
This study is being conducted by Genée Jackson, M.S. of Children's Hospital Oakland in conjunction with Dr. Marsha Treadwell, Ph.D., Elliott Vichinsky, M.D., and Herbert Schreier, M.D. of Children's Hospital Oakland, and Melanie Holladay, Ph.D. of Mary Bridge Hospital . Information for contacting any of the project sponsors is available on the following pages.

Procedures:
If you agree to participate in this study, the following will be scheduled:

(1) You will be contacted by phone or letter by Genée Jackson, M.S. and asked for your permission to participate during your regular clinic visit or at another agreed upon time.
(2) You will be given several questionnaires to complete in writing at one time. The questionnaires will be about recent family life events and changes; what behaviors help the family cope with stress; and how flexible and close to one another you view your family members.
(3) You will be interviewed by Genée Jackson, M.S. about who in the family gives you support, and what types of support they provide.
(4) The total time needed to complete the questionnaires and interview will be approximately one hour.
(5) You may be asked to complete the interview and questionnaires in four months, and then once yearly for five years.

Risks/Discomforts:
It is believed that there is minimal psychological or social risk involved in participation in this study. It is possible, however, that recalling stressful events and describing current stress will make you feel uncomfortable. Although the time commitment of one hour may be inconvenient for you, arrangements can and will be made to best suit your needs.

Families of newly diagnosed infants tend to feel particularly stressed at this time so every effort will be made to make this a smooth process. You may stop the interview at any time or refuse to answer any question. There are no foreseeable adverse consequences for voluntarily withdrawing from the project.

Benefits:
The information obtained by this study may help families of children with sickle cell disease who receive treatment and education. If you wish to recieve a copy of the results of this study let the investigator know. These results may be helpful in understanding how your family copes with stress.

Alternatives:
If you choose not to have your family included in this study, care routinely provided will continue to be made available to your child and any appropriate information will be provided to your family.

Confidentiality:
Your child's medical records and the records of this study will be handled as confidentially as other medical records, except that representatives of the sponsor and the FDA may need to see them. Your family will be assigned a code number (instead of using your name) at the onset which will be matched to a single master list which will be kept in a locked file and known only to the investigator. Only the code number will appear on the answer sheets. The information gathered in this study will be kept three years after the study ends, and then destroyed.

Treatment and Compensation for Injury:
In the unlikely event that you or any other family member is feeling anxious as a result of participating in this study, contact Genée Jackson, M.S. or Dr. Melanie Holladay at the address below and a consultation with a licensed therapist will be made available to you at no cost:

Melanie Holladay, Ph.D.
Pediatric Psychiatry Service
P. O. Box 5299
Tacoma, WA 98415
(206) 552-1465

Genée Jackson, M.S.
or
Marsha Treadwell, Ph.D.
Department of Psychiatry
Children's Hospital Oakland
770 53rd Street
Oakland, CA 94609
(510) 428-3356

Questions:
Before you agree to participate, you should talk with Genée Jackson, M.S. or Dr. Treadwell, who will answer your questions. If you have other questions during the course of this study, you may call:

Elliot Vichinsky, M.D.
Division Chief and Director
Dept. of Hematology/Oncology
Children's Hospital Oakland
747 52nd Street
Oakland, CA 94609
(510) 428-3651

Herbert Schreier, M.D., Chief
Dept. Of Psychiatry
Children's Hospital, Oakland
770 53rd Street
Oakland, CA 94609
(510) 428-3570

Additionally, if your wish to speak to a physician who is not involved with this research project and is available for reference, you may contact:

Robert Gerdsen, M.D.
Director of Medical Affairs
Children's Hospital Oakland
747 52nd Street
Oakland, CA 94609
(510) 428-3331

In the event that you have questions concerning your rights as a patient you may also contact the Multicare Medical Center Institutional Review Board at (206) 594-1085.

Participation in Research is Voluntary:
You have the right to refuse to take part in this study.

Consent To Be A Research Participant And List of Rights:
Your signature on the next page indicates that you consent to participate in this study. You will be given a copy of this form and a copy of the "Lists of Rights of a Participant in a Medical Experiment" to keep.

Signature of Consent:
If this project has been explained to you and you have had the chance to ask all the questions you want and you agree to take part, please sign below.

_____
Participant's Name (Please Print)

_____ _____
Participant's Signature Date

_____ _____
Participant's Address Phone

_____ _____
Witness Date

Appendix F

LIST OF RIGHTS OF A PARTICIPANT IN A MEDICAL EXPERIMENT

Under California law, if you are a person participating in a medical study, you must be told:

- The purpose of the study,
- The procedures which will be followed in the study, and the drugs or devices which will be used,
- If there are any other possible treatment, procedures, drugs or devices which can be given (instead of those offered in the study), and the risks or benefits of those other possibilities,
- If there are any discomforts or risks you may expect from participating in the study,
- If any medical treatment will be available to you if complications happen during or after the study,
- If you might benefit by taking part in the study,
- That you may ask any questions about the study and that you must receive answers,
- That you may leave the study at any time, without affecting the quality of care you would receive outside the study,
- That you may freely decide if you want to take part in the study and not be pressured into your decision,
- That you will be given a copy of the written, signed, and dated "Consent to be a Research Participant" form.

Dated: _____, 19____.

_____
PARENT OR GUARDIAN OF:

_____
(NAME OF MINOR CHILD)

Appendix G

Enhanced Intervention Protocol (EIP)

This protocol for families of new babies was used at the Children's Hospital as follows:

A. Initial contact between the family and the genetic counselor
   1. Initial phone contact with family is made by the genetic counselor (same as SIP)
   2. An office visit with the family is arranged (same as SIP)
   3. Fathers are encouraged to attend along with other extended family members and/or friends who are interested, or who are identified by the parents as possibly having responsibility for the baby's care in the future
   4. The family history or genogram (McGoldrick & Gerson, 1985) is constructed on this first visit. The usual information about the genetic transmission of medical condition is gathered, but additional information is obtained and coded:
      a) Patterns of communication about the disease
      b) Support network of primary caregivers
      c) Strategies of coping in other crisis situations
      d) Risk factors that might impede coping
   5. Primary caregivers are educated about signs and symptoms of sickle cell disease (same as SIP)
   6. Supportive counseling is given by the genetic counselor as needed (as in SIP)
   7. Family receives a copy of handbook, part 1 (same as SIP)

B. Initial Assessment
   1. A second appt is made for collecting information from the family within 48 hours
   2. The primary caregiver will be asked to fill out the F-COPES, FILE and FACES-III

C. First clinic visit
   1. Nurse specialist teaches the primary caregiver how to take the baby's temperature, situations in which they should take the temperature, feel for the spleen, when to call the clinic or hematologist on call, and lists immunizations the child will need (same as SIP)
   2. Hematologist reinforces education about medical aspects and future problems the child might encounter (same as SIP)
   3. The genetic counselor reviews previously presented material; goes into detail about any area as indicated (same as SIP)
   4. Social worker begins psychosocial assessment, provides information and assistance in addressing concrete and emotional needs (as in SIP)

D. All Follow-up visits
   1. Review information previously presented (same as SIP)
   2. Provide new information (same as SIP)
   3. Update genogram
      a) patterns of information exchange about the disease within the family system
      b) significant changes in the family since last meeting (births,deaths, pregnancies, moves, etc.)
   4. Assess how well goals for the family are being addressed
   5. Encourage the ongoing development of supports in the extended family and community
   6. Assess family's consolidation of information and coping after the first hospitalization

E. Four month Follow-up Assessment
   1. At 4 months after the initial assessment, the tests will be administered again for comparative data on any changes in their level of stress and coping.

