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FAMILY FUNCTIONING AND CHILD RESILIENCE AMONG CHILDREN WITH PHENYLKETONURIA

by

SHAUNE BORNHOLDT

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

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Abstract

FAMILY FUNCTIONING AND CHILD RESILIENCE

AMONG CHILDREN WITH PHENYLKETONURIA

by

Shaune Bornholdt

Advisor: Professor Vera S. Paster

A family systems version of Aaron Antonovsky's salutogenic model of stress management was applied in a study of 34 families of children with phenylketonuria (PKU), a metabolic disorder requiring a special diet to prevent mental retardation. The model predicts that individuals who perceive their familial/social world as coherent will manage tension in ways that promote health.

Two family strengths, (1) family sense of coherence, or mothers' perception that for the family the social world is comprehensible, manageable, and meaningful, and (2) family practice of regular routines, were correlated with one another and with child outcomes. A significant positive correlation was found between the two strengths. As a single predictor, mothers' perceived family sense of coherence correlated significantly and positively with measures of child psychosocial resilience and dietary adherence. Considered along with family social position, a strong predictor of adherence, sense of coherence retained significant association with measures of psychosocial resilience, and showed a trend in the predicted direction in

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its association with dietary adherence. As a single predictor, family practice of routines also correlated significantly and positively with child psychosocial resilience, but did not correlate significantly with dietary adherence. When considered with other variables, family practice of routines showed a trend towards association with the social skills variable.

The two family strengths were further examined in diet-related terms in interviews of 14 mothers. Mothers' descriptions of their experience reflected their measured family sense of coherence. Good adherence was associated with maternal belief in the PKU diet as comprehensible, manageable, and meaningful. Mothers of adherent children engaged them in diet-related activities, integrated the diet across social realms, and imbued these experiences with positive feeling. They were also better able to make use of PKU clinic support than were mothers of poorly adherent children.

Findings support the salutogenesis hypothesis that sense of coherence promotes good outcomes. Findings are also consistent with representational theories of internalization. The strength of influence of family social position on health outcome highlights the need for social outreach in this population.

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Family Functioning and Child Resilience among Children with Phenylketonuria

CHAPTER ONE

INTRODUCTION

What are the aspects of family functioning that contribute to the physical and psychosocial resilience and vulnerability of chronically ill children? In the past decades, advances in treatment have increased longevity for many such children and have brought about better health outcomes, reducing and in many cases eliminating the need for long term hospitalization and institutional care. Thus, more families are caring for their chronically ill children at home. Such families often bear the chief burden of implementing their children's medical treatment. They are called upon to cope with complicated medical and dietary regimens while attending to their children's social and emotional needs, the needs of other family members, and the daily and long term demands of family and social life.

Vulnerability and Resilience In Chronically Ill Children

While many of these children under outpatient and family care fare well, others do not. For example. compliance with prescribed pediatric regimens has been estimated at 50% (Litt and Cuskey, 1980), with problems in

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adherence greater for long-term as compared with short-term regimens (Sackett and Snow, 1979; Liptak, 1996). Health outcomes already compromised by the disease process are further affected by noncompliance with treatment.

The research picture of the children's psychosocial functioning is mixed. Some researchers have found that the children are at risk for poor psychosocial adjustment, vulnerable to problems in behavior, learning, self-esteem, social competence, and ego development (Pless, Roghmann and Haggerty, 1972; Satterwhite, 1978; Wallander, Varni, Bambani, Banis and Wilcox, 1988; Hauser et al., 1992). Others report few differences between chronically ill children and carefully matched healthy peers (Lavigne and Faier-Routman, 1993), and one reviewer concluded that "the generally positive psychosocial adaptation such youngsters display, in the face of very difficult life situations, has been quite impressive." (LaGreca, 1990, p. 287).

What is clear is that there is considerable variation in treatment outcomes on the physical and psychosocial dimensions influenced by family care within populations of chronically ill children. There is a need to account for these variations. We have much to learn not only from those families and children who do poorly, but from those who do well--the families and children who do adhere to treatment, the children whose health outcomes are optimal within the limiting terms of their illness, children who, in terms of

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their socioemotional development, are normal and in some cases thriving.

In recognition of this need, ecological systems models of health and illness have placed family functioning at the center of a bi-directional dynamic that has been studied from two main perspectives. On the one hand, the child's illness may be seen as a major stressor to the family, to which the family adapts more or less successfully through a restructuring of roles and routines, and a reallocation and mustering of economic, temporal, social, spiritual, and psychological resources. On the other hand, the family's functioning affects that of the chronically ill child, both directly by, for example, fostering or inhibiting prosocial behavior and treatment compliance, and indirectly by mediating the stresses experienced by the child, thereby intensifying, diminishing, or transforming their effect.

Sense of Coherence and Family Routines as Protective Factors

It is the second perspective, the influence of the family on the range of child functioning, that is the focus of the present study. A family systems model of the transmission of resilience is proposed as a means of accounting for differences in outcomes among chronically ill children. Based on the work of two sociologists, Aaron Antonovsky's "salutogenic" or health-generating theory of prevention of breakdown and promotion of health (Antonovsky,

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1979, 1987), and David Reiss' theory of the family's construction of social reality (Reiss and Oliveri, 1980; Reiss, 1981), the model posits family sense of coherence, a continuum ranging from weak to strong, as its central organizing construct. According to this model, individuals who perceive their family sense of coherence towards the strong end of the continuum have internalized a dynamic hope, a confidence in the relative predictability of internal, interpersonal, and social experience. These individuals have formed a pervasive and enduring belief that, for the family, the social world and its challenges are relatively comprehensible, manageable, and meaningful.

The model holds that the sense of coherence has motivating power, both for the family as a whole and for its members. Faced with difficult life demands, members of a family with a strong sense of coherence will seek to bring information and meaning into the system, secure in the belief that through their collective efforts the family can find ways to cope and to gain needed support from the social world. Such a family can selectively make the most of available resources of many kinds, thereby successfully managing tension in a way that is salutogenic--a way that further reinforces the sense of coherence and slows, prevents, or even reverses health breakdown. (Antonovsky, 1979, 1987; Sourani, 1983)

A major premise of the model is that the family sense of coherence is a pervasive dispositional orientation

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towards the world. It is shared by family members, and motivates perceptions and actions. According to the model as proposed in the present study, family sense of coherence, because of its globality and pervasiveness, looks both outward and inward. Looking outward, the family sense of coherence permeates the interactions between the family and larger social environment. Looking inward, family sense of coherence informs the simple repeated interactive patterns within family life, such as the enactment of everyday family rituals and routines.

The model holds that families with a strong sense of coherence, when faced with a challenge, will relate adaptively to the social surround. It follows that among families with chronically ill children, those with a strong sense of coherence are likely to seek and use information and support in service of a commitment to treatment, forming an efficacious working alliance with the medical team with regard to treatment adherence.

The model also holds that the family's abiding conception of the social world is reflected and enacted in the quality of the family's daily rituals and routines (Boyce, Jensen, James, and Peacock, 1983; Fiese and Sameroff, 1992; Reiss, 1981;). It follows that families with a strong sense of coherence whose conception of the world includes commitment to the child's treatment will be likely to integrate positive adherence behaviors and attitudes with enactment of their daily family routines and rituals.

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Insofar as the participatory learning implicit in such interactive routines draws the children into the families' felt vision of the world, children in families with strong sense of coherence will share in the positive adherence behaviors and attitudes towards illness, and in the family's overall sense of coherence, with concomitant adaptive engagement in the social world. Therefore, chronically ill children in families with a strong sense of coherence will show greater resilience under the stress of their illness than will children from families in which the sense of coherence is weak. Children in families with strong sense of coherence will be more likely to adhere to treatment, to have better health outcomes, and enjoy better psychosocial adjustment.

Advantages of a Family Systems Model of Resilience

An underlying assumption of this model is that the resilience of the children is fostered through family members'--and especially through parents'--belief that the family coherence is strong. The adults' positive attitudes and effective actions that are motivated by the family sense of coherence are internalized by the children in the context of family interactions.

This family systems model of the transmission of resilience has theoretical and clinical advantages over other models of stress and coping. First, in its focus on

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family strengths, it provides a framework for explaining and fostering good outcomes, rather than simply explaining and preventing poor ones, the focus of most past research. Second, the model provides a means of reconciling contradictory findings, as when one study finds that tight parental control and another finds parental fostering of child autonomy correlate with better treatment adherence. In each case, families with strong sense of coherence may have been more capable of evaluating various contextual factors, selectively choosing the form of family organization most adaptive to the particular situation. Third, in its emphasis on the family's characteristic style of adapting to and negotiating with the social environment, the model brings the social context into its central construct while maintaining, as some ecological models do not, a strong motivational family-based explanatory principle. Fourth, in describing and predicting a given family's likely mode of response to the clinical setting, the model provides information that may help the clinic team respond more sensitively to family style. Finally, the model specifies a simple, observable phenomenon, the repetitive patterned interactions of family life, such as bed time and family meal routines and rituals, as a means by which families transmit their view of the social world to their children (Boyce et al., 1983; Reiss, 1981; 1989). For clinicians, family routines and rituals may provide a simple and powerful means for observing and understanding family

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function, and a promising point of entry for the effectance of change.

Thus far, there have been a few empirical studies linking the family's view of the social world and a few linking family routines to health outcomes among chronically ill children (Boyce, Jensen, Cassel, Collier and Ramsey, 1977; Keltner, 1992; Reiss and Oliveri, 1980;). No study of children's health outcome, however, has examined both family variables together. In addition, most researchers agree that truly resilient children are those who, despite adversity, thrive not in just one but in many areas at once, physical, social, emotional, academic; yet studies of family influence on chronically ill children tend to focus on one realm of child outcome, while the present study looks at several. The present study looks at two poles of what might be called the family code or paradigm (Reiss and Oliveri, 1980, Reiss, 1989), family sense of coherence or general conception of the world, and family routines or the family's enactment of particular behaviors. The study then relates these two variables to dimensions of resilience in children who endure the significant stress associated with a treatable but serious chronic illness.

The Population Studied:
Children with Phenylketonuria

In order to examine relations among these variables most clearly, certain population criteria make sense.

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For the family, the effects of the child's illness and treatment should be perceived as a significant stressor, but the illness should be amenable to treatment in some measure, through the shared efforts of the family and clinic team. The illness should require regular communication and a close treatment alliance between family and clinic, but treatment itself should be largely implemented by the family. The effects of the illness and its treatment should be implicated in a range of attitudes and actions within the family, including the enactment of family routines, and should also affect the child and family's interactions with the larger social environment.

For the child, the effects of the illness and its treatment should be experienced potentially as stressful, and as similarly culturally embedded and extended. In keeping with current thought that considers resilience as a multi-faceted response, the illness and/or its attendant stress should have the potential to affect developmental competencies and functioning across physical, behavioral, cognitive, and psychosocial realms.

The population selected for this study, children with the rare genetic disorder phenylketonuria (PKU), meets these criteria. PKU is a metabolic disorder in which phenylalanine, an amino acid constituent of protein, cannot be metabolized to tyrosine, a precursor of the neurotransmitter dopamine. Children with PKU must follow a highly restricted diet from birth in order to prevent

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irreversible brain damage and severe mental retardation. The effects are most severe and most nearly universal for all children through age six or seven years, but while some older children who are "off diet" have seemingly minor or unidimensional impairment, or even no apparent impairment, others over time may suffer seizures, attentional problems, mood swings, behavior problems, and measured declines of intelligence of more than two standard deviations. Accordingly, the current medical recommendation is for individuals with PKU to stay on the diet for life.

The children get most of their nutrients from a liquid medical food mixed from a powder that has an odd odor (and, to some, an unpleasant texture and taste) that must be consumed in prescribed substantial amounts several times a day. There are severe restrictions on most other foods, but foods containing small "equivalents" of phenylalanine must be eaten in prescribed amounts; nearly everything must be weighed and measured. Changes in the diet are decided upon by the medical team, based on recorded food intake, developmental factors, and the child's phenylalanine levels as measured by frequent blood tests that are painful.

Families and children often find the treatment regimen stressful and many report having difficulty integrating the diet with their daily routines and normal social life. Advance planning, with cooperation of extended family, friends and school personnel may be needed to ensure that the diet is followed. Close cooperation with the medical

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team is essential to planning, but actual treatment is behavioral (following the diet), and is implemented entirely by the family. Thus, the success of treatment is largely dependent upon the family's and child's actions.

When the diet is rigorously followed from birth, and ideally for life, development is normal or nearly so; thus, reasonable hope as a motivating force has a basis in reality. Despite this justifiable hope, the potential for anxiety and stress among parents of children with PKU is high. Immediate effects of high phenylalanine levels may at first be invisible, and long term effects are insidious. Parents differ in their response to these characteristics of PKU. Even though they know that it is prolonged elevation of phenylalanine that is associated with lasting cognitive effects, parents who comply closely with the treatment often worry over the occasional high phenylalanine levels that occur when the children are ill or are growing rapidly, especially when it takes more than one diet change to regain control. Parents who are less compliant may dismiss effects they cannot readily see, but they too may feel nagging anxiety, and they often endure the stress of more frequent interventions by the medical team. For many children, long term poor compliance is associated with cognitive decline and learning difficulties, outcomes that create further stress for the children and their families. Thus, the both the metabolic disorder and the psychosocial aspects of its

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treatment may cause stress for the family and may affect the children adversely.

Many of the children following the diet regimen do quite well, with a range of functioning equal to that of their unaffected peers. However, when group comparisons are made, children with early and continuously treated PKU have been reported to have slightly lower intelligence (Dobson, Kushida and Williamson, 1976), more internalizing (but fewer externalizing) behavioral problems (Burgard, Armbruster, Schmidt, and Rupp, 1994), learning and attentional difficulties (Waisbren, Brown, de Sonneville, and Levy, 1994), and lower levels of social competency (Kazak, Reber, and Snitzer, 1988). In short, children with treated PKU meet the population criteria outlined above for investigating family correlates of vulnerability and risk under a condition of stress, within the context of a family systems salutogenic model of resilience.