Appendix H

Standard Intervention Protocol (SIP)

This protocol for families of new babies was used at the General Hospital as follows:

A. Initial contact between the family and the genetic counselor
   1. Initial phone contact with family is made by the genetic counselor
   2. An office visit with the family is arranged within a few days
   3. Fathers are encouraged to attend this and future visits
   4. A family history is taken, including information obtained about genetically transmitted medical conditions
   5. Primary caregivers are educated about the function of hemoglobin and the variation in hemoglobin types; individual and family hemoglobin test results and their significance; the inheritance of hemoglobin types; the clinical course of the condition, stressing the difficulty in predicting clinical severity; signs and symptoms of the disease; clinical management and preventative measures that may affect prognosis; and reproductive issues. Inquiries are made about the family's prior knowledge of sickle cell disease and acquaintance with affected persons.
   6. Supportive counseling by the genetic counselor is often necessary as the time of an initial diagnosis can be very stressful and frightening for families.
   7. The family is given a copy of A Parents' Handbook For Sickle Cell Disease, Part 1 (Lessing and Vichensky, 1990).

B. Initial Assessment
   1. A second appt is made for collecting information from the family within 48 hours
   2. The primary caregiver will be asked to fill out the F-COPES, FILE and FACES-III

C. First clinic visit
   1. Nurse specialist teaches the primary caregiver how to take the baby's temperature, situations in which hey should take the temperature, feel for the spleen, when to call the clinic or hematologist on call, and lists immunizations the child will need
   2. Hematologist reinforces education about medicalaspects and future problems the child might encounter
   3. The genetic counselor reviews previously presented material; goes into detail about any area as indicated
   4. Social worker begins psychosocial assessment, provides information and assistance in addressing concrete and emotional needs

D. All Follow-up visits
   1. Review all information previously presented
   2. Provide new information
   3. Update family history as to significant changes in the family since last meeting (births, deaths, pregnancies).
   4. Families are brought back to clinic once every two to three months initially, and then annually after the age of one year.

E. Four month Follow-up Assessment
   1. At 4 months after the initial assessment, the tests will be administered again (F-COPES, FILE, and FACES III) again for comparative data on any changes in their level of stress and coping.

Appendix I

FAMILY STRESS COPING AND HEALTH PROJECT
[illegible] Linden Drive
University of Wisconsin-Madison
Madison, WI 53706

Family Health Program
FORM A
1981
H. McCubbin

F-COPES

FAMILY CRISIS ORIENTED PERSONAL SCALES

Hamilton I. McCubbin David H. Olson Andrea B. Larsen

PURPOSE

The Family Crisis Oriented Personal Evaluation Scales is designed to record effective problem-solving attitudes and behavior which families develop to respond to problems or difficulties.

DIRECTIONS

First, read the list of "Response Choices" one at a time.

Second, decide how well each statement describes your attitudes and behavior in response to problems or difficulties. If the statement describes your response very well, then circle the number 5 indicating that you STRONGLY AGREE; if the statement does not describe your response at all, then circle the number 1 indicating that you STRONGLY DISAGREE; if the statement describes your response to some degree, then select a number 2, 3, or 4 to indicate how much you agree or disagree with the statement about your response.

WHEN WE FACE PROBLEMS OR DIFFICULTIES IN OUR FAMILY, WE RESPOND BY:	Strongly Disagree	Moderately Disagree	Neither Agree Nor Disagree	Moderately Agree	Strongly Agree
1 Sharing our difficulties with relatives	1	2	3	4	5
2 Seeking encouragement and support from friends	1	2	3	4	5
3 Knowing we have the power to solve major problems	1	2	3	4	5
4 Seeking information and advice from persons in other families who have faced the same or similar problems	1	2	3	4	5
5 Seeking advice from relatives (grandparents, etc.)	1	2	3	4	5
6 Seeking assistance from community agencies and programs designed to help families in our situation	1	2	3	4	5
7 Knowing that we have the strength within our own family to solve our problems	1	2	3	4	5
8 Receiving gifts and favors from neighbors (e.g. food, taking in mail, etc.)	1	2	3	4	5
9 Seeking information and advice from the family doctor	1	2	3	4	5
10 Asking neighbors for favors and assistance	1	2	3	4	5

	WHEN WE FACE PROBLEMS OR DIFFICULTIES IN OUR FAMILY, WE RESPOND BY:	Strongly Disagree	Moderately Disagree	Neither Agree Nor Disagree	Moderately Agree	Strongly Agree
11	Facing the problems "head-on" and trying to get solution right away	1	2	3	4	5
12	Watching television	1	2	3	4	5
13	Showing that we are strong	1	2	3	4	5
14	Attending church services	1	2	3	4	5
15	Accepting stressful events as a fact of life	1	2	3	4	5
16	Sharing concerns with close friends	1	2	3	4	5
17	Knowing luck plays a big part in how well we are able to solve family problems	1	2	3	4	5
18	Exercising with friends to stay fit and reduce tension	1	2	3	4	5
19	Accepting that difficulties occur unexpectedly	1	2	3	4	5
20	Doing things with relatives (get-togethers, dinners, etc.)	1	2	3	4	5
21	Seeking professional counseling and help for family difficulties	1	2	3	4	5
22	Believing we can handle our own problems	1	2	3	4	5
23	Participating in church activities	1	2	3	4	5
24	Defining the family problem in a more positive way so that we do not become too discouraged	1	2	3	4	5
25	Asking relatives how they feel about problems we face	1	2	3	4	5
26	Feeling that no matter what we do to prepare, we will have difficulty handling problems	1	2	3	4	5
27	Seeking advice from a minister	1	2	3	4	5
28	Believing if we wait long enough, the problem will go away	1	2	3	4	5
29	Sharing problems with neighbors	1	2	3	4	5
30	Having faith in God	1	2	3	4	5

Appendix J

FAMILY STRESS COPING AND HEALTH PROJECT
1300 Linden Drive
University of Wisconsin-Madison
Madison, WI 53706

Family Health Program
FORM C
1983
© H. McCubbin

IID	☐	☐	☐	☐
GID	☐	☐	☐	
FID	☐	☐	☐	☐

FILE

Family Inventory of Life Events and Changes

Hamilton I. McCubbin Joan M. Patterson Lance R. Wilson

PURPOSE

Over their life cycle, all families experience many changes as a result of normal growth and development of members and due to external circumstances. The following list of family life changes can happen in a family at any time. Because family members are connected to each other in some way, a life change for any one member affects all the other persons in the family to some degree.

> "FAMILY" means a group of two or more persons living together who are related by blood, marriage or adoption. This includes persons who live with you and to whom you have a long term commitment.

DIRECTIONS

"DID THE CHANGE HAPPEN IN YOUR FAMILY?"
Please read each family life change and decide whether it happened to any member of your family—including you.

- DURING THE LAST YEAR
  First, decide if it happened any time during the last 12 months and check YES or NO.

During Last 12 Months	
Yes	No
☐	☐

FAMILY LIFE CHANGES		DID THE CHANGE HAPPEN IN YOUR FAMILY? During Last 12 Months Yes	No	Score
I. INTRA-FAMILY STRAINS				
1. Increase of husband/father's time away from family	46	☐	☐	
2. Increase of wife/mother's time away from family	51	☐	☐	
3. A member appears to have emotional problems	58	☐	☐	
4. A member appears to depend on alcohol or drugs	66	☐	☐	
5. Increase in conflict between husband and wife	53	☐	☐	
6. Increase in arguments between parent(s) and child(ren)	45	☐	☐	
7. Increase in conflict among children in the family	48	☐	☐	
8. Increased difficulty in managing teenage child(ren)	55	☐	☐	
9. Increased difficulty in managing school age child(ren) (6-12 yrs.)	39	☐	☐	
10. Increased difficulty in managing preschool age child(ren) (2½-6 yrs.)	36	☐	☐	
11. Increased difficulty in managing toddler(s) (1-2½ yrs.)	36	☐	☐	
12. Increased difficulty in managing infant(s) (0-1 yr.)	35	☐	☐	
13. Increase in the amount of "outside activities" which the child(ren) are involved in	25	☐	☐	
14. Increased disagreement about a member's friends or activities	35	☐	☐	
15. Increase in the number of problems or issues which don't get resolved	45	☐	☐	
16. Increase in the number of tasks or chores which don't get done	35	☐	☐	
17. Increased conflict with in-laws or relatives	40	☐	☐	
II. MARITAL STRAINS				
18. Spouse/parent was separated or divorced	79	☐	☐	
19. Spouse/parent has an "affair"	68	☐	☐	
20. Increased difficulty in resolving issues with a "former" or separated spouse	47	☐	☐	
21. Increased difficulty with sexual relationship between husband and wife	58	☐	☐	

Subtotal 1 ____

Please turn over and complete

FAMILY LIFE CHANGES	DID THE CHANGE HAPPEN IN YOUR FAMILY? During Last 12 Months Yes	No	Score
III. PREGNANCY AND CHILDBEARING STRAINS			
22. Spouse had unwanted or difficult pregnancy 45	☐	☐	
23. An unmarried member became pregnant 65	☐	☐	
24. A member had an abortion 50	☐	☐	
25. A member gave birth to or adopted a child 60	☐	☐ a	
IV. FINANCE AND BUSINESS STRAINS			
26. Took out a loan or refinanced a loan to cover increased expenses 29	☐	☐	
27. Went on welfare 65	☐	☐	
28. Change in conditions (economic, political, weather) which hurts the family business 41	☐	☐	
29. Change in Agriculture Market, Stock Market, or Land Values which hurts family investments and/or income 43	☐	☐	
30. A member started a new business 50	☐	☐	
31. Purchased or built a home 41	☐	☐	
32. A member purchased a car or other major item 19	☐	☐	
33. Increasing financial debts due to over-use of credit cards 31	☐	☐	
34. Increased strain on family "money" for medical/dental expenses 23	☐	☐	
35. Increased strain on family "money" for food, clothing, energy, home care 21	☐	☐	
36. Increased strain on family "money" for child(ren)'s education 22	☐	☐	
37. Delay in receiving child support or alimony payments 41	☐	☐ a	
V. WORK-FAMILY TRANSITIONS AND STRAINS			
38. A member changed to a new job/career 40	☐	☐	
39. A member lost or quit a job 65	☐	☐	
40. A member retired from work 48	☐	☐	
41. A member started or returned to work 41	☐	☐	
42. A member stopped working for extended period (e.g., laid off, leave of absence, strike) 61	☐	☐	
43. Decrease in satisfaction with job/career 45	☐	☐	
44. A member had increased difficulty with people at work 32	☐	☐	
45. A member was promoted at work or given more responsibilities 40	☐	☐	
46. Family moved to a new home/apartment 43	☐	☐	
47. A child/adolescent member changed to a new school 24	☐	☐ a a	
Subtotal 3			

FAMILY LIFE CHANGES	DID THE CHANGE HAPPEN IN YOUR FAMILY? During Last 12 Months Yes	No	Score
VI. ILLNESS AND FAMILY "CARE" STRAINS			
48. Parent/spouse became seriously ill or injured 44	☐	☐	
49. Child became seriously ill or injured 35	☐	☐	
50. Close relative or friend of the family became seriously ill 44	☐	☐	
51. A member became physically disabled or chronically ill 73	☐	☐	
52. Increased difficulty in managing a chronically ill or disabled member 58	☐	☐	
53. Member or close relative was committed to an institution or nursing home 44	☐	☐	
54. Increased responsibility to provide direct care or financial help to husband's and/or wife's parent(s) 47	☐	☐	
55. Experienced difficulty in arranging for satisfactory child care 40	☐	☐ a	
VII. LOSSES			
56. A parent/spouse died 98	☐	☐	
57. A child member died 99	☐	☐	
58. Death of husband's or wife's parent or close relative 48	☐	☐	
59. Close friend of the family died 47	☐	☐	
60. Married son or daughter was separated or divorced 58	☐	☐	
61. A member "broke up" a relationship with a close friend 39	☐	☐ a	
VIII. TRANSITIONS "IN AND OUT"			
62. A member was married 42	☐	☐	
63. Young adult member left home 43	☐	☐	
64. A young adult member began college (or post high school training) 28	☐	☐	
65. A member moved back home or a new person moved into the household 42	☐	☐	
66. A parent/spouse started school (or training program) after being away from school for a long time 38	☐	☐ a	
IX. FAMILY LEGAL VIOLATIONS			
67. A member went to jail or juvenile detention 68	☐	☐	
68. A member was picked up by police or arrested 57	☐	☐	
69. Physical or sexual abuse or violence in the home 75	☐	☐	
70. A member ran away from home 61	☐	☐	
71. A member dropped out of school or was suspended from school 38	☐	☐ a a	
Subtotal 4			
Grand Total			

F-9245

Appendix K

FACES III

David H. Olson, Joyce Portner, and Yoav Lavee

1	2	3	4	5
ALMOST NEVER	ONCE IN AWHILE	SOMETIMES	FREQUENTLY	ALMOST ALWAYS

DESCRIBE YOUR FAMILY NOW:

____ 1. Family members ask each other for help.

____ 2. In solving problems, the children's suggestions are followed.

____ 3. We approve of each other's friends.

____ 4. Children have a say in their discipline.

____ 5. We like to do things with just our immediate family.

____ 6. Different persons act as leaders in our family.

____ 7. Family members feel closer to other family members than to people outside the family.

____ 8. Our family changes its way of handling tasks.

____ 9. Family members like to spend free time with each other.

____ 10. Parent(s) and children discuss punishment together.

____ 11. Family members feel very close to each other.

____ 12. The children make the decisions in our family.

____ 13. When our family gets together for activities, everybody is present.

____ 14. Rules change in our family.

____ 15. We can easily think of things to do together as a family.

____ 16. We shift household responsibilities from person to person.

____ 17. Family members consult other family members on their decisions.

____ 18. It is hard to identify the leader(s) in our family.

____ 19. Family togetherness is very important.

____ 20. It is hard to tell who does which household chores.

FAMILY SOCIAL SCIENCE, 290 McNeal Hall, University of Minnesota, St. Paul, MN 55108

FACES III: Ideal Version

David H. Olson, Joyce Portner, and Yoav Lavee

1	2	3	4	5
ALMOST NEVER	ONCE IN AWHILE	SOMETIMES	FREQUENTLY	ALMOST ALWAYS

IDEALLY, how would you like YOUR FAMILY TO BE:

_____ 21. Family members would ask each other for help.

_____ 22. In solving problems, the children's suggestions would be followed.

_____ 23. We would approve of each other's friends.

_____ 24. The children would have a say in their discipline.

_____ 25. We would like to do things with just our immediate family.

_____ 26. Different persons would act as leaders in our family.

_____ 27. Family members would feel closer to each other than to people outside the family.

_____ 28. Our family would change its way of handling tasks.

_____ 29. Family members would like to spend free time with each other.

_____ 30. Parent(s) and children would discuss punishment together.

_____ 31. Family members would feel very close to each other.

_____ 32. Children would make the decisions in our family.

_____ 33. When our family got together, everybody would be present.

_____ 34. Rules would change in our family.

_____ 35. We could easily think of things to do together as a family

_____ 36. We would shift household responsibilities from person to person.

_____ 37. Family members would consult each other on their decisions.

_____ 38. We would know who the leader(s) was in our family.

_____ 39. Family togetherness would be very important.

_____ 40. We could tell who does which household chores.

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Appendix L

Caregiver Evaluation

In your meetings with the nurse, genetic counselor, and social worker or psychologist, you talked about many different topics. For the topics listed below, circle the amount of time you spent talking about each one:

1) How family members viewed sickle cell disease before your child's diagnosis

never very little some a lot always don't know

2) How family members view sickle cell disease since your child's diagnosis

never very little some a lot always don't know

3) Why family members should know that your child's diagnosis of sickle cell disease involves them

never very little some a lot always don't know

4) How much family members know about sickle cell disease

never very little some a lot always don't know

5) How family and friends can help if they know more about sickle cell disease

never very little some a lot always don't know

6) Getting written information to family members

never very little some a lot always don't know

7) Getting family and friends to come to the clinic to get more information

never very little some a lot always don't know

8) How to involve family and friends in the care of your child

never very little some a lot always don't know

9) Getting information from others when needed

never very little some a lot always don't know

10) Getting advice and assistance from others when needed

never very little some a lot always don't know

Appendix M

CAREGIVER INTERVIEW

Interview Date _____ Interviewer_____ Subject #_____

Diagnosis_____

When Diagnosed?: prenatally____ at birth____ age at dx ____
Patient's Date of Birth_____
Length of Time in Hospital Program _____

A. Demographic Information

1. Who lives in your household? (indicate any other family members with sickle cell disease):

Name	Age	Relationship to Patient

Caregiver: _____
Patient: _____
_____
_____
_____
_____
_____
_____

2. What kind of work do you usually do?

3. Are you presently working outside of the home?

4. What kind of work does the baby's father usually do? (if involved)

5. Is he presently working? (If involved)

6. What is your household's approximate monthly income? (AFDC?)

7. What is the last grade you completed in school (and degree)?

8. What is the last grade the baby's father completed in school (and degree)?

9. Who do you consider to be your "family"? The definition of "family" = persons who are important in your life, to whom you are related by blood, marriage, adoption, or friendship, and/or to whom you have made a long term commitment.

Name	Age	Relationship to Patient

(Indicate all "family" members who have sickle cell disease)

B. Family Support

FAMILY SUPPORT SCALE: Listed below are sources that often times are helpful to members of families raising a young child. This questionnaire asks you to indicate how helpful each source is to your family.

Please circle the response that best describes how helpful the sources have been to your family during the past 3 to 6 months. Cross out any sources of help that have not been available to your family during this period of time.

	Not at all Helpful	Sometimes Helpful	Generally Helpful	Very Helpful	Extremely Helpful
1. My parents.....	0	1	2	3	4
2. The child's father's parents....	0	1	2	3	4
3. My relatives/kin.....	0	1	2	3	4

	Not at all Helpful	Sometimes Helpful	Generally Helpful	Very Helpful	Extremely Helpful
4. The child's father's relatives..	0	1	2	3	4
5. Spouse or partner.....	0	1	2	3	4
6. My friends.....	0	1	2	3	4
7. The child's father's friends.....	0	1	2	3	4
8. Other children in my home.....	0	1	2	3	4
9. Other parents.....	0	1	2	3	4
10. Church.....	0	1	2	3	4
11. Social groups/clubs.....	0	1	2	3	4
12. Co-workers.....	0	1	2	3	4
13. Parent groups.....	0	1	2	3	4
14. Family or child's physician..	0	1	2	3	4
15. Professional helpers (social workers, teachers, etc.).....	0	1	2	3	4
16. School/day care center.....	0	1	2	3	4
17. Professional agencies (public health, social services, mental health, etc.).....	0	1	2	3	4
18. Sickle Cell Program at hospital.....	0	1	2	3	4
19. Other (specify).....	0	1	2	3	4

C. Sharing Information

1. Make a list (on the following page) of those people who fall into any of the following categories:

***You have you talked to them about sickle cell disease**

***You have given them written information to read about sickle cell disease**

***They've gone to the hospital or clinic with you to learn more about sickle cell disease**

These may include people who are in your family or people who are not in your family. This list should not include people who work in the sickle cell clinic.

2. How would you rate their understanding about sickle cell disease? (Use the following scale):

5 - Knows an exceptional amount about sickle cell disease and has a very good understanding of the information (what scd is compared to normal blood, inheritance, signs and symptoms)

4 - Has some knowledge about the information covered by the genetic counselors (what scd is, signs and symptoms)

3 - Has knowledge to know what to look for (when to call the doctor, take the baby to the hospital)

2 - Does not know enough to be helpful

1 - Has the wrong information

SHARING INFORMATION

NAME	RELATIONSHIP	Talked to about SCD	Gave written information to	Has been to the hospital or clinic	Knowledge rating

D. Support

1. Type of Support

Now we are going to make a list make a list of all the people from whom you get any type of support. You will then indicate the type of support you have received from them by rating how helpful or supportive they have been on the following scale:

5 - Extremely supportive or helpful
4 - Very supportive or helpful
3- Supportive or helpful
2- Not very helpful
1- Extremely unhelpful
0 - Did not turn to for this

The types of support are defined as follows:

EMOTIONAL SUPPORT - affection, comfort, caring, love, being on your side

ENCOURAGEMENT - praises, compliments, makes you feel important

ADVICE/INFORMATION ABOUT SCD - provides advice and information about scd

ADVICE/INFORMATION ABOUT RAISING CHILDREN - provides advice and information about child-rearing, nutrition, etc.

HELPS WITH PROBLEM-SOLVING - provides advice, information, and assistance when problems come up

SOCIALIZING - spends time with you, does things with you, visits

HELPS WITH FAMILY RESPONSIBILITIES - helps with chores, family responsibilities, babysitting

PROVIDES FINANCIAL ASSISTANCE - loans or gives money when needed

HELPS IN EMERGENCIES - would give a ride to the hospital when the baby is sick, would take care of the other children if you got sick

OVERALL HELPFULNESS - general impression of their helpfulness

TYPE OF SUPPORT

NAME and RELATIONSHIP	Emotional support	Encouragement	Advice/Information about SCD	Advice/Information about raising children	Help with problem solving	Spends time together	Helps with family responsibilities	Financial support	Emergency support	Overall Helpfulness

2. Amount of Support

How would you rate your satisfaction with the amount of support you receive in your life overall?

5 - extremely well satisfied
4- very satisfied
3 - satisfied
2 - dissatisfied
1 - not satisfied at all

E. Ratings of Family Health

For the first interview, answer in terms of current status, or "since diagnosis". For all interviews following the first, answer in terms of "since the last interview".

1) Has any one in the family suffered from any form of physical distress such as headaches, fatigue, loss of weight, etc. (since diagnosis or last interview)?

Yes_____ No_____

1a) Which family member(s)? (indicate relationship to the child)

_____

_____

1b) How distressed is/are the family member(s)?

5 - not at all distressed
4 - a little distressed
3 - somewhat distressed
2 - very distressed
1 - extremely distressed

2) Has any pre-existing illness (e.g. high blood pressure, asthma, arthritis) of any family member worsened (since diagnosis or last interview)?

Yes_____ No_____

2a) Which family member(s)? (indicate relationship to child)

_____

_____

2b) If Yes, to what extent has their condition become worse?

5 - has not worsened at all
4 - has become a little worse
3 - has worsened somewhat
2 - has become much worse
1 - has become extremely worse

3) Has any family member suffered from any emotional problems such as a suicide attempt, depression, or anxiety (since diagnosis or last interview)?

Yes_____ No_____

3a) Which family member(s)? (indicate relationship to child)

_____

_____

3b) Did the family member(s) talk to a professional about the(ir) problem?

Yes_____ No_____

3c) If yes, how bad is their emotional problem?

5 - not at all severe
4 - a little severe
3 - somewhat severe
2 - very severe
1 - extremely severe

4) Has any family member suffered from sleeplessness, irritability, or frustration (since diagnosis or last interview)?

Yes_____ No_____

4a) Which family member(s)? (indicate relationship to child)

_____

_____

4b) If yes, how bad did it become?

5 - not at all bad
4 - a little bad
3 - somewhat bad
2 - very bad
1 - extremely bad

5) Was anybody in the family involved excessively with alcohol or any other drug (since diagnosis or last interview)?

Yes_____ No_____

5a) Which family member(s)? (indicate relationship to child)

_____

_____

5b) How excessive was their use of alcohol or other drugs?