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STATEMENT OF PURPOSE

Antonovsky's salutogenic hypothesis proposes that individuals with a positive view of the social world and their place within it are able to manage tension in ways that promote health. This study of families of children with chronic illness presents a family systems model of salutogenesis. It describes how family sense of coherence or the degree to which a family sees the social world as comprehensible, manageable, and meaningful, may be related to the quality of the family's everyday routines and to the family's interactions with others. The study then investigates whether family sense of coherence correlates positively with family adherence to everyday routines, and the degree to which each of these two variables contributes to resilient outcomes among children with the serious but treatable chronic illness, phenylketonuria.

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CHAPTER TWO

LITERATURE REVIEW

Resilience is usually defined as the capacity to do well despite adversity. Reflecting a shift in recent years away from a pathogenic model of human behavior in which researchers sought antecedents of negative outcomes, research on resilience is part of a larger "wellness" (Cowen, 1994) or "salutogenic" (Antonovsky, 1979, 1987) model that seeks antecedents or correlates of health. In the study of stress-resilient children, sometimes termed "invulnerable" or "hardy", neither the severity of the stressors nor the extent of their effect is minimized, but the focus is on successful adaptation.

Family Correlates of Resilience: The Larger Framework

It has now been nearly two decades since Michael Rutter first decried a "regrettable tendency to focus gloomily upon the ills of mankind and all that can and does go wrong" to the exclusion of "what goes right" and, more important, to the exclusion of identifying and fostering those factors that protect and support what goes right among children who have experienced severe psychosocial stress. (Rutter, 1979, p. 49). His findings, repeatedly verified by others, remain fresh, direct, and rich in clinical implication.

In a series of archival studies, Rutter, a British epidemiological psychologist with a strong interest in

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primary prevention, found that the serious psychosocial stressors affecting urban children, such as poverty, marital discord, parental criminality and mental illness, potentiated one another; that is, their harmful effects on child function were synergistic, with the combination of chronic stresses producing a much greater effect than the sum of the separate stresses considered individually. When in addition to the environmental factors, there were risk factors in the child, such as a genetic defect or low self esteem, there was a kind of double synergy, rendering the child especially vulnerable to, and further potentiating, the already interacting environmental factors, an effect Rutter called, after Sameroff (1975), transactional.

Rutter then looked at the obverse side of this dispiriting account, seeking out subsets of children who were doing well psychosocially and academically despite high environmental risk, looking for the specific environmental and personal attributes that might protect children from stress and, possibly, interact to promote resilience and mental health. Based on his own work and a review of that of others, Rutter detailed eight variables likely to be conducive to resilient outcomes and high function in children: "the patterning of stresses, individual differences caused by both constitutional and experiential factors, compensating experiences outside the home, the development of self-esteem, the scope and range of available opportunities, and appropriate degree of environmental

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structure and control, the availability of personal bonds and intimate relationships, and the acquisition of coping skills." (Rutter, 1979, p. 70). Although many of these factors are outside the family or are specific to the child, within a transactional framework all may be mediated by the family.

Continuing in the tradition laid out by Rutter and others, Garmezy (1983), in work with American mid-Western school children subject to multiple environmental stressors, identified a triad of factors leading to adaptive outcomes among children under stress: (a) a socially responsive and adaptive temperament; (b) supportive family relationships, and (c) availability of extended family and other adults. These conclusions are founded upon multiple longitudinal studies involving several different populations of children, conducted under the name Project Competence. Garmezy's group also specified the methodological strategies that have since been employed in studies of resilience, delineating compensatory, challenge, and immunity versus vulnerability models of the relation between the presence of protective factors and child outcomes under stress (Garmezy, Masten, and Tellegen, 1984).

Similar family milieu correlates of child resilience have been found in numerous studies over the current decade by investigators in the Rochester Child Resilience Project (Cowen, Wyman, Work and Parker, 1990; Work, Cowen, Parker and Wyman, 1990; Wyman, Cowen, Work and Parker, 1991; Cowen

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et al., 1992; Cowen, 1994). Stress in this project was defined in terms of traumatic life events experienced by these urban children, such as violence, poverty, family turmoil, separation, illness and death (Work et al., 1990). Among children experiencing more than four major stressful life events or circumstances, groups of "stress resistant" and "stress affected" children were identified, based on teacher report and multiple measures of child adjustment. Summarizing findings of family milieu correlates of resilience in these urban children, Wyman and colleagues state that, compared with parents of children affected by stress, parents of children resistant to stress scored higher on variables reflecting "parent perceptions of a nurturant caregiver-child relationship and self-views as effective caregivers, in the context of positive discipline practices, a child's positive early temperament, and support for primary caregivers." (Wyman et al., 1991).

Of particular interest, in the light of the transactional model of family resilience that the present study proposes, is the importance of the parents' self view as effective caregivers, and the fact that family variables in the Rochester study are identified through parent interviews, and thus reflect the parents' reported perceptions of their familial and social world. These parents, in highly stressful circumstances, have strengths that enable them to represent and interpret many aspects of life in a positive manner. In the socio-cultural world,

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attitudes influence actions, and as they are interpreted these actions "pull for" certain kinds of responses by the social surround. In the transactional framework of mutually interacting systems, the family may exert a strong direct and indirect effect even on those factors seemingly outside the realm of family-based perceptions, such as the compensating experiences outside the home that Rutter lists. A family that sees itself as effective--in the terms of this study, a family with a strong sense of coherence--may be more alert to such compensating experiences, and may therefore be better able to encourage the child to make use of them.

In general, these researchers in community psychology agree as to the sources of strength among resilient children, and as to the importance of family perceptions and attitudes. They also agree on the general definition of child resilience itself, though they differ in their methodological approach to its measurement. Rutter, for example, frequently cites children's good school performance and freedom from behavioral problems as indices of resilience, usually describing individual studies that relate multiple protective predictive factors to just one or two child outcome variables, each considered singly. For the researchers at Project Competence and the Rochester Child Resilience Project, resilience is usually defined in terms of age and stage specific competencies in social, school, and cognitive realms, as well as through global teacher and

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parent ratings of child adjustment (Garmezy, 1983; Wyman et al., 1991). For these researchers, children are categorized as resilient only if they meet criteria in several realms at once (Cowen et al., 1990; Wyman et al., 1991); the resilient child is one who, despite deep adversity, "works well, loves well, and expects well." (Werner and Smith, 1982; quoted in Cowen et al., 1990, p. 193). There is a focus on adaptive function; most measurements are based on reports of children's behavior, but Cowen notes that resilience defined as "wellness" may include a wide range of physiological, behavioral, and psychological markers (Cowen, 1994).

In summary, these broad, environmental school-based studies have identified categories of family function, such as nurturance and support, organization and structure, and parental sense of efficacy as caregivers as major family correlates of child resilience. In view of their measurement largely through report of parents' perceptions, and interpreted through the lens of transactional theory, these as well as other family variables may be seen as aspects of the family's underlying sense that core aspects of family and social life important to the family are perceived as coherent, meaningful, and relatively manageable. The argument will be developed that this sense of coherence or motivational disposition affects family behavior in ways that promote child resilience.

For these researchers in community psychology, there is also agreement that child resilience, or the capacity to do

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well despite adversity, may be defined in terms of age and stage-specific competencies across physical, behavioral, social, and cognitive realms. Some studies measure one aspect of child function at a time, while others take a more stringent approach, requiring competency in, say, three out of four realms before a child is classified as resilient.

Resilience in Childhood Chronic Illness

Among children with chronic illness, researchers have considered both general and illness-related outcomes under the rubric of resilience, sometimes selecting for study those aspects most likely to be affected by the particular illness (Hauser, Vieyra, Jacobson, and Wertlieb, 1985). Thus, when children chronically ill with the same disease are measured on illness related variables, resilient as compared with non-resilient children might show better functional response to treatment, more adaptive treatment adherence behaviors, and better adjustment to illness. In terms of more general psychosocial function they might have fewer behavioral problems, better social competencies, higher self esteem, and better cognitive function and school performance. For example, authors of one set of longitudinal studies of preadolescent and adolescent children with diabetes, establishing a plan that has guided ten years of research, stated that "markers of outcome include physical measures such as metabolic control (hemoglobin Alc), medical providers' ratings of diabetic adjustment and compliance,

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and psychological measures such as the Diabetic Adjustment Scale (Sullivan, 1979), and behavioral symptoms (Achenbach and Edelbrock, 1979). Those with superior outcomes on these measures can be considered our resilient adolescents." (Hauser, Yiera, Jacobson, and Wertlieb, 1985). The broad areas usually explored are thus (1) treatment adherence and health outcome and (2) psychosocial function.

A General Model of Family Correlates of Resilience in Childhood Chronic Illness

A useful conceptual framework for organizing research findings on family correlates of resilience among chronically ill children is that adopted by Hauser and his colleagues at the Harvard Medical School's Joslin Clinic Health and Illness Project (Hauser et al., 1985). In a manner similar to that of Garmezy (1983), this group identifies a triad of factors affecting resilience: child personality, family, and social milieu. At the center is the family, exerting direct influence on, and being affected by, the child's emerging competencies and adjustment to illness, through parental attitudes and the overall family atmosphere of shared beliefs, behaviors, and values.

In addition to its direct influence, the family also indirectly affects child resilience through its response to and effect upon the child's personality and the social milieu, both of which in turn affect the child's adaptation. Both the direct and indirect family effects operate at what Hauser et al. call "macro" and "micro" levels. (Hauser et

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al., 1985). The group defines "macro" as "all studies that examine the family as a global entity" including studies of "(1) influence of family structure and home environment [i.e. demographic influences, crowding etc.]; and ; (2) influences of parental attitude and support." The "micro" level refers to "looking in detail at how aspects of family communication and interaction may be linked with child or adolescent vulnerability and resilience. The unit of analysis here is the individual family member, in relation to, and in interaction with, other members of the family." (Hauser et al., 1985 p. 86, emphasis added.) This concept of levels of family function is consistent with and lends support to a major argument of the present study, which holds that the family's global dispositional orientation to the familial and social world is mirrored and enacted in the family's time-bounded interactive everyday routines.

Studies of Families of Children with Diabetes

Research on family functioning and juvenile diabetes is relevant to a study of children with phenylketonuria (PKU), because the behavioral aspects of treatment are similar for both conditions, and because there are relatively few studies on the family and PKU. With both PKU and juvenile diabetes, the family is deeply involved in the management of treatment, and the regimen involves the child's dietary needs and food choices, affecting many family activities and routines. There are important differences, however. For the diabetic child, dietary and medication lapses may result in

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serious, even life threatening consequences and hospitalization. For the child with PKU, occasional lapses often have no immediately discernible effect; deterioration is gradual. PKU as a condition is "invisible" but the treatment regimen is ever present in family life; this combination may be baffling to parents, children, and those outside the family circle. A second important difference is age of onset. Children with PKU are diagnosed in the first weeks of life, and supervised treatment is most intensive through infancy and childhood; the onset of juvenile diabetes occurs at varying ages, with most research centered on the largest age group, older school age children and adolescents.

Focus on the families of children with diabetes began with Minuchin's work with psychosomatic families of crisis-prone children whose metabolic control was superlabile. His group identified poor outcomes with reciprocal family interaction patterns of enmeshment and rigidity (Minuchin, Rosman, and Baker, 1978). These patterns are frequently seen in families referred for psychological counseling; however, in studies of the range of child outcomes among families of more normal clinic populations, such extreme family enmeshment and rigidity are rare. Among normal populations, healthy child outcomes are usually positively associated with family cohesion and organization.

There is now considerable consensus, based on convergent research at the "macro" or global level, as to

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the family systems characteristics conducive to good outcomes among children with diabetes on dimensions of treatment outcome, adherence, and psychosocial function. Early studies focused on mothers' attitudes toward the treatment, rather than on mother's perceptions of family strengths or on family factors. Mothers' attitudes reflecting warmth, support, consistency, flexibility, and encouragement of adolescent autonomy in these early studies were found to be associated with good psychosocial adjustment (e.g. Crain, Sussman, and Weil, 1965) and with good metabolic control (e.g. Anderson, Miller, Auslander, and Santiago, 1981).

More recent studies of families have found both illness-related family support and general family adaptability to be associated with dietary adherence and with psychosocial adaptation (Hanson et al., 1992). Hauser and colleagues (1990) found that clear family organizational rules and boundaries, family cohesion, expressiveness, and low family conflict fostered adherence and good treatment outcome. These authors also found that when high cohesion (a measure of support, warmth, and commitment among family members) was present at the early stages of illness, the likelihood of long-term treatment adherence increased. Child and adolescent psychosocial adjustment is fostered by similar family patterns, with some evidence that among children with diabetes, as distinct from comparison families, the effects of family organization and conflict

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are stronger (Wertlieb, Hauser, and Jacobson, 1986). With regard to family coping, better child outcomes are associated with active family problem solving, and the family's capacity to perceive the environment as potentially supportive (Hauser, Powers, and Jacobson, 1988).

Except for work done with clinically referred families, such as that by Minuchin, there has been relatively little research at the "micro" level of interaction among families of chronically ill children. An exception is the recent work by the Hauser group. Hauser and colleagues (1986) have investigated such transactional effects using a "micro" methodology, a linguistic analysis of family conversational exchanges during a family interview following a stress event, the diagnosis of the child's diabetes. In this cross sectional study comparing families of children with diabetes with families of children with a recently diagnosed acute illness, results showed that the diabetic children's families, and especially their mothers, engaged in more "enabling" conversational interactions such as focusing, problem solving, and active understanding than they did in "constraining" conversational patterns such as distracting, excessive gratifying, and devaluing. This in turn encouraged the children's increased use of similar enabling patterns, especially focusing and problem solving.