5 - not at all excessive
4 - a little excessive
3 - somewhat excessive
2 - very excessive
1 - extremely excessive

6) Has there been a change in the general climate or mood within the family (since diagnosis or last interview)?

Yes_____ No_____

6a) How noticeable has it been?

5 - not at all noticeable
4 - a little noticeable
3 - somewhat noticeable
2 - very noticeable
1 - extremely noticeable

7) Does your family worry about something else bad happening in the future (since diagnosis or last interview)?

Yes _____ No_____

7a) To what degree is your family worrying about this?

5 - not at all
4 - a little
3 - somewhat
2 - very much
1 - extremely so

8) Have you or your partner stayed away from home, or wanted to stay away from home for long periods (since diagnosis or last interview)?

Yes_____ No_____

8a) How often have either of you stayed away or wanted to stay away?

5 - not at all
4 - rarely
3 - sometimes
2 - often
1 - extremely often

F. Ratings of Burden

21) How burdened is your family by having a family member who has sickle cell disease in the following categories:

Financially___
Emotionally___
Time/Convenience___
Overall___

5 - Not in the least burdened
4 - Not particularly burdened
3 - Somewhat burdened
2 - Very burdened
1 - Extremely burdened

22) By having a family member who has sickle cell disease, how burdened do you feel in each of the following categories:

Financially___
Emotionally___
Time/Convenience___
Overall___

5 - Not in the least burdened
4 - Not particularly burdened
3 - Somewhat burdened
2 - Very burdened
1 - Extremely burdened

23) How do you feel your family is coping with having a member with sickle cell disease?

5 - Coping extremely well
4 - Coping very well
3 - Coping as well as can be expected
2 - Not coping well
1 - Coping extremely poorly

24) How do you feel you are coping with having a family member with sickle cell disease?

5 - Coping extremely well
4 - Coping very well
3 - Coping as well as can be expected
2 - Not coping well
1 - Coping extremely poorly

Feedback/Debriefing

1. Using the page of adjectives as a guide, which emotions best describe how you often feel? (choose as many as you like)

2. What types of things have generally helped you to adjust to your child's diagnosis?

3. What about the sickle cell program has been particularly helpful to you in adjusting to your child's diagnosis?

4. What would you like us to change in our program?

5. How do you feel about this interview?

*****END OF INTERVIEW*****

Appendix N

Summary of Instruments

Instrument	Predictor or Outcome	Standardized?	Administration Who	Administration When	Qualitative or Quantitative?	Reliability & Validity
CAREGIVER INTERVIEW Appendix G CI	P and O	No	Investigator	Before diagnosed child is 5 years old	Both	Not yet available
FAMILY SUPPORT SCALE Part of App G FSS	O	Yes	Caregiver	Before diagnosed child is 5 years old	Quantitative	Coefficient alpha .77 Rel. .75
FACES - III Appendix J	O	Yes	Caregiver	Before diagnosed child is 5 years old	Quantitative	Cronbach's alpha .62 Rel. .75
F-COPES Appendix H	P	Yes	Caregiver	Before diagnosed child is 5 years old	Quantitative	Cronbach's alpha .86 Coefficient alpha .63-.95
FILE Appendix I Form C	P	Yes	Caregiver	Before diagnosed child is 5 years old	Quantitative	Cronbach's alpha .81 Test retest .80 Only reliable if complete form is used
CAREGIVER EVALUATION Appendix K CE	O	No	Caregiver	Before diagnosed child is 5 years old	Quantitative	not yet available

Summary of Instruments

Instrument	Scoring	Theoretical Basis	Variable Measured	Dimensions or Subscales	Items
CAREGIVER INTERVIEW Appendix G C1	Ratings and scaled scores are summed Higher scores yield more desireable ratings	ABC-X Model Bronfenbrenner's Social Ecology Model	Caregiver's perception of: Support Knowledge Burden Coping	Demographic Info: household make-up parent's education SES "family" make-up Sharing Information type of sharing knowledge rating Support type amount Family Health Ratings physical emotional (caregiver) emotional (family) Burden Ratings Feedback	A1-A9 A1 A7-A8 A6 A9 C1 C2 D1 D2 E1-E6 E7-E14 E15-E20 F21-24 G1-G5
FAMILY SUPPORT SCALE Part of App G FSS	Helpfulness index = sum of ratings of items Number of sources of support available = sum of items not rated as n/a Sub category scores can be standardized using adjusted scores	Bronfenbrenner's Social Ecology Model (Operationalized by Dunst)	Support	Sources of Support: informal kinship social groups professionals professional groups formal kinship Types of support: formal informal	6, 7, 9, 12 10, 11, 13 14, 15 16, 17, 18 1-5, 8
FACES - III Appendix J	Total of scores assigned to each item are summed Discrepancy between perceived and ideal = inverse assessment of satisfaction	Circumplex Model of Family Functioning	Psychosocial Adjustment of Families	Cohesion: Disengaged vs. Enmeshed Adaptability: Rigid vs. Flexible	1-5, 7, 9-13, 17, 19 6, 8, 14, 16, 18, 20
F-COPES Appendix H	Sum the circled scores for each item Reverse item #s: 17, 26, 28	ABC - X Model Integrates 2 factors in model: Family resources (factor B) Meaning/perception (factor C)	Coping	Acquiring social support Reframing Seeking spiritual support Mobilizing family to accept help Passive appraisal	1, 2, 5, 6, 10, 16, 20, 25, 29 3, 7, 11, 13 15, 19, 22, 24 14, 23, 27, 30 4, 6, 9, 21 12, 17, 26, 28
FILE Appendix I Form C	Provides an index of family vulnerability Weighted values of all "yes" responses are summed.	ABC - X Model "aA" (Double A) factor or "pile-up" factor Derived from family and psychobiological stress research Pile-up stress depletes family resources	Family Stress	Intra-family strains Marital strains Pregnancy/childbearing strains Finance/business strains Work/family transitions Illness and family care Losses Transitions in/out home Family legal violations	1-17 18-21 22-25 26-37 38-47 48-55 56-61 62-66 67-71
CAREGIVER EVALUATION Appendix K	Cross-site comparison of ratings in each category	Content areas were derived from the enhanced intervention protocol employed at CHO	Strength of the Intervention	Total Support Seeking Family Support Seeking	10 items total

Table 1

Means and Standard Deviations of All Measures

	Enhanced Group			Standard Group		
Measure	N	Mean	S.D.	N	Mean	S.D.
CARESUPPORT						
Total	40	33.0	7.1	27	32.0	7.1
Emotional	40	20.7	4.6	27	20.6	4.0
Material	40	12.4	4.0	27	11.0	3.6
CARECOPE (range 1–5)	40	4.2	0.83	27	4.1	0.78
FAMCOPE (range 1–5)	40	3.8	0.99	27	4.2	0.83
CAREBURDEN Overall (range 1–5)	40	4.2	0.8	27	4.1	0.8
FAMBURDEN Overall (range 1-5)	40	3.8	1.0	27	4.2	0.8
FACES (range 0–80)	34	14.8	10.1	27	12.0	10.5
FILE Total (range 0–3,307)	40	449.8	328.5	27	516.2	309.5
FSS						
Total (range 0– 76)	34	28.3	9.8	27	31.0	10.0
Informal (range 0–14)	34	11.4	0.6	27	11.6	0.7
Formal	34	12.0	0.8	27	12.2	0.7
FCOPES						
Acquiring (range 0–45)	40	27.7	8.1	27	30.0	5.0
Reframing (range 0–40)	40	32.3	6.2	27	34.0	3.3
Spiritual (range 0–20)	40	14.8	3.8	27	15.0	4.0
Mobilizing (range 0–20)	40	13.8	4.0	27	14.4	2.7
Passive (range 0–20)	40	12.8	3.1	27	14.4	2.0
CE Total (range 0–4)	33	2.4	0.73	26	2.1	0.71