Commenting on this study in their explication of a transactional family process theory, Fiese and Sameroff (1992) place the Hauser group's (1986) study in the context

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of a social regulatory model, emphasizing that the parents' underlying perception of their children as vulnerable guided these reinforcing transactions. Their point is that the effect of child behavior on parents' responses is filtered through the interpretive light of the parents' internalized representational structures. This affects the parents' responses, which in turn affect the child's self-representation and subsequent child behavior (Fiese and Sameroff, 1992). In a later "micro" study, this time an analysis of the family coping strategies of appraisal, problem solving and emotion management as revealed by conversation content and patterns, the Hauser group did find that long-term stable underlying constructs, such as parental ego development, correlated positively with specific effective coping behaviors (Hauser et al., 1993).

In summary, the research on family correlates of child resilience among families of children with diabetes indicates that the earlier findings of the community and epidemiological psychologists also apply to this population. The same triad of social environmental, family, and child characteristics are found to contribute to child resilience, with the family milieu seen as a central force. As in the community studies, family warmth and cohesion, as well as organization and structure, are identified as major factors in all aspects of good physical, behavioral, and psychosocial outcomes for these children with diabetes. Although these studies of families with chronically ill

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children do not explicitly address the role of parents' perceptions of the family's effectance and place within the social world, studies by Hauser's group (Hauser et al., 1986; Hauser et al., 1993) and theoretical work by Fiese and Sameroff (1992) have shown that global or underlying family attitudes and parental psychological structures relate to specific behavioral enactments in the context of the children's treatment. Fiese and Sameroff argue further that such enactments form a matrix for the transmission of parental attitudes and representations to the children.

Studies of Families of Children with PKU

The few existing studies of the influence of family functioning on child outcomes in PKU, like those on diabetes, have focused on (1) treatment adherence and metabolic control, which are more closely causally related for PKU than for juvenile diabetes and (2) psychosocial function. Two early studies, both dating from 1967 and each including 22 children, looked at parent-child interaction in play situations (Keleske, Solomons, and Opitz, 1967; Wood, Friedman, and Steisel, 1967). The researchers found that parental worry and restrictions inhibited the children's play; however, many of the children in these samples were retarded because of late-diagnosed PKU. Thus the retardation itself rather than the children's PKU and associated aspects of its treatment probably affected the parent-child interaction.

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A review of the literature found no further studies of PKU and family functioning until five appeared in the past decade, three based upon the same subject pool. Each of these studies reflects the current interest in resilience and range of function, and they are also representative of the three main models (social support, stress and coping, and family systems) that have been most frequently applied to family issues in pediatric psychology.

Kazak, Reber, and Carter (1988), applying classic social network theory, found that PKU families (N=45) did not differ from comparison families (N=49) in terms of network size and density, but that PKU mothers were significantly less likely to name their spouses, parents, and extended family members as part of their network than were comparison mothers. PKU mothers were more likely to name their children. Both PKU mothers and fathers reported receiving less emotional help from family members than did comparison mothers and fathers.

A second descriptive comparison study by the Kazak group, using the same samples, found "lower levels of adaptability and cohesion for the families with a child with phenylketonuria and evidence of lower levels of social competence in the children with phenylketonuria than in the comparison group." (Kazak, Reber, and Snitzer, 1988, p. 224).

This finding did not, however, imply a significant correlation between family function and child social

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competence, one of the several relationships investigated in a related study with the same sample of PKU families (Reber, Kazak, and Himmelberg, 1987). Indeed, in the 1987 study, few relationships of any kind were found between family function and child resilience. The model that was applied, using standardized self-report measures, was loosely based on premises of structural family systems theory. Areas of family adjustment assessed were parents' individual psychological functioning (Langer Symptom Checklist), marital satisfaction (Dyadic Adjustment Scale), parent-child functioning (Parenting Stress Index), and family system functioning as whole (Family Adaptability and Cohesion Evaluation Scale-Version 2). Child outcome measures included level of metabolic control and cognitive function for all 41 child subjects, and measures of social competence and behavioral adjustment for 14 child subjects based on parent report on the Child Behavior Checklist. Neither metabolic control nor child social competence correlated significantly with any of the family psychosocial measures. Cohesion as perceived by mothers and adaptability as perceived by fathers did correlate as expected with the children's cognitive functioning, but accounted for only a small part of the variance (4%). Children's behavior problems as measured by the CBCL also correlated negatively with mothers' perceived family cohesion; but the variability of the behavior problems scores was mostly in the normal range. Accounting for the relatively slim findings, the authors

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conclude that the measures used may have been too general to capture the differences among families.

Fehrenbach and Peterson (1989) designed a procedure to measure parental problem solving skills specific to PKU dietary compliance. The authors used a stress and coping model based on the well known work by Folkman and Lazarus on cognitive appraisal, coping, and outcomes (Folkman, Lazarus, Dunkell-Schetter, DeLongis and Gruen, 1986). Using an experimental procedure involving simulated everyday parent-child problem situations conducive to noncompliance, they found that compliant as compared to noncompliant parents gave higher quality verbal and written problem solving solutions; their responses were non-punitive, effective, and integrated several behaviors into a single solution. These parents also experienced less subjective stress than noncompliant parents in the experimental situation. The authors note that the findings provide support for the Folkman model, which posits problem solving as an effective mediator of stress in chronic illness. On a measure of general family function included in the study, the Family Environment Scale (Moos, 1974) higher scores on the Control scale, a measure of family adherence to fixed rules connoting relative rigidity, also correlated positively with compliance.

A similar moderate inflexibility was found in a study of families of children with PKU by Shulman, Fisch, Zempel, Gadish, and Chang (1991). The functioning and coping of 43

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families with children with PKU was investigated. A primary measure of family functioning was the Reiss' Card Sort Procedure, an experimental family problem solving task yielding a family typology comprising three main types of family interaction, each reflecting the family's conception of their social reality (Reiss, 1981). Unfortunately, an insufficient number of mother-father dyads participated for the results of the Card Sort Procedure to be included in the statistical analysis relating this aspect of family function to treatment adherence, child psychosocial function and metabolic control. Qualitative analysis suggested, however, that nearly all the PKU families reached a quick consensus, opting for lower quality solutions without exploring options. The authors interpret this as functional cooperation that may have some price. Parent self-reports of general coping styles on a general measure, Family Crisis Oriented Personal Evaluation Scales (McCubbin, Larson, and Olson, 1981), did not correlate with dietary adherence or metabolic control. Family cohesion, however, measured on FACES-III (Olson, Portner and Lavee, 1985), did correlate with child IQ, dietary adherence, metabolic control and some aspects of child behavior.

What conclusions may be drawn from these disparate findings? First, simply, that the research on this particular population is in the early stages; studies are sparse, so the full patterning of family effects upon child resilience is not yet clear; also, there is little work

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done at the "micro" level. Second, the relative inflexibility suggested by the high control and low adaptability noted in separate studies may be adaptive with regard to dietary control, but results suggest it takes a toll on the children's cognitive development. Third, there is some evidence that family cohesion benefits children with PKU as it does children with diabetes, though the results across the PKU studies are inconsistent. For the children with PKU, family cohesion was associated with good outcomes, and was the only family variable to correlate positively with all three measured outcome realms, physical, cognitive, and psychosocial, albeit in separate studies, with one study finding a correlation where the other did not.

Comparison across studies also suggests that this potentially beneficial pattern of cohesive interaction enjoyed by some families with a child with PKU may be attenuated in many other PKU families: the Kazak descriptive studies discussed indicate that, compared with others, many of these families present as less directly emotionally supportive of one another within the nuclear and extended family contexts, and fathers, especially, may be distanced from both family and friends. Considered together, the Kazak (1988) finding on lower family adaptability, the Fehrenbach (1989) finding on higher control, and the Shulman (1991) finding on constriction in problem solving all suggest that, compared with other families, a greater proportion of families of children with PKU may be less

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expressive, less creative and flexible in meeting environmental demands, with a more rigid organizational structure in which consensus is not negotiated, but is achieved through ready-made rules and hierarchical controls. (Shulman et al., 1991).

Shulman comments that the need for clear procedural rules in the everyday decisions about routines involving the diet in daily life has probably influenced the parental coping style, speculating that it may be different, but adaptive. Recall, however, that adaptability among families of diabetic children was associated with concurrent adjustment, and cohesion among families of young diabetic children predicted to adolescent adjustment and adherence. The relative inflexibility of families of children with PKU, especially in conjunction with the lower support and cohesion that may characterize some of these families, could be problematic for the children's long term adjustment and adherence (Hauser et al., 1990). In seeking antecedents of resilience it may, therefore, be especially important to identify those families that can imbue both their larger overall environment and their everyday routines with meaning as well as order.

Family Sense of Coherence and Family Routines: A Transactional Ecological Perspective

Family Sense of Coherence

The construct of family sense of coherence as adapted for the present study is based on and combines the theories

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of two sociologists, Aaron Antonovsky (1979, 1987) and David Reiss (1981). It refers to the extent to which family members, individually and in agreement, have a "pervasive, dynamic, and enduring" belief that, for the family, the social world is comprehensible, manageable, and meaningful (Antonovsky, 1987).

Antonovsky, a medical sociologist, like Rutter sought an explanation of "what goes right," developing what he called the salutogenic hypothesis. This hypothesis states that, under the right conditions, the multiple stressors in the world cause not stress, but successful management of tension. Antonovsky thought that such successful management of tension could be salutogenic, preventing breakdown on the continuum of "health ease/dis-ease," and promoting health. (Antonovsky, 1979).

The general salutogenesis model proposed by Antonovsky (1979, 1987) describes sense of coherence as an enduring feeling of confidence that, for the individual or group, the inner and outer world "makes sense," that life's challenges are worthy of engagement, and that resources are available to meet them. This dynamic orientation is founded upon long term integration of life experiences characterized by consistency, a reasonable balance between demands and resources, and a sense of participatory agency in the shaping of outcomes. Sets of coherent, meaningful life experiences are regularly provided by "generalized resistance resources," those characteristics of individuals

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or groups that facilitate avoiding or combating a broad range of stressors. Generalized resistance resources are of many types--material, cognitive, psychological, spiritual, attitudinal, interpersonal, sociocultural--including assets such as wealth, intelligence, ego strengths, flexible coping strategies, family cohesiveness, social supports, religious beliefs, and the like. The sense of coherence has many sources, and individuals and groups differ as to which are most necessary in its formation, but, as Antonovsky notes, in all cultures wealth is an especially potent means of avoiding and combating stress.

Set against these resistance resources are stressors of many kinds including life events, effects of illness, intrapsychic or interpersonal or social conflicts, horrors of history, culture shock associated with immigration, and so forth, as well as deficits in the major resistance resources. Antonovsky defines a stressor as any event or condition that seriously disrupts homeostasis because a routine response would be unavailable or would be inadequate to its restoration (1979). He makes a crucial distinction between tension and stress. Stressors cause tension, a state of felt need for the resolution of the precipitating situation. If a stressor is overcome, tension resolves; if not, stress results, with accompanying biological, psychological, and behavioral responses that can maladaptively interact with pathogens and "weak links" to precipitate "breakdown" or movement toward the "dis-ease"

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end of the health-ease/dis-ease continuum. This continuum is a model of health that takes into account the individual's subjective experience of pain and functional limitations of an illness, together with prognosis and need for intervention as defined by health professionals (Antonovsky, 1979).

Stressors that may potentially cause breakdown are, Antonovsky argues, ubiquitous in life. In a striking image, he compares life to a rushing river in which all are ultimately swept out to sea, but in which some are drowning and others are swimming with relative ease. How well one swims, he argues, is largely determined by one's sense of coherence. Those with a strong sense of coherence expect that order and meaning can be brought into confusing or trying situations. Motivated by reasonable hope founded upon experience, they selectively seek resources that will achieve this end, thereby resolving tension and promoting health (Antonovsky, 1979).

It is the sense of coherence, or the internalized belief in the reasonable predictability and consistency in the world, and in its general meaningfulness, that enables an individual to mobilize the resources to cope with a given adversity. Antonovsky felt that an individual's sense of coherence forms over a lifetime, founded upon experiences that promote a sense of participatory agency in a social world that presents as relatively consistent, manageable, and rich in meaning. As an enduring sense of confidence, the

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sense of coherence helps one to persevere and to overcome adversity.

Acknowledging the importance of the stress and coping and social support research, Antonovsky thought these models were inadequate without such a strong, person-based mediating principle such as the sense of coherence. For example, Antonovsky argued that cognitive appraisal and coping style, both primary predictors of stress management acccording to Folkman, Lazarus and colleagues (1986), are actually secondary to an underlying sense of coherence. That is, it is the individual's sense of coherence that permits the informed flexibility of such appraisal and coping styles. The sense of coherence is related to more recently developed person-based models that explain successful coping, such as Scheier and Carver's (1985) and Seligman's (1991) construct of optimism, Bandura's (1989) of self-efficacy, and Snyder's (1989) of hope; but it differs from them in its emphasis on the self as deeply social in nature, and in its concern with valuation, commitment, and meaning in life.

As developed by Antonovsky, the concept of sense of coherence refers to individuals, but he felt it could apply to groups and to families as well, and he acknowledged the similarity of his theory, independently developed, with that of the sociologist David Reiss (Antonovsky, 1987; Reiss, 1981). Reiss's model is constructivist; he presents a typology of family problem solving styles that reflect three

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differing family views of the social world and the family's place within it (Reiss, 1981).

According to Reiss, families differ in their conception of the social world and their customary way of responding to it along dimensions of configuration or the capacity to grasp subtle and complex patterns, coordination or the ability to form shared solutions, and closure or the ability to defer quick solutions while drawing from the richness of available information. Family function and the family's construction of the social world may be described in terms of their being high or low on these three dimensions. For example, "consensus-sensitive" families tend to close ranks against the social environment, placing a high value on cohesion to the exclusion of examination of the richness of information; "distance-sensitive" families value personal independence and self control, rarely reaching a coordinated solution; and "environment-sensitive" families remain open to the information from the environment while sharing information with one another in order to reach an optimal solution (Reiss, 1981; Reiss and Oliveri, 1980). When the concept of individual sense of coherence is recast in family terms, it resembles Reiss's notion of the environment-sensitive family; though, again, the sense of deep valuation that Antonovsky emphasizes is less prominent in Reiss's work.