Table 2

Demographic Characteristics of Caregivers

Variable	Enhanced Group (N=40) %	n	Standard Group (N=27) %	n
Education				
> 7 years	2.5	1	0	0
J.H.S. Graduate	10.0	4	0	0
Some H.S.	15.0	6	7.4	2
H.S. Graduate	40.0	16	14.8	4
Some College	30.0	12	51.9	14
College Graduate	2.5	1	22.2	6
Graduate/Professional	0	0	3.7	1
Annual Income				
$0–$7,200	20.0	8	7.7	2
$7,200–$15,000	50.0	20	42.3	11
$15,000–$30,000	22.5	9	15.4	4
$30,000–$50,000	5.0	2	26.9	7
>$50,000	2.5	1	7.7	2
Work				
Unskilled	52.5	21	11.1	3
Semiskilled	20.0	8	22.2	6
Skilled	7.5	3	7.4	2
Clerical, Technical	15.0	6	22.2	6
Administrative	0	0	7.4	2
(Executive lesser)	5.0	2	29.6	8
(Executive major)	0	0	0	0
Working Now?				
Yes = 1	17.5	7	66.7	18
No = 2	82.5	33	33.3	9

Table 3

Means and Standard Deviations of Caregiver Network Size

	Enhanced Group (N=40)		Standard Group (N=27)	
Network	Mean	S.D.	Mean	S.D.
Total Network	11.4	6.2	16.4	10.0
Support Network	6.8	4.2	9.4	8.6
Info Sharing Network	6.8	5.6	8.3	5.9
Family Network	10.3	4.7	10.9	6.7
Household Network	4.7	2.1	3.9	1.1

Table 4

Demographic Characteristics of Caregiver Network Type

	Total		Support		Sharing		Family		Household	
Group	n	%	n	%	n	%	n	%	n	%
Enhanced (N=40)										
Blood Nuclear	0	–	0	–	0	–	0	–	22	55.0
Blood Extended	11	27.5	13	32.5	14	35.0	16	42.1	12	30.0
Non Blood Extended	0	–	1	2.5	2	5.0	1	2.6	0	–
Mixed Extended	18	45.0	18	45.0	16	40.0	21	55.3	5	12.5
Formal/Foster	11	27.5	8	20.0	8	20.0	0	–	1	2.5
Standard (N=27)										
Blood Nuclear	0	–	0	–	0	–	2	7.4	17	63.0
Blood Extended	3	11.1	4	14.8	4	14.8	13	48.1	10	37.0
Non Blood Extended	0	–	0	–	0	–	0	–	0	–
Mixed Extended	16	22.2	16	22.2	10	37.0	11	40.7	0	–
Formal/Foster	18	66.7	17	63.0	13	48.1	1	3.7	0	–

Table 5

Preliminary Group Comparisons Using the Kruskal-Wallis Chi Square Test

Variable	N	H	DF	P
Caregiver Work Status	66	16.7	1	0.001
Caregiver Education	67	15.2	1	0.001
Caregiver Work	67	15.8	1	0.001
Income	66	5.1	1	0.025
Patient Sex	67	0.2	1	n.s.

Table 6

Preliminary Group Comparisons Using Mann-Whitney Test

	Enhanced			Standard			Standardized U	
Variable	N	Mean	S.D.	N	Mean	S.D.	Z	P
Program Time	37	13.3	17.0	23	30.0	17.4	-3.7	0.002
Caregiver Age	40	29.3	11.4	27	31.0	6.4	-1.7	n.s.
Network Size	40	11.4	6.2	27	16.4	9.8	-2.2	0.026

Table 7

Group Comparison on the Intervention (Hypothesis 1)

Test	Enhanced N	Enhanced Mean	Enhanced S.D.	Standard N	Standard Mean	Standard S.D.	Standardized U Z	Standardized U P
Caregiver Evaluation								
Family Involvement	34	2.3	1.0	27	1.9	0.9	1.40	n.s.
Total (Active Coping)	33	2.4	0.7	27	2.1	0.7	1.36	n.s.
Functioning Measures								
FAMCOPE	40	3.8	1.0	27	4.2	0.8	-1.55	n.s.
CARECOPE	40	4.2	0.8	27	4.1	0.8	0.42	n.s.
SATISPORT	40	4.1	1.2	27	3.9	0.8	0.97	n.s.
FACES	34	14.8	10.1	27	12.0	10.5	1.24	n.s.
F-COPES								
Acquiring	40	27.7	8.1	27	29.6	5.1	-1.04	n.s.
Reframing	40	32.3	6.2	27	33.6	3.3	-0.31	n.s.
Spiritual	40	14.8	3.8	27	14.7	3.6	0.36	n.s.
Mobilizing	40	13.8	4.0	27	14.4	2.7	0.03	n.s.
Passive	40	12.8	3.1	27	14.4	2.2	-2.14	p<.03

Table 8

Pearson Correlations (r) For Support Variables (Hypothesis 2)

			FCOPES						
	CARECOPE	FAMCOPE	Acquiring	Reframing	Spiritual	Mobilizing	Passive	FACES	SATISPORT
FSS									
Total	0.41[c]	0.26	0.12	0.04	0.16	0.002	0.13	-0.20	0.41[c]
Informal	0.35[b]	0.23	0.20	0.05	0.10	-0.06	0.03	-0.22	0.38[b]
Formal	0.30[a]	0.18	0.003	-0.02	0.20	0.10	0.25	-0.06	0.22
CARESUPPORT									
Total	-0.03	0.13	0.21	0.36[b]	0.21	0.05	-0.10	-0.27	0.25
Emotional	-0.16	0.02	0.09	0.27	0.13	0.08	0.24	-0.22	0.11
Material	0.13	0.22	0.26	0.32[b]	0.20	-0.03	0.10	-0.20	0.32[b]
Size									
Household	0.28	0.09	0.21	0.03	0.17	0.11	0.33	-0.04	0.30[b]
Family	0.25	0.10	0.10	0.05	0.16	-0.06	0.16	-0.07	0.32[b]
Support	0.23	0.08	0.31[a]	0.02	0.20	0.11	0.24	-0.08	0.29[a]
Network	0.20	0.10	0.21	0.03	0.17	0.11	0.33[b]	-0.02	0.30[b]

[a]$p<0.05$

[b]$p<0.01$

[c]$p<0.001$

Table 9

Pearson Correlations (r) For Stress Variables (Hypothesis 3)

			FCOPES						
	CARECOPE	FAMCOPE	Acquiring	Reframing	Spiritual	Mobilizing	Passive	FACES	SATISPORT
FILE									
Total	-0.03	-0.24	-0.10	-0.03	0.27	0.22	0.006	0.26	-0.32[b]
Financial	-0.25	-0.15	0.20	-0.10	0.30[b]	0.16	0.10	0.03	-0.25
Marital	0.04	-0.38[b]	0.19	0.16	0.21	0.21	-0.12	0.18	-0.06
Intra Family	-0.03	-0.18	-0.15	-0.06	0.15	0.20	0.005	0.30[a]	-0.36[b]
Illness	0.27	0.10	-0.10	-0.24	-0.07	-0.03	-0.09	0.19	0.31[b]
Pregnancy	-0.03	-0.21	0.07	0.09	0.08	0.10	-0.12	0.07	-0.16
CAREBURDEN									
Financial	0.03	0.31[c]	0.11	0.28	0.17	-0.13	0.08	-0.22	0.05
Emotional	0.15	0.14	0.08	0.26	0.13	0.05	0.23	-0.08	0.14
Time	-0.003	0.01	0.05	0.19	0.18	0.001	0.21	-0.04	0.13
Overall	0.16	0.08	-0.02	0.22	0.03	0.01	0.22	-0.07	0.006
FAMBURDEN									
Financial	0.06	0.21	-0.11	0.29	-0.17	-0.13	0.08	-0.03	-0.06
Emotional	0.07	0.24	-0.04	0.14	-0.15	-0.12	0.11	-0.01	0.07
Time	-0.11	0.26	-0.08	-0.05	-0.10	-0.19	0.17	0.04	0.08
Overall	0.01	0.24	-0.08	0.05	-0.06	-0.05	0.15	-0.03	-0.03