The present study combines the ideas of Reiss and Antonovsky in the construct family sense of coherence. An

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important theoretical assumption in this construct is its contextual nature: families are part of, interact with, influence and are influenced by, the surrounding community and culture. Family sense of coherence is an ecologically based dispositional orientation formed over time through the mutually influencing transactions within the family and between the family and the larger environment. Once formed, this orientation or family paradigm is slow to change, forming a core set of assumptions that further construct the family's social reality, influencing how the family perceives "the relative safety and stability of the social world, the degree to which it is experienced as novel or precedented [sic] by past experiences, and the conviction that it treats the family as an integrated group or as a group of unrelated individuals." (Reiss, 1989)

Because both the general and illness-related care of children is embedded within the family, members' sense of the strength of family coherence may influence many aspects of treatment of chronically ill children, including, for example, how the family reacts to health professionals (Fiese and Sameroff, 1992). Reiss (1981) has linked the family typologies derived from his model to outcome for schizophrenic adolescents and Hauser and his colleagues have found that coping strategies consistent with these typologies reflect higher levels of parental ego function and better outcome for adolescent diabetics (Hauser, DiPlacido, Jacobson, Willett, and Cole, 1993). In studies

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based on Antonovsky's work, higher levels of individual sense of coherence have correlated with better health, general sense of well being, and adjustment to illness (Antonovsky, 1993); and family sense of coherence has correlated positively with adjustment in disabled men (Antonovsky and Sourani, 1988; Sourani, 1983). High sense of coherence has thus been empirically linked to resilience.

Reconceived for the present study in family terms that include Reiss's view of the family's construction of social reality, the term family sense of coherence refers to the extent to which members believe that, for the family, the world makes logical, social and emotional sense. The events and demands of family and social life have a relatively predictable structure, resources are available to meet these demands, and the demands are challenges that are worthy of family emotional engagement and commitment. (Antonovsky, 1987)

In short, family members, perceiving their family to have a strong sense of coherence, have faith in their family as a system that can reliably access support as needed from the individual, familial, and societal realms. Within such a family, members experience their personal internal and external world, their interpersonal relations with family members, and the family's relations with the larger social surround as relatively comprehensible, manageable, and meaningful.

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The three components of family sense of coherence are comprehensibility, manageability, and meaningfulness. Each component applies to members' perceptions about relations within the family and to those about relations between the family and the larger social world, including members' perceptions about how that world views the family. Family sense of coherence is thought to form a foundation for hope and optimism, and to precede and structure how the family perceives and reacts to stressors, mobilizes internal and external resources, chooses strategies, and applies solutions. Formed through integration of long-term group experience, it underlies these processes as a global, dispositional orientation.

Family sense of coherence is conceived as a continuum from weak to strong. Theoretically, the three components of comprehensibility, manageability and meaningfulness are separable. Each may range from weak to strong, but in life the components are usually intertwined, clustered at a similar level on the general sense of coherence continuum (Antonovsky, 1987).

Comprehensibility refers to the "understandability" of one's internal and external world, the belief that the world has a lawfulness and structure that makes sense--and that the individual and the family "makes sense" to others; the family is definable and its relationship to the larger society is understandable to others. For the family, the predictability of the world can include lawful change; the

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order of the world may include sad and traumatic events, but families can make sense of these events, discern their structure, and place them in perspective.

Manageability refers to family members' belief that resources are available to the family to meet the challenges of life. Such resources may be of many types--money, flexible coping strategies, family cohesiveness, individual member's ego strength, social support, religious beliefs, trustworthy professionals, to name a few. This component differs from the concept of internal locus of control. Family sense of coherence may be strong when resources and outcomes are within the control of the family, but also when they are within control of others whom the family actively acknowledges as legitimate. In both cases members experience a sense of agency and effectancy through the family's sense of mutual participation in outcomes. Members are confident that familial and societal resources can be accessed, and that, within reason, their use will be effective. Conversely, members respect the actions and expectations of others outside the family whom they have deemed legitimate. Antonovsky notes that "to the extent that one has a high sense of manageability, one will not feel victimized by events or feel that life treats one unfairly. Untoward things do happen in life, but when they do occur, one will be able to cope and not grieve endlessly." (Antonovsky, 1987, p. 18.)

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Meaningfulness refers to the deep emotional investment, engagement, and commitment--the valuing--that family members and the family as a whole bring to life and its challenges. As the motivational element it is a central aspect of the sense of coherence, and has a strongly Existentialist quality, in that the meaning of an event is in part "made" by the family's active engagement. Life's problems are seen as worthy of the investment of emotional energy. Conversely, the family feels that the investment it makes is valued by the groups in which it participates. Family members with a strong family sense of meaningfulness will have faith in the family's ability to rise to a challenge, to actively seek and find shared meaning even in times of trouble. They will trust that the family can make emotional sense of the world.

For younger children as well as for adolescents and adults, family sense of coherence, translated into daily behavior, provides direct experience of the extent to which the world is comprehensible, manageable, and meaningful across a broad range of situations. It is also the lens through which the child's illness, within the family and in the family's relation to the larger social world, is interpreted to the child.

For children of all ages, but especially for preschool and elementary school age children, a productive way to link family sense of coherence to resilience is through family routines, both because such routines increase in frequency

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during this family phase (Fiese, Hooker, Kotary, and Schwagler, 1993), and because family routines provide a developmentally appropriate channel through which family sense of coherence may be internalized. Both Antonovsky and Reiss recognized the importance of family routines as related to their models. Antonovsky felt that the continuity and permanence symbolized by such routines was related to the comprehensibility component of sense of coherence (Antonovsky, 1987), and Reiss notes that family routines are behavioral, capsule versions of the family's abiding view of the world (Reiss, 1981).

Family Routines

The protective effect of family routines and rituals on family members' psychosocial well being and physical health has received increasing attention in recent years. In their typology of family rituals, Wolin and Bennett (1984) describe how, through their power to transform, communicate, and stabilize family life, family routines and rituals come to establish and enact the family identity, or the family's collective sense of self. They describe three types of rituals: family celebrations, in which cultural holidays such as Thanksgiving, and rites of passage such as baptisms, weddings and funerals affirm membership in the larger culture and signify the family's developmental phase; family traditions, shaped by the culture, but given particularity by the family, such as celebration of birthdays, the regular family vacation, or "Sunday Dinner;" and patterned family

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interactions such as bed time routines, customary ways of marking departures and arrivals of family members, weekend activities, organization of chores. Boyce and Jensen have called such patterned interactions family routines, describing them as the observable, recurring interactions of daily family life that provide sense of order, stability, and predictability that fosters a "sense of permanence" and belonging (Boyce, Jensen, James, and Peacock, 1983; Jensen, James, Boyce, and Hartnett, 1983).

Such routines, even when they are simply instrumental, intended to get the day's work done or manage transitions smoothly, often take on symbolic significance through their regular recurrence, prescription of roles, and ascription of meaning, thereby helping to organize and define the family's identity and the place of members within that identity (Boyce et al., 1983). Defining routines from an ethnographic perspective, Boyce and Jensen suggest ways their practice may foster family well-being and the management of stress. Because they recur regularly, family routines provide a structured continuity to daily interactive life. As repeatedly enacted symbols of family relatedness, such routines promote stability during transitional periods, fostering the strengths needed to cope with stress, and thereby protecting family morale and health (Boyce et al., 1983; Jensen et al., 1983).

Endorsement of and adherence to valued family routines and rituals has been associated with a wide range of health

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benefits, including fewer respiratory illnesses (Boyce et al., 1977) and better general health among children (Keltner, 1992), and lower rates of alcoholism among adult children of alcoholics (Wolin, Bennett, and Jacobs, 1988). Among the documented psychosocial benefits that have been found are better social competence among preschool children (Keltner, 1990), and, especially when ascription of meaning is paramount, higher self-esteem and lower anxiety among adolescents (Fiese and Kline, 1993; Fiese, 1992), and greater marital satisfaction among parents of young children (Fiese, Hooker, Kotary, and Schwagler, 1993).

Antonovsky theorized that family practice of daily routines and the sense of coherence are interrelated, in that both create a sense of predictable continuity in life (Antonovsky, 1987). Similarly, Reiss has theorized that the content, quality, and process of interactive family routines are guided and informed by the family paradigm, a construct similar to the sense of coherence. Reiss saw these routines as one behavioral component of the family paradigm. (Reiss, 1981). Family routines and rituals may be seen as an enactment, writ small, of the family sense of coherence. As structured, enacted, predictable events, they organize time, memory, and anticipation, placing the participants within a larger structure; they render experience <u>comprehensible</u>. As the behavioral units of family interaction, often ways of getting things done or managing the stress of transitions smoothly, they make experience <u>manageable</u>. As symbolic

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expression of family identity, often including heightened feelings of interpersonal relatedness, they render experience meaningful. Family routines and rituals thus bear an analogous relation to family paradigm or sense of coherence, in that they reflect and embody the family's felt vision of itself and its relation to the larger world (Antonovsky, 1987; Reiss, 1981; Fiese and Sameroff, 1992).

While family sense of coherence is a global dispositional orientation that rests ultimately on individual members' perceptions of family strength, family routines are specific, temporally bounded behavioral enactments that are inherently shared, participatory events. As such, they are a potentially important means for the child's internalization of family attitudes and values. Such routines are the "organizational units of ordinary life in families."(Boyce et al., p. 194.) They are inherently regulatory and transactional. As homely, affectively patterned interactions, they are guided by aspects of the family's overall sense of coherence. As rituals, family routines may represent the family paradigm in symbolic form with heightened clarity. When the child shares in the actions of routines, participatory learning binds cognitive, affective, and behavioral elements together in a way that renders the order and meaning implicit in the family milieu more accessible to the child. Increased apprehension of the larger meaning may then re-inform and enrich engagement in the routine, beginning a new cycle of interpretation. In

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this way, children come to share in the overall family sense of coherence in its relative strength or weakness, and thereby to develop more, or less, resilient ways of managing tension and meeting the everyday challenges of life.

Within a transactional framework, the family's practice of meaningful, supportive family routines may be seen as promoting positive developmental experience through the feedback that occurs through their repeated enactment. Sprunger, Boyce, and Gaines (1985), using the Family Routines Inventory (Boyce et al., 1983; Jensen et al., 1983) found that family adherence to valued routines in homes with infants was associated with overall family life satisfaction, and mothers' sense of competency as a parent. In a transactional paradigm, a mother's sense of efficacy could further enhance pleasurable, synchronous exchanges with her child, fostering security. According to attachment theory research, the synchronous exchanges typical of securely attached children enable such children to respond more adaptively in situations calling for compliance (Rocissano, Slade and Lynch, 1987). Thus we see two connections: a family routine reflects and affirms the larger family milieu, and a family routine may serve as a "holding environment" for development of specific role behaviors, including compliance.

For parent and child, there are many ways that a rich repertoire of valued family routines might enhance probability of treatment compliance and adherence. Ideally,

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there should be a good balance in the integration of treatment with routines; treatment should neither overwhelm nor be entirely excluded from the dailiness of life, but should be flexibly integrated in those routines in which such integration seems desirable. Thus, many routines and rituals might function as those pleasurable times when life is not affected by illness and treatment. Others may provide an ongoing script for incorporation of treatment routines, or they may serve as cues, temporal prompts, and models for new routines.

A clinical example may also illustrate this point. Children with PKU cannot eat bread made with regular flour, and some parents bake bread made with flour that has been chemically altered to remove phenylalanine. As part of a routine language assessment, a therapist in session with a three year old boy whose mother was present attempted to elicit a sequential narrative of daily events as the child busily pushed a tiny shopping cart with a toy dinosaur. No "shopping script" was forthcoming, and the mother said, "Tell her about Donut Day!"

Child: (smiling, to therapist, emphatically) Daddy buys the donuts!

Mother: And Mommy...?

Child: And Mommy makes the donuts.

Mother: (quickly, to therapist) PKU donuts.

Child: And Tommy (his fraternal twin brother)...We ice the donuts.

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Mother: And...

Child: And we <u>eat</u> them! (smiling hugely and spreading fingers)

Mother: (laughs) Your fingers get sticky, don't they?

The mother then explained that Donut Day is a Sunday morning ritual from before the twins were born, when her husband would always bring home donuts from the local bakery. He still does, but in the morning she makes special ones for the child with PKU, and her husband always gets some plain donuts so Tommy has a bakery one to ice (frost), too. The ritual reflects family tradition, continuity, flexible change, warmth, and comfortable acknowledgment of the children's shared and differing needs; a morning activity that might have excluded the PKU child has been modified and expanded to include him, to everyone's evident pleasure. The diet is followed, roles are clarified and enacted, participation of each member is high, and Donut Day is observed.

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Definitions

Family Sense of Coherence

Family sense of coherence is a global dispositional orientation that expresses the degree to which family members have internalized a "dynamic feeling of confidence" (Antonovsky, 1979, p. 10) that, for the family, the events arising in the course of family life and in its reciprocal relation to the social world are (1) comprehensible: these events are explicable and predictable; they can be interpreted in a context in which they make sense; (2) manageable: resources are available to meet the challenges of life; the family can cope or gain needed support; and (3) meaningful: in a context of mutual, reciprocal commitment, these demands are challenges, worthy of shared emotional investment and engagement (Antonovsky, 1979, 1987; Antonovsky and Sourani, 1988; Reiss, 1981); and that "there is a high probability that things will work out as well as can reasonably be expected."(Antonovsky, 1979, p. 10) Formed over time through the integration of experience, family sense of coherence is an emotionally funded, dynamic and enduring representation of the social world and the family's place within it, structuring family perception and response to stress, and providing a foundation for hope.

In this study, family sense of coherence as perceived by mothers was measured by maternal report on the Family Life Questionnaire, a scale adapted by the writer, based on the work of Antonovsky (1979, 1987), Antonovsky and Sourani,

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(1988), Sourani (1983) and Reiss (1981). This scale measures general family sense of coherence; it relates to family life in general and includes no questions related to any specific illness (See Appendix). It was completed by all the mothers in the study.

Factors associated with mothers' perceptions of family sense of coherence in its relation to the PKU diet were further explored through clinical evaluation of mothers' responses to a semi-structured interview, the PKU Parent Interview, written for this study (See Appendix). A subset of mothers in the study agreed to be interviewed.

Family Routines

Family routines are structured, repeated behavioral events involving participation of two or more family members, that occur with predictable regularity, giving order and continuity to daily family life. (Boyce et al., 1983; Jensen et al., 1983). They are the observable, common events that may express affect, effect goals, and mark transitions during the days and weeks, consisting of sequenced acts and roles that form "what usually happens" in such time-bounded, scripted family events as parents' return from work, a trip to the store, performance of chores, family meal time, children's bed time, and the like. Family routines are repeated but evolving behavioral units that facilitate procedures and shape family experience, and that may promote family sense of stability and continuity,

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providing a locus for the ascription of shared meaning. These functions of family routines may foster the family strengths conducive to successful management of tension in ways that promote health and well-being.

Family practice of regular routines was measured by maternal report of the frequency of such family interactions, based on the frequency score of the Family Routines Inventory (Jensen et al., 1983). This measure concerns the kinds of conventional, normative routines outlined above, and does not include any routines related to illness or diet. All mothers in the study completed the frequency scale of this measure.

Family routines specifically related to the diet, and the balance between diet-related routines and other family interactions, were topics that explored through clinical evaluation of mothers' responses to the PKU Parent Interview, developed for the study.

Child Resilience

The resilient child is one who "works well, loves well, expects well," despite life's adversities. (Werner and Smith, 1982) Compared with other children who develop problems of coping and adjustment when exposed to similar life stressors, resilient children cope and adapt well. They exhibit age and stage specific competencies across physical, behavioral, social, and cognitive realms. Among children with chronic illness, resilient children are those

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who show better function in areas likely to be affected by their illness. For example, resilient children with PKU could be expected to exhibit better social skills, better behavior adjustment, better school performance, and more adaptive treatment behavior such as adherence to prescribed regimens.

Three dimensions of general psychosocial resilience were considered in the study: social skills, behavioral adjustment, and school competence. One measure of adaptive treatment behavior, dietary adherence, was considered in the study.

Social skills refer to children's prosocial daily interactions with others. Such skills include cooperating with others in work, play and daily family life; being appropriately assertive in social situations; acting responsibly in the performance of social tasks; showing empathy for others; and demonstrating appropriate self-control in social situations. Social skills were measured by maternal report on the parent form of the Social Skills Scale of the Social Skills Rating System (SSRS) by Gresham and Elliot (1990).

Behavioral adjustment refers to degree of freedom from externalizing problems such as behaviors involving poor control of temper, and verbal or physical aggression toward others; internalizing problems such as behaviors indicating anxiety, sadness, loneliness, and poor self esteem; and (for elementary school age children), hyperactivity behaviors

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involving excessive movement and impulsive reactions. Behavioral adjustment was measured by maternal report on the Behavior Problems Scale of the Social Skills Rating System (SSRS) by Gresham and Elliott (1990).

School competence refers to degree of successful performance at school as indicated by a history of yearly promotions into the next grade; lack of need for special educational services; and average or better academic achievement. School competence was measured by maternal report on the School Competence sub-scale of the Competence Scales of the Child Behavior Checklist (CBCL/ 4-18) by Achenbach (1991).

Dietary treatment adherence refers to the degree to which the family and child follow the correctly prescribed, individualized treatment diet. Adherence to the diet involves a set of interrelated behaviors. It includes food intake behavior, such as child's drinking the formula and eating permitted foods in prescribed amounts while refraining from drinking and eating non-permitted foods. It also includes those family and child behaviors effectively supporting correct food intake.

In this study an indirect measure, or index, of dietary adherence was used: the child's long-term mean phenylalanine level, based on medical record of blood tests results for each child during the full year immediately preceding the study. Physicians and clinicians consider phenylalanine control over time a "best available" index of patient

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dietary adherence because phenylalanine intake by patients with PKU is very closely related to blood phenylalanine control. Researchers have also taken individual patient mean phenylalanine levels over time as a measure of dietary adherence (Fehrenbach and Peterson, 1989).

Formulation of Research Question

The central argument of the study is that the family's sense of coherence, together with and as enacted through family routines and rituals, facilitates the effective family coping that fosters child resilience under a condition of stress. "Effective family coping" comprises a broad set of intermediary variables, such as the use of generalized and specific resistance resources, inferred through correlations between the family predictor and child criterion variables. While information on selected intervening variables was sought, the focus of the empirical study is on the relations among family sense of coherence, family routines, and child resilience. The association between family sense of coherence and family routines, proposed by Antonovsky (1987) and Reiss (1989), is tested as a means exploring the construct validity of family sense of coherence. Insofar as each of these variables represents the two poles of the family paradigm (Reiss, 1989), the representational and the behavioral realms of experience, they are separate but related, and should show a moderate

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correlation. Because the two family variables each access differing realms of experience, family sense of coherence and family practice of regular routines are each hypothesized to contribute separately to child resilience; but if they are mutually reinforcing, their greatest effect may occur when both are strong. It is further expected that mothers' descriptions of living with PKU will reflect the two family strengths in terms related to the diet, and that such "diet related" sense of coherence and "diet related" family routines will be associated with treatment adherence.

The working formulation of the research question is:

> Among the population of families with children who have the chronic metabolic disorder phenylketonuria, how strongly do the variables of family sense of coherence and family practice of regular routines correlate with one another; and, how strongly do the variables family sense of coherence and family practice of regular routines each correlate with child psychosocial resilience and dietary adherence?

Hypotheses

The following three hypotheses will be tested:

Hypothesis #1:

> Family sense of coherence will correlate positively with family practice of regular routines.

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Hypothesis #2:

Family sense of coherence will correlate positively with dimensions of child resilience including (a) social skills, (b) behavioral adjustment, (c) school competence, and (d) dietary adherence.

Hypothesis #3:

Family practice of regular routines will correlate positively with dimensions of child resilience including (a) social skills, (b) behavioral adjustment, (c) school competence, and (d) dietary adherence.

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CHAPTER THREE

METHOD

Design

The study is correlational and retrospective in design. Within a sample of families of children who have the chronic metabolic condition phenylketonuria (PKU), the study first examined the association between two aspects of family functioning: family sense of coherence and family practice of regular routines, both as perceived by mothers. Each of these family variables was then separately correlated with measures of child outcomes including social skills, behavioral adjustment, school competence, and dietary adherence. The study drew upon information from pencil and paper questionnaires completed by mothers of children with PKU collected from each individual respondent on a single occasion, and from each child's medical records covering the continuous year preceding parental completion of the questionnaires. Qualitative aspects of families' experience coping with the PKU diet were then assessed through individual, semi-structured interviews with a subset of mothers.

Subjects

Subjects for the study were 34 families with children ranging from preschool age through grade six (mean age in months=90.03) on a low phenylalanine diet for treatment of phenylketonuria (PKU), a metabolic disorder requiring strict

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dietary control in order to prevent mental retardation. Families were recruited from three PKU treatment centers in metropolitan hospitals in two Northeastern states. Children were eligible for the study if they had begun the diet within three weeks of birth, remained on the diet continuously, and had been in the same primary caregiver's care for 11 of the 12 months during the year from which treatment adherence data were drawn. In the three families in which there were siblings with PKU, one child from each of these families was randomly selected to participate.

The three treatment centers were comparable in terms of kind of location, frequency of dietary counseling, policies regarding frequency of treatment outcome assessment, treatment goals, and method of measuring treatment effectiveness. Medical directors described each set of participating families as generally representative of their patient populations.

The subset of mothers participating in the qualitative follow-up interview (N=14) included mothers from each of the three centers.

Demographic information as reported by parents is given in Table 1 for the whole sample. Geographically, the families reside in rural, suburban, and urban areas scattered throughout the two states. The ethnic distribution reflects the incidence of this rare genetic disorder in the differing ethnic groups.

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Table 1

Sample Demographic Characteristics

Variable	N	%
Sex of child		
Male	14	41.2
Female	20	58.8
Ethnicity of child		
White	30	88.2
Black	2	5.9
Hispanic	1	2.9
Other	1	2.9
Social class*		
I	3	8.8
II	7	20.6
III	12	35.3
IV	12	35.3
Family structure		
Single parent family	4	11.8
Two parent family	30	88.2
Mother's education		
Graduate school	2	5.9
College graduate	9	26.5
Partial college	6	17.6
High school graduate	16	47.1
Partial high school	1	2.9

*Hollingshead, 1957 (Father's education included in formula)

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In the present study, no family reported information meriting a socioeconomic Class V status (Hollingshead, 1957; Miller, 1991); however, some families in Class IV suffered periods of unemployment or curtailment of telephone service and one family was living in a shelter for homeless families for several months. Some in class IV were also uninsured and either did not choose or did not qualify to obtain Medicaid.

At each center, all families with children meeting eligibility requirements were invited to participate. No incentives were offered for participation in the study. Number of participating families, number of eligible families, and response percentages for each center are given in Table 2. Center #1, with the highest response rate of participation, is the center sponsoring the research and with which the researcher is associated.

Table 2

Participation and Response Rate, by Center

Center	Participating	Eligible	% response
#1	25	28	89%
#2	5	13	38%
#3	4	9	44%
Total	34	50	68%

Children with this rare condition are treated only in PKU treatment centers, each covering a wide geographic area. For this study, approximately 68% of families of children

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with PKU who met age and eligibility requirements in an area comprising all of one state and half of another participated in the study.

Measures

Quantitative Family Measures

Family Sense of Coherence

Family sense of coherence, or the extent to which mothers perceive that, for the family, the social world is comprehensible, manageable, and meaningful was measured by the Family Life Questionnaire (FLQ). This 36 item Likert type scale was adapted for this study from a scale measuring individual sense of coherence by Antonovsky (1987)and a family scale by (Sourani, 1983; Antonovsky and Sourani, 1988). A copy of the FLQ, together with definitions of its components, is provided in the Appendix.

Antonovsky's original scale has been widely used, and the research indicates good reliability and validity. Cronbach's alpha on 26 separate studies ranged from .82 to .95 (Antonovsky, 1993). The scale correlated as predicted with Rotter's Internal-External Locus of Control Scale, and Studies of clinical populations of individuals show that the measure predicts to better adjustment and better recovery from illness (Langius, Bjorvell and Antonovsky, 1992; Antonovsky, 1993).

For the family scale adapted and developed for the present study, items originally referring to the individual were rewritten, taking the family as referent. About one-

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third of the items were based on the original scale; the remainder were based on a similar scale by Sourani (1983) or newly written for the study based on Reiss's theories of the family's view of the social world (1981, 1989) and Antonovsky's discussion of sense of coherence as a group property (1987). As in the original scale, items address the central components of the sense of coherence construct: comprehensibility, manageability, and meaningfulness. There is a balance between items referring to "within family" perceptions, and family perceptions about relations between the family and the larger social world. The time frame reference of the measure is extended, including the general past, present, and future. Scores on the scale have a theoretical range of 36-252. On a pilot (N=26) conducted for the present study, scores ranged from 171-231 with a mean of 205. The single, summary raw score was the measure used for this variable. Higher scores reflect a stronger family sense of coherence.

The content validity of each item of this newly adapted Family Life Questionnaire has been approved by two reviewers. For the pilot study, parents of school age children completed the FLQ and the Family Environment Scale (FES; Moos and Moos, 1994). As predicted, scores obtained on the FLQ correlated positively ($r=.40$, $p<.05$) with a factor on the FES identified by Kronnenberger and Thompson (1990) as relevant to care of children with chronic illness, in which family conflict is mediated by family organization and

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support. Split half reliability (R11) on this pilot of the FLQ was .87.

Family Routines

The frequency of the family's regular practice of shared everyday family routines involving two or more members was measured by a parental questionnaire, the Family Routines Inventory (Jensen, James, Boyce, and Hartnett, 1983).

The Family Routines Inventory (FRI) describes 28 routines thought to promote family strength and solidarity (Boyce, Jensen, James, and Peacock, 1983; Jensen et al., 1983), drawn from ten domains of family life: workday, weekend, children's routines, parents' routines, bedtime, mealtime, extended family, leaving and homecoming, discipline, and household chores (Jensen et al., 1983). Time reference for each routine is the general present, with the understanding that the routine is customary and enduring.

Each item presents a routine as a brief statement, such as "Whole family eats dinner together almost every night," "Parent(s) have sometime each day just for talking with the children," "Children do regular household chores," (Jensen et al., 1983). The parent indicates one of four levels of frequency of practice for each routine. Scoring of these frequency ratings is weighted (e.g. daily practice=3 points; 3-5 times a week=2 points; 1-2 times a week=1 point, almost never=0) to yield a maximum possible frequency score of 84. The original inventory also includes importance

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ratings for each routine; however, the authors recommend the frequency score, based on the summed frequency ratings, as the best scoring option and the most meaningful measure of adherence to routines (Jensen et al., 1983). For the present study, importance ratings were omitted and mothers completed only frequency ratings. The single, summary frequency score was the measure of the family practice of regular routines variable. Higher scores reflect more frequent practice of routines.

In development and validation of the FRI, families from a range of ethnic, racial, and socioeconomic backgrounds were included. Test-retest reliability for the frequency score of the FRI (N=271) was measured by its authors at .79 (Jensen et al., 1983). The FRI was validated through comparison studies with an instrument of known validity and reliability, the Family Environment Scale (FES, Moos and Moos, 1981; Jensen et al., 1983). The frequency score has demonstrated predictive validity in studies of preschool children's health and social functioning (Keltner, 1990, 1992). The FRI is considered suitable for families of children in the age range of the present study (Jensen, personal communication).