[a]$p<0.05$

[b]$p<0.01$

[c]$p<0.001$

Table 10

Sets of Variables Used in the Multiple Regression Analyses

Dependent (Outcome) Variables
FAMCOPE – Perceived level of family coping (range 1–5, higher score is better)
CARECOPE – Perceived level of caregiver coping (range 1–5, higher score is better)
FACES – Caregiver satisfaction with family functioning (range 0-80, lower score is better)
Independent (Predictor) Variables
Block 1: Resources
Diagnosis (0=SS, 1=other)
Income (1=<7,200, 2=<15,000, 3=<30,000, 4=<50,000, 5=>50,000)
Education (1=Graduate/Professional, 2=College Graduate, 3=Some College, 4=H.S. Graduate, 5=Some H.S., 6=Junior H.S. Graduate, 7=<7 years of education)
Patient Age – Age of diagnosed child at the time of interview (range 0–65 months)
Block 2: Life Stress
FILE Total – Total life stress affecting family over previous year (range 1–3,307, lower score is better)
CAREBURDEN – Overall score on perceived caregiver burden (range 1–5, higher score is better)
FAMBURDEN – Overall score on perceived family burden (range 1–5, higher score is better)
Block 3: Support
FSS Total – Sources of support helpful to family (range 0–76, higher score is better)
CARESUPPORT – Caregiver ratings of specific types of support (range 0–5, higher score is better)
Network Size – Total members in extended kinship network
Household Size – Total members living in household
Block 4: Active Coping
F–COPES (FC) Subscales:
FC Acquiring Social Support – Ability to actively seek support from network (range 0–45, higher score is better)
FC Reframing – Ability to reframe problems in positive terms (range 0–40, higher score is better)
FC Seeking Spiritual Support – Ability to get support through spiritual means (range 0–20, higher score is better)
FC Mobilizing Family to Acquire/Accept Help – Ability to seek out resources and accept help from others (range 0–20, higher score is better)
FC Passive Appraisal – Use of passive coping strategies (range 0–20, lower score is better)
Block 5: Intervention Strength
Caregiver Evaluation Total – Evaluation of enhanced program (range 0–4, higher score is better)

Table 11

Summary of Multiple Regression Analysis Predicting Family Coping (FAMCOPE)

		Step Change			Total Equation			Bivariate (Zero Order) Correlation
Step	Variable	Step	Beta	P	Total	R^2 Total	Adj. R^2 P	r
Block 1: Resources								
	Diagnosis							0.07
	Income							0.21
1	Education	0.081	-0.284	0.029	0.081	0.065	0.029	-0.28[a]
	Patient Age							0.05
Block 2: Life Stress								
	FILE Total							-0.24
	FAMBURDEN	No Variables Entered			All P>0.05			0.18
	CAREBURDEN							0.03
Block 3: Support								
2	FSS Total	0.063	0.256	0.047	0.144	0.113	0.013	0.30[a]
	CARESUPPORT							0.17
	Network Size							0.09
	Household Size							0.11
Block 4: Active Coping								
	FC Acquiring							-0.14
	FC Reframing							-0.02
	FC Spiritual							-0.26
3	FC Mobilizing	0.161	-0.413	0.001	0.305	0.267	0.001	-0.34[b]
	FC Passive							0.19
Block 5: Intervention								
	CE Total	No Variables Entered			All P>0.05			0.24

[a]p<0.05 [b]p<0.01

Table 12

Summary of Multiple Regression Analysis Predicting Caregiver Coping (CARECOPE)

		Step Change			Total Equation			Bivariate (Zero Order) Correlation
Step	Variable	Step	Beta	P	Total	R^2 Total	Adj. R^2 P	r
Block 1: Resources								
	Diagnosis							0.05
	Income	No Variables Entered			All P>0.05			0.21
	Education							-0.24
	Patient Age							0.03
Block 2: Life Stress								
	FILE Total							-0.04
	FAMBURDEN							0.10
1	CAREBURDEN	0.067	0.258	0.049	0.067	0.050	0.049	0.26[a]
Block 3: Support								
2	FSS Total	0.142	0.381	0.003	0.208	0.180	0.003	0.41[b]
	CARESUPPORT							-0.04
	Network Size							0.28
	Household Size							0.20
Block 4: Active Coping								
	FC Acquiring							0.13
	FC Reframing							0.02
	FC Spiritual							0.07
	FC Mobilizing							0.10
3	FC Passive	0.086	0.302	0.012	0.294	0.256	0.001	0.37[b]
Block 5: Intervention								
	CE Total	No Variables Entered			All P>0.05			0.21

[a]p<0.05 [b]p<0.001

Table 13

Summary of Multiple Regression Analysis Predicting Family Functioning (FACES)

		Step Change			Total Equation			Bivariate (Zero Order) Correlation
Step	Variable	Step	Beta	P	Total	R^2 Total	Adj. R^2 P	r
Block 1: Resources								
	Diagnosis							0.14
1	Income	0.083	-0.288	0.027	0.083	0.067	0.027	-0.29[a]
	Education							0.09
	Patient Age							-0.08
Block 2: Life Stress								
	FILE Total							0.23
	FAMBURDEN	No Variables Entered			All P>0.05			-0.04
	CAREBURDEN							-0.10
Block 3: Support								
	FSS Total							-0.19
2	CARESUPPORT	0.097	-0.313	0.013	0.180	0.151	0.004	-0.28[a]
	Network Size							-0.04
	Household Size							0.17
Block 4: Active Coping								
	FC Acquiring							0.02
	FC Reframing							-0.07
	FC Spiritual	No Variables Entered			All P>0.05			-0.07
	FC Mobilizing							0.19
	FC Passive							-0.99
Block 5: Intervention								
	CE Total	No Variables Entered			All P>0.05			-0.13

[a]p<0.05

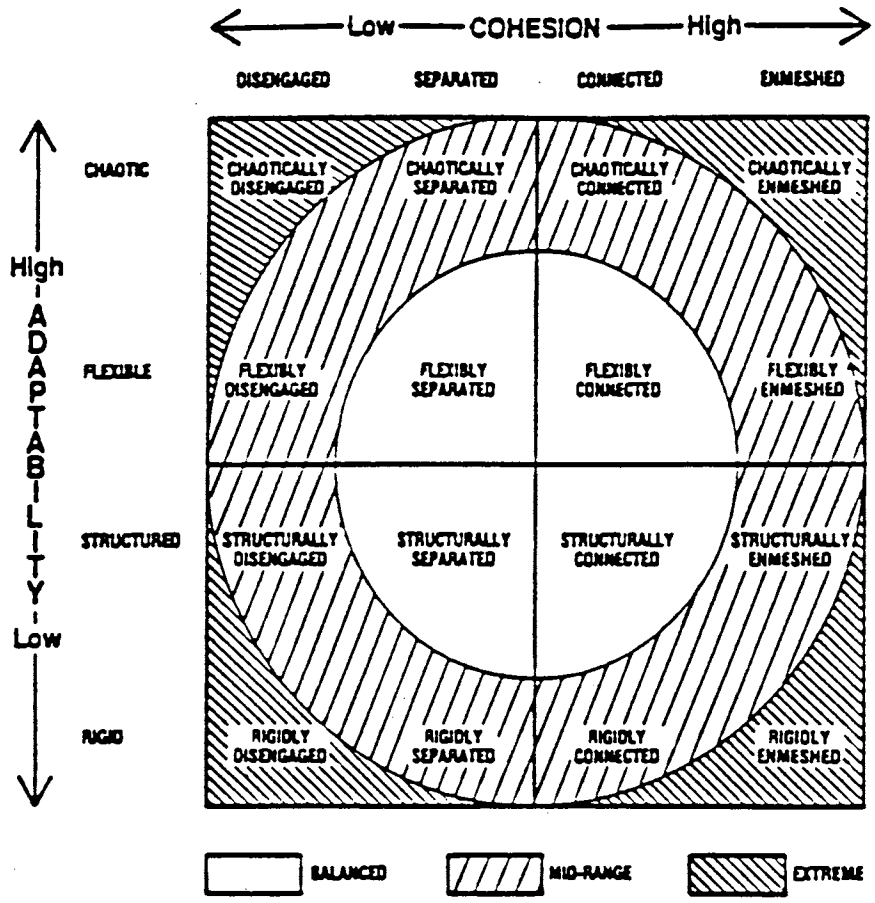

Figure 1. Sixteen possible types of marital and family systems derived from the circumplex model. (From Olson, Russell and Sprenkle (1983). Circumplex Model of Marital and Family Systems: VI. Theoretical Update, Family Process, 22, pp. 69-83.)