Qualitative Family Measure

According to the salutogenic model of child resilience proposed, family sense of coherence and the enactment of family routines foster resilience among children through a variety of intervening processes. Some of these processes

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were explored through the PKU Interview, written for the study. A copy of the interview is provided in the Appendix.

The interview was intended to provide information about qualitative aspects of families' experience in living with PKU. It is semi-structured, and includes open ended questions that relate to the two family variables, sense of coherence and family routines. Questions are usually presented in general terms to elicit the mother's spontaneous description of how the diet and its management relate to daily life. The interview also gave parents a chance to talk about how the diet affects family members, siblings, and extended family, and to describe the diet in relation to ethnic traditions, special celebrations, and personalized routines. Questions such as "Could you describe a typical weekday for X and how you do things in your family to make the diet work?" and "When you first learned about X's having PKU, what were your thoughts and feelings about what this would mean for your family?" Responses were interpreted in terms of the sense of coherence construct as it relates to the diet, and of the quality of integration of the diet routines with daily life. General areas explored are described briefly below.

Diet-Related Sense of Coherence

Diet-related sense of coherence refers to the degree to which the mother described the family's experience of the diet as comprehensible (e.g. the diet is believed to be effective and, therefore, worthy of commitment); manageable

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(e.g. resources are available for support with the diet); and meaningful (e.g. the experience of living with PKU has positive value, with the diet seen primarily as a challenge rather than a burden).

Integration of Diet Routines and Daily Life

The integration of diet routines and daily life refers to the degree to which the mother described a qualitatively "good fit" between the demands of the diet and every day family life. This means, for example, that the diet is an accepted and important part of family life, but does not overwhelm life or become an all consuming mission. Necessary diet-related routines are performed as a matter of course, the child is appropriately engaged in these activities, the diet is integrated with ongoing social life, and these experiences are imbued with positive feeling.

Child Measures

Child Social Skills

Child social skills were assessed by parental report. Mothers rated the frequency of child prosocial behaviors, using the parent form of the Social Skills scale of the Social Skills Rating System (SSRS, Gresham and Elliott, 1990), a standardized, nationally normed measure.* Each item of the Social Skills scale is presented as a brief statement of child behavior. The parent rates the frequency

*Because the SSRS, available through the American Guidance Service, Inc., is considered a secure test, sample items may not be reproduced in this context.

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with which the behavior occurs on a scale of 0-2 for each item (never; sometimes; very often). Sub-scale scores measuring behaviors indicating cooperation, assertion, responsibility, and self control are summed and converted to a total score.

The child's standard score, based on parent ratings, for the total Social Skills scale was used for this variable. Higher scores on this measure reflect better social skills.

Child Behavioral Adjustment

Each child's relative freedom from behavioral problems was assessed by parental questionnaire. Primary caregivers rated the frequency with which each child shows internalizing problems, externalizing problems, and (for children in elementary school) hyperactivity, using the parent form of the Problem Behaviors scale of the Social Skills Rating System (SSRS, Gresham and Elliott, 1990). Internalizing problems reflect anxiety, depression, and poor self-esteem; externalizing problems reflect aggression and poor self control. Each item on this scale is presented as a brief statement of a problem behavior, and the parent rates its frequency on a scale of 0-2 (never; sometimes; very often).

The child's standard score, based on the parent ratings, for the total Problem Behaviors scale was used for this variable. Lower scores on this measure reflect fewer reported problems and better behavioral adjustment.

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The SSRS is a relatively new instrument, now widely used to assess school age children, including some studies of children with chronic illness. It was standardized on a national sample of 4,170 preschool and school age children representative of the U.S. population. The Preschool and Elementary School Levels were used for this study. Each Level includes the two scales, Social Skills and Behavior Problems. The primary emphasis is on prosocial behavior; for example, the Preschool Level has 39 items measuring social skills and 10 items measuring problems. The manual reports extensive validity and reliability data (Gresham and Elliott, 1990). For example, test-retest reliability coefficients were .87 for Social Skills and .67 for Problem Behaviors. Median internal consistency reliablities were .90 for Social Skills and .84 for Problem Behaviors. The SSRS is especially appropriate for this study because it measures positively stated prosocial skills and behaviors, which is in keeping with a study of resilience. Each of the scales has a mean of 100 and a standard deviation of 15. Separate norms are provided for boys and girls in each of the two age groups, preschool (ages 3 to 5 years) and school age (6 to 11 years). For each of the scales, the standard scores are comparable across sex and age groups and may be treated as such for statistical purposes (Personal communication, American Guidance Service Test Consultation Department).

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Child School Competence

For school age children ages 6 and older, each child's school performance in academic subjects, regular or special class placement, and normal or delayed progression through grades was assessed by parental questionnaire. Mothers completed the School Competence sub-scale of the Competence Scales of the Child Behavior Checklist (CBCL/4-18, Achenbach, 1991; see Appendix). The child's T score on the School Competence sub-scale was used for this variable. Higher scores reflect better school competency. Separate norms are provided for boys and girls in this 6-11 age bracket, and the generated T scores are comparable across sexes.

The CBCL/4-18 has been widely used among both normal children and children with chronic illness to assess social competency and behavior problems. Extensive validity and reliability information is reported in the manual (Achenbach, 1991). Only the School Competence sub-scale was used for this study. According to one study (Reber, Kazak, and Himmelberg, 1987), the scale that includes this sub-scale, the Competence (formerly Social Competency) Scale, may have special utility in assessing differences between children with PKU and their peers.

Dietary Adherence

Degree of long term phenylalanine control (phe level) was taken as an index of dietary treatment adherence. Mean phenylalanine (phe) level for the year for each child was

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chosen as the summary measure for the dietary adherence variable. Data for this variable were drawn from each child's medical record of phenylalanine levels measured in mg/dL (milligrams per deciliter) by an amino acid analyzer in a standard laboratory test. Lower phenylalanine levels represent better adherence. For each child, all measures taken over the year as a routine part of treatment prior to and including the time the parent completed the questionnaires were included, and the mean for each child was calculated. Children with PKU in the present study receive such serum tests on the average of once per month; but for a given child, intervals may range from once per week to several times a year. Prescribed frequency of tests takes into account the child's age, past stability of metabolic control, growth pattern, treatment adherence history, and response to prescribed dietary changes.

While the ideal assessment of phenylalanine control for research purposes might involve random monthly assessments of phenylalanine in all study children, this would place an undue burden on those children whose history of long term good control and high stability permits the use of fewer of these painful tests. In some cases, random monthly assessment would also be incompatible with the clinical needs of those children from whom more frequent tests are needed to better assess compliance and nutritional issues. The retrospective nature of the present study allowed assessment of hypotheses without disrupting clinically

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indicated schedules and without burdening patients, since no blood was drawn specifically for the study.

The pattern, stability, and overall level of a child's phenylalanine levels over time are considered a good approximate measure of dietary adherence. If the diet is followed, levels can be controlled. There are individual differences in response to dietary adjustments, and families must make differing adjustments to achieve control, but all receive individualized counseling and an adjusted diet that suits the child's needs. Illness can cause elevated levels, and parents are instructed not to draw blood during this time, or to indicate when they have done so. Otherwise, phenylalanine levels in young and school age children are relatively stable, affected by diet but not by time of day or amount of exercise. In short, phenylalanine level and control over time is considered a "best available" index of dietary treatment adherence and is clinically interpreted as such. Researchers have also taken individual mean phenylalanine level as a valid measure of compliance (Fehrenbach and Peterson, 1989), and physicians' ratings of compliance show high correlation with metabolic control (Shulman, Fisch, Zempel, Gadish and Chang, 1991).

The choice of mean phenylalanine level as the best available measure of overall phenylalanine control was based on analysis of an existing yearly review of data of 32 children at one center during 1996. In order to assess the reliability of the mean as a measure, it was compared with

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another measure, the percentage of values >10 mg/dL for the year for each child, using the 1996 yearly review data. Although >10 mg/dL exceeds the ideal range, this level is often interpreted as a marker for the need of clinical intervention for children the age of those in the study. For the 32 children evaluated in the 1996 preliminary study, child mean phenylalanine level and percentage over 10 mg/dL for each child were highly correlated, with Pearson's correlation calculated at .92. While not identical, these two measures were generally interchangeable. This meant that, in using the mean for the correlational statistical tests, one did not need to be concerned that a single extreme value would have a marked effect on overall conclusions to be drawn about phenylalanine control. In summary, the mean phenylalanine level for each child was considered to have adequate reliability and validity.

As a continuous variable that reflects each child's phenylalanine control relative to that of all other children, the mean permits more powerful use of correlational statistical procedures than would other measures of child phenylalanine control. The mean was therefore treated as a continuous variable in all statistical tests involving the whole study sample. For the descriptive interpretation of interview data based on a subset of the sample, data were dichotomized, with children classified as in good (M phe <10 mg/dL) or poor (M phe >10 mg/dL) dietary adherence.

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Control Measures

Child age

Child age of subjects in the study ranged from 3 years to 11 years 11 3/4 months, with a mean age of 90.03 months. Because of this very broad range, it was important to rule out age as a confound through including it as needed in statistical tests. In addition, standardized measures that included child age in norms were chosen whenever possible.

Family social position

Hollingshead's Two Factor index of Family Social Position (Hollingshead, 1957; reprinted in Miller, 1991, pp. 351-359) was used as a measure of position the family occupies in the socio-economic status (SES) structure. The index is based on the head of household, and weights two factors, education (weighted with a factor of 4) and occupation (weighted with a factor of 7), to yield a classification in one of five social classes. Class I is the highest, Class V the lowest. Statistical tests were used to determine the role of social class as a potential confound in measures of association, and its role as a predictor of child outcomes.

Procedure

The study was approved by the Internal Review Board of the sponsoring site, Center #1, and by the medical directors of all participating sites. Parents at Center #1 were contacted initially by the researcher. Parents at other centers made the initial contact with the researcher

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following receipt of mailings sponsored by their treatment programs. Packets of questionnaires were mailed to each primary caregiver and returned by mail to the researcher. Informed consent was obtained from each participating parent.

Thirty-four mothers completed a demographic sheet, consent form, and questionnaires: the Family Life Questionnaire (FLQ) measuring family sense of coherence; the Family Routines Inventory (FRI) frequency scale measuring family practice of conventional routines; and the Social Skills and the Behavior Problems scales of the Social Skills Rating System (SSRS). Twenty-three mothers from within this group, parents of study children age six years or older, also completed the Competence Scale including the School Competence sub-scale of the Child Behavior Checklist (CBCL 4/18). Fourteen mothers who agreed to be interviewed by the researcher using the PKU Parent Interview were seen individually by the researcher in their homes. Please see the Appendix for copies of materials developed for the study, consent form, and demographic sheet.

Medical directors provided records of each participating child's phenylalanine readings from all blood specimens taken during the previous year dating from, and continuing to, the date the parent completed the questionnaires. Medical directors or designated staff resolved questions about interpretation of blood phenylalanine data, and provided information about the

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number of eligible families and the representativeness of the sample from each center.

Data Analysis

Descriptive analysis of all study variables is given in Table 3. For each variable, distribution was essentially normal and the degree of skew was within the limits appropriate for the statistical tests that were used.

Table 3

Descriptive analysis of study variables (N=34)

Variable	Measure	M	SD	Min	Max
Fam sense coherence**	FLQ	191.27	29.11	126	245
Family routines	FRI	62.35	9.80	38	77
Social skills	SSRS	102.15	15.93	75	130
Problem behaviors-	SSRS	97.79	12.97	85	131
School competence***	CBCL	42.91	9.28	27	55
Diet adherence**-	M Phe	10.24	3.50	3.7	16.4

-Lower scores indicate better function

**Data missing for one case

***Includes only those children age 6 years and older (N=23)

Pairwise missing value method was used for all statistical tests. Missing values included the FLQ score for one subject, and the M phe value for another. This meant, for example, that for the whole group, 32 comparisons were made between the M phe and the FLQ and FRI measures, and 34 between the FRI and the SSRS measures.

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Before the hypotheses were tested, intercorrelations were checked to identify potentially confounding variables. All tests for confounds were conducted on the entire sample, two tailed, with alpha=.05. In accordance with standard statistical practice, a control variable was to be considered a confound if it correlated with both predictor and outcome variables with a strength of r>.25, or was significantly correlated with both. None of the four control variables (child age, child sex, family social position, mother's education) correlated at this strength with either of the family predictor variables (family sense of coherence and family practice of routines). Absolute values of correlations ranged from .01 (p=.96) to .17 (p=.36). Neither child age nor child sex control variables correlated at criterion level with the child outcome variables; however, family social position and mother's education did correlate with some of the child outcome variables. The association of these two control variables with all child outcome variables is given in Table 4.

All tests for hypotheses I, II, and III were one sided (one tail), using alpha of 0.05. The use of one tail tests is appropriate in a study such as this, in which the direction of all hypothesized outcomes is predicted a priori on theoretical ground.

Bivariate correlations with Pearson's r were used to test Hypotheses I, the predicted positive association of the two family strengths. For Hypotheses II and III, bivariate

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correlations with Pearson's r were also used to test the strength of association of each of the family predictors, singly, with each of the child outcome measures. No adjustment for control variables was made, because in no case did a control variable correlate at or above criterion level with both predictor and outcome variables. Thus, probable confounds were ruled out and the use of zero order symmetrical correlational tests was appropriate.

Table 4

Correlation of Mother's Education and Family Social Position with Child Outcome Variables (N=34; School Competence N=23)

M education

Soc Skills	Prob beh	School	M Phe
-.4821**	-.0246	-.0746	.2891
p=.004	p=.890	p=.735	p=.103

Fam Soc Position

Soc Skills	Prob beh	School	M Phe
-.2628	.1321	-.2011	.4709**
p=.133	p=.456	p=.358	p=.006

**p<.01 (Two tail tests used to determine confounds)

Note: On mother's education and family social position, lower scores= higher education and higher social position. On Mean phenylalanine level (M Phe) lower scores=better control.