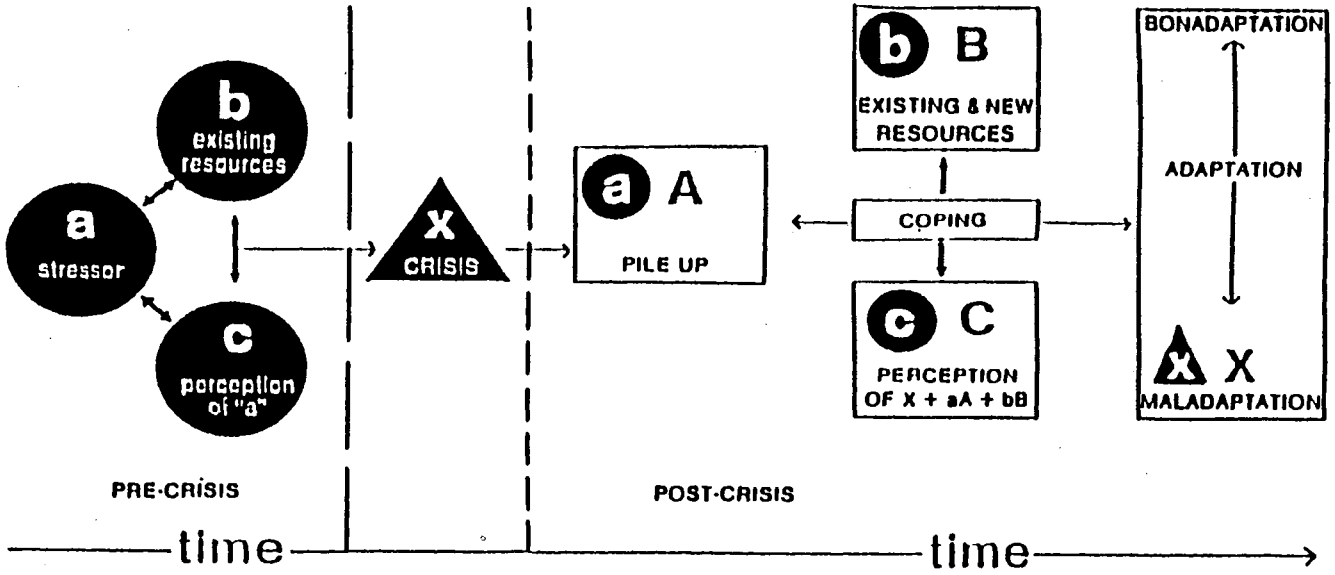

Figure 2. The Double ABC-X Model (FromMcCubbin, H. and Patterson, J. (1983). The Family Stress Process: The Double ABC-X Model of Adjustment and Adaptation, in H McCubbin, M.Sussman, & J. Patterson (Eds.), Social Stress and the Family: Advances and Developments in Family Stress Theory and Research (pp.). New York: The Haworth Press, Inc.)

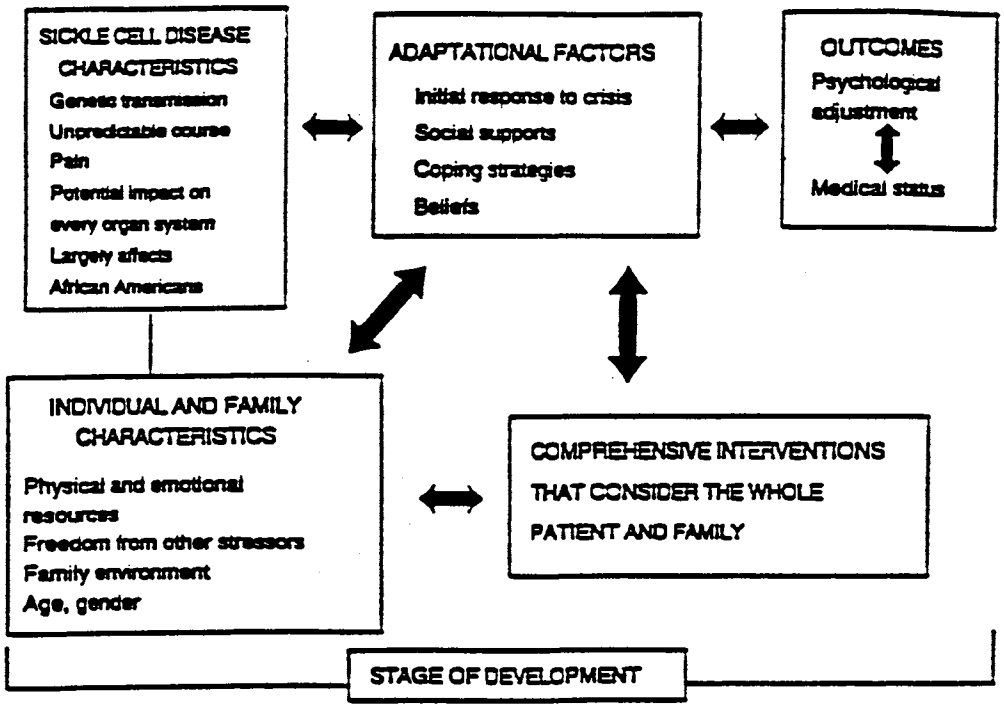

Figure 3. Stress amd adaptation model. (From Treadwell and Gil (1994). Psychosocial aspects. In S.H. Embury, R. Hebbel, N. Mohandas, & M. Steinberg (Eds.) Sickle Cell Disease: Scientific Principles and Clinical Practice pp. 517-529). New York: Raven Press.)

VITA

Genée D. Jackson

Candidate for the Degree of

Doctor of Philosophy

Thesis: FAMILY COPING AND HOSPITAL-BASED EXTENDED KINSHIP NETWORK INTERVENTIONS WITH CAREGIVERS OF CHILDREN DIAGNOSED WITH SICKLE CELL DISEASE

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OKLAHOMA STATE UNIVERSITY
INSTITUTIONAL REVIEW BOARD
FOR HUMAN SUBJECTS RESEARCH

Proposal Title: INTERVENTIONS WITH EXTENDED FAMILY MEMBERS: IMPACT ON COPING IN FAMILIES OF PATIENTS WITH SICKLE CELL DISEASE

Principal Investigator: B. HELM, G.D. JACKSON

Date: FEBRUARY 16, 1992 IRB # AS-92-028

This application has been reviewed by the IRB and

Processed as: Exempt [XX] Expedite [ ] Full Board Review [ ]

Renewal or Continuation [ ]

Approval Status Recommended by Reviewer(s):

Approved [XXX] Deferred for Revision [ ]

Approved with Provision [ ] Disapproved [ ]

Approval status subject to review by full Institutional Review Board at next meeting, 2nd and 4th Thursday of each month.

Comments, Modifications/Conditions for Approval or Reason for Deferral or Disapproval:

PROVISIONS RECEIVED 7/20/92

Signature: Marcia L. Tilley Date: 7-20-92

Chair of Institutional Review Board