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Following hypothesis testing, multiple regression analyses were used to explore possible interactions and to assess the relative strength of family and social variables as joint predictors of child psychosocial resilience and dietary adherence. Using a combination of forced entry and stepwise analyses, each of the five child outcome variables was regressed on the family variables and the social control variables. Because of small sample size and weak statistical power, these regressions were considered exploratory, with effect size (Beta >.20) used in addition to F significance as a guide to assess influence of a predictor variable (Morris and Fitz-Gibbon, 1978). Relations between family social position, family sense of coherence, and dietary adherence were further explored through description of patterns within the data set.

Qualitative data from follow-up interviews were then related to Hypotheses IId and IIId. That is, mothers' descriptions of diet-related sense of coherence and mothers' descriptions of integration of diet routines with daily life were each related to the corresponding general family predictor variables, and to dietary adherence as indicated by child phenylalanine level.

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CHAPTER FOUR

RESULTS

Review of the Study

This study of 34 families with children who have a phenylketonuria (PKU), a rare metabolic disorder, hypothesized that the two general family strengths, family sense of coherence and family practice of regular routines, would be positively associated with one another; and, that each of the two strengths would be positively associated with children's psychosocial resilience and dietary adherence. The study further proposed that mothers' descriptions of living with PKU would reflect the two general family strengths in terms that were specific to the diet, and that such "diet-related" sense of coherence and "diet-related" family routines would be associated with dietary treatment adherence.

Family predictor variables, based on summary scores of questionnaires completed by mothers, included: (a) family sense of coherence, measured by the Family Life Questionnaire (FLQ), and (b) family practice of regular routines, measured by the Family Routines Inventory (FRI).

Child outcome variables were based on summary scores of scales completed by mothers, and upon each child's medical records. The child outcome variables based on maternal report included: (a) prosocial skills, measured by the SSRS Social Skills scale, (b) behavioral adjustment, measured by

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the SSRS Problem Behaviors scale, and (c) school competence, measured by the CBCL 4/18 School Competence sub-scale of the Competence Scale. The fourth child resilience outcome variable, (d) dietary adherence, was based on the medical record of each child's mean phenylalanine level in mg/dL over a year's time. Control variables included family social position (SES) and mother's education as measured by Hollingshead's Two Factor Index.

Information about family attitudes towards diet-related aspects of each of the general family strengths was provided by a sub set of mothers (N=14) in the study who participated in a semi-structured follow up individual interview, the PKU Parent Interview, written for the study.

Direction of all hypothesized outcomes was predicted a priori on theoretical ground. All statistical tests used alpha of 0.05, one sided. Bivariate correlations were used to test hypotheses, including the correlation between the two family strengths, and the correlation between each of these strengths and the child outcome measures. In hypothesis testing, a relatively even influence of social position was assumed.

Relations among family social position, family sense of coherence, family practice of regular routines, and dietary adherence were then explored through multiple regression analyses and through descriptive methods. Finally, clinical interpretation of interview and anecdotal material was used to show how mothers' descriptions of diet-related sense of

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coherence and their descriptions of the quality of diet-related routines were each associated with the general family strengths and with child dietary adherence.

Hypothesis Testing

Hypothesis I: Association between Family Strengths

Hypothesis I stated: Family sense of coherence will correlate positively with family practice of regular routines.

As predicted, there was a significant positive correlation between these two areas of family function as measured by the Family Life Questionnaire (FLQ; adapted from Antonovsky, 1987, and Sourani, 1983) and the Family Routines Inventory (FRI, Jensen et al., 1983), respectively, ($r=.5005$, $p<.01$). The moderately strong correlation obtained indicates that mothers who viewed family life and the place of the family within the social structure as coherent in terms its of manageability, meaningfulness and comprehensibility also tended to report more frequent practice of conventionally valued everyday family routines. Conversely, mothers who saw their familial and social world as less coherent tended to report less frequent practice of everyday family routines. Hypothesis I was sustained.

Hypothesis II: Association between Family Sense of Coherence and Child Outcomes

Hypothesis II stated: Family sense of coherence will correlate positively with dimensions of child resilience including (a) social skills (SSRS Social Skills scale), (b)

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behavioral adjustment (SSRS Problem Behaviors scale), (c) school competence (CBCL 4/18 School Competence sub scale, and (d) dietary adherence (child year mean phenylalanine level).

The relation of mothers' perception of family sense of coherence as reported on the Family Life Questionnaire (FLQ) to the four child outcomes was examined. The predictor variable was the FLQ summary score. In bivariate correlations, family sense of coherence as perceived by mothers correlated significantly with all four child resilience outcome measures, and all were in the predicted direction. Pearson product moment correlations are reported in Table 5. Higher scores reflect better child function on measures of social skills and school competence; positive correlations were, therefore, expected between family sense of coherence and each of these measures. Lower scores reflect better child function on measures of the remaining child outcomes, behavioral adjustment as indexed by frequency of reported behavior problems, and dietary adherence as indexed by mean phenylalanine level; negative correlations were, therefore, expected between family sense of coherence and each of these measures.

Hypothesis IIa

Mothers' perceived family sense of coherence as measured by the summary score of the Family Life Questionnaire (FLQ) correlated positively with their report of their children's prosocial skills as measured by the

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SSRS Social Skills scale summary score comprising measures of cooperation, assertion, responsibility, and self control (r=.46, p<.01; see Table 5). Hypothesis IIa was sustained.

Table 5

Hypothesis II a, b, c, d: Correlation of Family Sense of Coherence with Child Outcomes

(Social skills, problem behaviors, dietary adherence variables N=34; School competence variable N=23

Soc Skills	Prob Beh	School Com	Diet Adh (M Phe)
.4583**	-6802***	.3599*	-.3047*
p=.004	p=.000	p=.050	p=.045

*p=.05 **p<.01 ***p<.001

Hypothesis IIb

Mothers' perceived family sense of coherence correlated negatively with problem behaviors as measured by the summary score of the SSRS Problem Behaviors scale (r=-.68, p<.001; see Table 5); mothers who saw family life within society as more strongly coherent reported that their children had significantly fewer internalizing and externalizing behavior problems than did mothers who saw family life as less coherent. Hypothesis IIb was sustained.

Hypothesis IIc

Mothers' perceived family sense of coherence correlated significantly and positively with school competency as

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measured by the School Competence sub scale of the CBCL 4/18 (r=.36, p=.05; see Table 5). Mothers who saw family life as more strongly coherent reported that their children were achieving higher grades, making better progress, and requiring fewer special educational services than did mothers who saw family life as less coherent. Hypothesis IIc was sustained.

Hypothesis IId

Finally, as predicted, mothers' perceived family sense of coherence correlated significantly and negatively with children's serum phenylalanine levels, measured by each child's mean phe level in mg/dL for the preceding year (r=-.30, p<.05; see Table 5). This means that, in general, children of mothers who reported stronger family sense of coherence had lower phenylalanine levels, reflecting better dietary adherence, than did children from families whose mothers reported weaker family sense of coherence. Hypothesis IId was sustained.

Taken together, these bivariate correlational findings provide consistent support for Hypothesis II. When it is considered as a single predictor and a relatively even influence of other variables such as social position is assumed, mothers' perceived family sense of coherence correlates significantly as predicted with mothers' report of the three child psychosocial resilience outcomes, and with dietary adherence as measured by mean phenylalanine level.

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Hypothesis III: Association between Family Practice of Regular Routines and Child Outcomes

Hypothesis III stated: Family practice of regular routines (FRI) will correlate positively with dimensions of child resilience including (a) social skills (SSRS Social Skills scale, (b) behavioral adjustment (SSRS Behavior Problems scale), (c) school competence (CBCL/4-18 School Competence sub scale), and (d) dietary adherence (child year mean phenylalanine level).

The relation of family practice of regular routines, reported by mothers, to each of the four child outcome variables was examined. The predictor variable was the frequency score on the Family Routines Inventory (FRI, Jensen et al., 1983). Routines measured by the FRI are not specifically related to chronic illness, but are the shared, conventionally valued actions any family might practice, such as taking regular trips, performing chores, and eating dinner together on a regular basis. Pearson product moment correlations for Hypothesis III are given in Table 6. Positive correlations were predicted between the family routines and two child outcomes, social skills and school competence. Because lower scores represent better child function on the measures of behavioral adjustment and dietary adherence, negative correlations were predicted between these variables and the measure of family routines.

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Hypothesis IIIa

Family practice of regular routines as measured by the FRI correlated significantly in the predicted direction with children's prosocial behaviors as measured by the SSRS Social Skills scale ($r=.41$, $p<.01$; see Table 6). Mothers who reported that their families frequently engaged in conventionally valued routines also reported that their children more frequently engaged in prosocial behaviors involving cooperation, assertion, responsibility, and self control than did mothers who reported less frequent practice of regular family routines. Hypothesis IIIa was sustained.

Hypothesis IIIb

Family practice of regular routines as measured by the FRI correlated significantly in the predicted direction with child problem behaviors as measured by the SSRS Problem Behaviors scale ($r=-.30$, $p<.05$; see Table 6). Mothers who reported more frequent practice of conventionally valued routines reported that their children had significantly fewer internalizing and externalizing problems than did mothers who reported lower levels of shared family interactions. Hypothesis IIIb was sustained.

Hypothesis IIIc

Family practice of regular routines as measured by the FRI correlated in the predicted direction with their report of the children's school competence as measured by the CBCL/ 4-18 School Competency sub-scale at the level of a slight trend ($r=.23$; $p=.15$). Thus, compared with mothers who

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reported less frequent practice of routines, mothers who reported frequent practice of routines were slightly more likely to report that their children were making better progress in school; however, the correlation did not reach statistical significance. Hypothesis IIIc was, therefore, not sustained.

Hypothesis IIId

Family practice of regular routines as measured by the FRI did not correlate with dietary adherence (r=-.04; p=.41; see Table 6). For Hypothesis IIId, the correlation obtained was close to the level of chance. The performance of predictable routines that are not specifically related to the PKU diet, but that are the conventionally valued activities that any family might practice, did not predict to lower (i.e. better) levels of phenylalanine, the measure of dietary adherence. Hypothesis IIId was, therefore, not sustained.

Table 6

Hypothesis III a, b, c, d: Correlation of family routines with child outcome variables

(Social skills, problem behaviors and dietary adherence N=34; School competency N=23)

Soc skills	Prob beh	School prog	Diet Adh (M Phe)
.4127**	-.3032*	.2273	-.0434
p=.008	p=.041	p=.149	p=.405

*p<.05 **p<.01

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These results provide moderate support only for those aspects of Hypothesis III that relate mothers' report of family routines to their report of their children's social skills and problem behaviors. In each case, the absolute value of the correlation, while significant, is weaker than that of the corresponding value found for Hypothesis II. This suggests that, compared with frequency of family routines as measured by the FRI, degree of family sense of coherence as measured by the FLQ is the better predictor of the child outcomes.

Related Findings

Relative Strength of Family Predictor Variables in the Presence of Social Variables

In order to assess the relative strength of the two family predictors in the presence of the two social variables (mother's education and family social position) that had been found to correlate with specific child outcomes in the preliminary data analysis, a series of multiple regressions was conducted, using a combination of forced entry and stepwise methods.

This second approach to the data differs the first approach, taken in testing Hypotheses II and III. The first approach asked the question, what is the strength of association, and how important are family sense of coherence and family practice of routines when each is used alone to predict child outcomes? (Norusis, 1993) Asked in this

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manner, the question forefronts the clinically relevant variables while keeping other factors that might influence child outcomes in the background. This question was answered by looking at the bivariate correlations between each of the two family variables and the child outcomes (Norusis, 1993).

The second approach asked the question, how important are family sense of coherence and family practice of routines when they are used together along with other variables, such as parents' education and social position, to predict child outcomes? (Norusis, 1993) Asked in this manner, the question contexts the clinical variables within a social frame that is itself considered part of the picture to be interpreted. The second question may be answered by drawing inferences from the outcome of regression equations taking the child outcomes as dependent variables, and including the family predictors along with other relevant variables (Norusis, 1993).

Findings are considered exploratory, because the power for such analysis is weak in a sample of this size. All tests were one-sided with alpha of 0.05. For the purposes of evaluation in the context of sample size, effect sizes (Beta) with absolute values >.20 were considered to indicate influence as trends, even when significance was not reached. (Morris and Fitz-Gibbon, 1978).

The possibility that, despite its relatively weak showing in the bivariate correlations, the family routines

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variable might function synergistically with family sense of coherence was considered. An interaction variable was tested to see if it had sufficient strength ($p<.05$) to enter the regression equations, first in the presence of the two family predictor variables, and then in the additional presence of any subsequently entered social variables that remained in the equations. On no occasion did the interaction variable gain entry under the stepwise condition, therefore the evidence argues against a synergistic effect.

In the presence of the family sense of coherence variable and family social variables, family routines retained influence as a trend for only one of the dependent variables, child social skills (Beta=.244, $p=.067$). For the remaining dependent variables, family routines effect size was low, with no Beta greater than .085 and probabilities at or near the level of chance.

In contrast, family sense of coherence retained significance or influence as a predictor of child psychosocial variables, even when effects of other variables were taken into account. Effect size (Beta) and significant F statistics for family sense of coherence and child psychosocial outcome variables are given in table 7.

Family sense of coherence effect size was significant for social skills (Beta=.274, $p<.05$ and for problem behaviors (Beta=-.672, $p<.001$). Family sense of coherence effect size retained influence as a trend for school

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competency (Beta=.312, p=.12). Social control variables including social position and mother's level of education played no significant role in prediction of child psychosocial resilience with one exception: in the presence of the two contributing family predictor variables (family sense of coherence and family routines), mother's education had a significant effect on prediction of social skills (Beta=.457, p<.01).

Table 7

Family Sense of Coherence Effect Size and F Significance in Multiple Regressions for Child Psychosocial Resilience

Variable	Beta	p
Social skills	.273519*	.0484
Problem behaviors	-.672068***	.0001
School competency	.312193	.1205

*p<.05 ***p<.001

Social skills & prob behaviors N=34; school competency N=23.

The situation was different for the dietary adherence child outcome variable. Here, family social position emerged as the strongest predictor of dietary adherence, with a significant effect size (Beta=.381, p<.05). In the presence of this relatively strong predictor, family sense of coherence did not reach significance, but retained influence as a trend predicting adherence (Beta=-.237, p=.12); with greater effect size when an outlier in this regression was removed (Beta=-.303, p=.057).

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Taken together, these exploratory regression analyses tend to support and qualify findings obtained in hypothesis testing using bivariate correlations. The regression analyses indicate that the evidence does not support an association between frequency of practice of family routines and child dietary adherence. Family sense of coherence emerges as the better of the two variables under study as a predictor of measures of child psychosocial resilience and dietary adherence. Its effect size as a predictor of adherence is at the level of a trend, overshadowed by that of family social position.

Family sense of Coherence, Family Social Position, and Dietary Adherence in the Sample

In order to further explore relations among the family sense of coherence, family social position, and dietary adherence variables, patterns in the study sample were examined. These patterns are based on descriptive rather than upon inferential statistics, and apply to the sample.

Correlations between family sense of coherence and phenylalanine level/dietary adherence were based on 32 comparisons. When these comparisons were examined descriptively by social position (SES, based on Hollingshead's two factor index), breakdown by SES Class membership was: Class I, N=2; Class II, N=7; Class III, N=11; Class IV, N=12.

Taking the clinically relevant M phe >10 mg/dL as the division between "good" and "poor" phenylalanine control

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groups, and the mean group FLQ (family sense of coherence) score of 191 as the division between "strong" and "weak" family sense of coherence, two statements comparing subjects above and below middle class (Class III) could be made:

Of those subjects of higher SES (Classes I and II) with strong family sense of coherence, 83% (5 of a total of 6) had good phenylalanine control.

Of those subjects of lower SES (Class IV) with weak family sense of coherence, 83% (5 out of a total of 6) had poor phenylalanine control.

In the two adjacent classes of similar size (Class III, N=11; and Class IV, N=12), no family with a family sense of coherence (FLQ) score below 180 was in the good dietary control group, regardless of class membership. That is, of the 8 families with family FLQ score below 180, all 3 Class III and all 5 Class IV subjects were in the poor control group.

Of the remaining 15 Class III and IV families each with an FLQ score >180, 60% were in the good control group. In this set of 15 families with FLQ scores each >180, the proportion of those in good control differed between the two classes. In Class III, 75%, or 6 of these 8 subjects were in good control; in Class IV, 42% or 3 of these 7 subjects were in good control.

Inferences about these descriptive statements are drawn in the discussion.

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Relations among General Sense of Coherence, Diet-Related Sense of Coherence, and Dietary Adherence

The systems theory proposed by this study implies that mothers' general perceived family sense of coherence will be reflected in their attitudes towards the diet, and that such "diet-related" sense of coherence will be associated with dietary adherence. Interpretation of qualitative data addressed this assumption.

Many mothers spoke spontaneously of their experience with the diet during the course of data collection, and a subset of 14 mothers were individually interviewed, 7 with children with good adherence (M phe <10 mg/dL) and 7 with children with poor adherence (M phe >10 mg/dL). Among those interviewed, distribution of FLQ scores fell into two modal groups, 9 with "higher" scores (>185) and 5 with "lower" scores (<170). Six of the 7 mothers in the good adherence group had higher FLQ scores; 3 of the 7 mothers in the poor adherence group had higher FLQ scores.

Mothers of children differed in the kinds of attitudes towards the diet they expressed, and in their diet related behaviors. As illustrated in the following section, these differences could be classified in terms of degree of diet-related sense of coherence, using the dimensions of the sense of coherence construct. Mothers of adherent children saw the diet as comprehensible; it made sense, was predictable, and was worthy of commitment. They saw the diet

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as manageable; resources were available to help cope with problems. They saw the diet as meaningful; they maintained a positive attitude towards it and perceived it as a challenge rather than a burden. These mothers had a strong diet-related sense of coherence. In contrast, mothers of children with poor adherence were likely to be experience difficulty in one or more of these dimensions; in the terms of this study; these mothers had weaker diet-related sense of coherence.

The quality of mothers' diet-related speech was usually consistent with the general level of sense of coherence measured by the FLQ; however, 2 of the 3 mothers with higher FLQ scores whose children were poorly adherent spoke of their experience with the diet in terms that were more similar to those of mothers with lower FLQ scores. In 12 out of 14 cases, however, diet-related sense of coherence was closely related to family sense of coherence as measured by the FLQ. When there were differences between the two, diet-related sense of coherence was the better predictor of adherence.

Qualitative aspects of diet-related sense of adherence associated with good and poor adherence are given below. In the vignettes, names, initials, and personally identifying information have been altered to preserve confidentiality. The qualitative aspects manifest in the "good adherence" group are discussed first.

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Diet-Related Sense of Coherence and Good Adherence

High commitment and comprehensibility. All mothers of children with good adherence, without exception, made statements indicating that they believed that the diet made sense and was worthy of commitment. Belief in the diet and commitment to it were closely intertwined. These mothers believed that the diagnosis was correct, that adherence affected phenylalanine levels, and that levels within treatment range were a necessary condition for the child's normal development. They believed their behavior affected treatment, and that they could usually predict their children's phe levels. These mothers' commitment included expectation of success and a sense of efficacy in its attainment. The following cases exemplify the commitment and belief in the diet of mothers of adherent children.

> Mrs. S., a middle class working mother whose youngest child has PKU, describes herself as "100 per cent" committed to the diet, even though there are constant adjustments because her 4 year old daughter is a "picky eater" with a low phe tolerance. During her pregnancy, Mrs. S. had a premonition that "I would have a special needs child, and that it would be something we could handle and it would be O.K." She and her husband never doubted the diagnosis or their ability to follow the diet. Convinced that lower phe levels meant better outcome, they successfully negotiated with the PKU center to devise a diet that would keep their daughter in the lower half of the accepted treatment range. When test results arrive, they are almost always what she expects them to be. Mrs. S. credits the diet with saving her child from retardation. She describes her child as very social and intellectually gifted. "If she were not on this diet, she would not be who she is."

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For some mothers of adherent children, belief in the diet and commitment to its efficacy were hard won.

> Mrs. L., a waitress, struggled to cope with the demands of the diet after several years of poor compliance were associated with measured cognitive decline. She believes that she has benefited from treatment interventions including support from the PKU center and the local CMHC. Her son J., now in second grade, is now doing well on the diet. Mrs. L. expresses some anxiety about capacity to deal with problems when J. becomes a teen ager. Nevertheless, she is strongly committed ("to the utmost") to the diet. She believes that her son's lowered phe levels, due to improved care over the past two years, have resulted in J.'s no longer needing special class placement. "It [the diet] works...he can read now...He is doing really good in school." Of the diet's fit with every day life she says, "It has to fit. It just does." Her actions and words imply a developing sense of agency and efficacy. "I write [all foods] down...I keep after him with the milk [formula]...I believe if he drinks all the milk, he will be successful."

Satisfaction with resources. Mothers of children with good adherence were satisfied with the resources available for support with the diet.

The social position (SES) of interviewed mothers of children with good adherence ranged from Class I to Class IV, with a modal position of III. Satisfaction with financial resources was implied rather than stated. Well to do parents used the computer network, bought bread machines, and worried that they might run low on special foods, not whether they could afford them. No mother in this group, including those in Class III and the mother in class IV, cited financial constraints affecting dietary adherence.

The mothers in this group described a variety of characterological, spiritual, and social resources that

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contributed to their effective coping with the diet: "Faith in God and a sense of humor." "We [her husband] balance each other; he's like, just do it, and I am more emotional." In this good adherence group, just one mother, the only mother in the group with a low FLQ score, expressed feelings of isolation and anxiety about familial social support. Among the remaining mothers, degree of reliance on social support varied. Some mothers felt that, while they did most of "diet work" themselves, they received adequate emotional and material support from an immediate family member. Others relied on a broad network. All these remaining mothers believed that support they received was predictable; they could call upon it if it were needed. Every mother in this group who was asked said that she would turn to the clinic for help should her child have a series of high blood levels. Social supports included family, grandparents, friends, teachers, clinic staff, visiting nurses, and social workers. An example illustrates the variety of resources contributing to the good coping among these families.

> Mrs. C., a nurse's aide, felt deeply supported by her family and especially her husband at the time of diagnosis and thereafter. She described his helping with the formula, "many a night, at this very counter," and his decorating the birthday PKU cakes that she baked. Her parents and brother took turns accompanying the couple and newborn to the clinic to learn more about the diet. She trusts her sister, who lives nearby and knows the diet ("She even subscribes to the PKU Newsletter") to give her 9 year old child a meal "any day." Mrs. C. is glad that state provision of formula has prevented the family from having to "double and triple mortgage the house." She describes the clinic experience as "having this whole backbone." "It wasn't 'Here's you PKU baby, we can't help you. It was 'Here's all this information...And here's this

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> doctor...a great doctor...and here's a nutritionist!'" When she was told that new research recommended lifelong diet (rather than discontinuation at age 7, found to be associated with cognitive decline), this mother sensed an ultimate support that had coordinated the timing of the research and her child's birth: "It was like another, 'Thank you, God!'"

Positive Attitude towards Living with PKU. Mothers of children with good adherence described their experience of living with PKU in a generally positive manner, as a challenge rather than as a burden. They admitted that the diet complicated life and involved work and inconvenience, but 6 out of 7 offered positive or neutral descriptions of its fit with daily life: "It fits easily." "A challenge--not a bad challenge, just a challenge." "It's routine." "Comfortable." The most negative comment by the seventh mother was that the diet fit into life "with difficulty."

Those mothers in this group who had endured a time of deep grief at the time of the child's diagnosis felt they had gotten past that time, and some felt that the family's strengths had been revealed through the experience. ("It made us closer." "My mother amazed me.") Several mothers spontaneously offered detailed, positive statements about the child, often invoking family identity and conveying acceptance of the child's PKU condition, such as "I'm a reader, and he's reading now, I am so thrilled," "She has such beautiful skin, I think it's the PKU..."; and, of an adopted child, "She does everything, Brownies, trampoline. I wish you could see her, oh, here's her picture [with the

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other children], see, she fits right in." Virtually every mother in this good adherence group stated, in one way or another, that the child was living proof of the worth of the diet and the effort it involved.

In summary, mothers of children with good adherence expressed a strong diet-related sense of coherence. They saw the family experience of the diet as comprehensible, manageable, and meaningful.

Diet-Related Sense of Coherence and Poor Adherence

Compared with the good adherence group, mothers of children with poor adherence displayed greater variation in the quality of their diet-related attitudes. For example, a given mother might express dissatisfaction with her social support system, while stating a high commitment to the diet. Each mother's pattern was different, but each had one or more areas in which their attitudes were less adaptive, or their situation was more difficult, than those of the mothers in the good adherence group.

Ambivalence of commitment and belief in the diet. Two of the mothers in the poor adherence expressed a level of commitment to the diet that was comparable to that of that of the mothers of adherent children. The remaining 5 mothers professed commitment, but their comments and described behavior suggested ambivalence. In this group, one mother with a with a high FLQ score professed full commitment to the diet but subtly distanced herself from its enactment,

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leaving adult responsibility to her child in such matters as negotiating with the school staff about dietary conflicts. One mother permitted occasional "cheating" on the diet. Two delayed doing the blood work essential in monitoring the diet. Two questioned the nature of the diagnosis, one stating an expectation that her child would outgrow PKU and the need for formula, another that the PKU could only have come from the other side of the family. One mother doubted that her child's high phe levels related to diagnosed cognitive deficits.

Although all these mothers worked very hard at some aspects of the diet, they ignored or worked ineffectively at others, such as regularly adjusting the day's phenylalanine intake. They were then mystified and frustrated by the treatment outcome. For these mothers, as for mothers of adherent children, belief in the diet and commitment to it were closely intertwined; but for most mothers in the poor adherence group, the diet did not "make sense." These mothers also were less likely to describe a sense of personal agency in their handling of the diet.

> Mrs. E. attributes her 7 year old daughter's grade repetition and learning disabilities partly to her elevated phenylalanine levels. Nevertheless, she doubts that more assiduous adherence to the diet will lower the levels, believing that there are many other factors that contribute. Because her child drinks the formula and she works hard to provide the right foods, Mrs. E. is convinced that she is fully adherent, but to little avail: "I do everything right, and [she never cheats on the diet], but she still keeps having the high levels...it's like a puzzle, you got to put it together, but if the pieces are wrong, you can't." Mrs. E. is unable to see how some of her own behavior might contribute to these puzzling outcomes. For

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example, she rarely calls the clinic for needed advice, and finds measuring food too difficult. Without some success, she is denied the positive feedback that might enhance her sense of effectiveness and participatory agency, and the therapeutic alliance suffers. Much of the work of the PKU diet is experienced as imposed upon her by others. "They tell you what you should do, they tell you how to do it." Like many other parents in her situation, she expresses doubt that anyone who does not have a child with PKU can understand the difficulties of the diet, which must be endured day after day.

Conflicts in support and dissatisfaction with resources. Mothers of children with poor adherence were less satisfied with resources, and, in some cases, less likely to rely upon support when it was seemingly available. They were also far more likely to experience conflict within one or more support systems.

Family social position (SES) of interviewed mothers of children with poor adherence ranged from Class II to Class IV, with an equal number of families in Classes III and IV. Two out of 3 mothers in Class III and 2 out of 3 in Class IV cited financial constraints affecting the diet. They received formula without difficulty, but had trouble obtaining the special foods. This was a special hardship for mothers of active school age children who needed solid foods to provide calories and sense of fullness not provided by the otherwise nutritious diet formula. These mothers frequently expressed their fear that state funds for formula could be cut ("You say to yourself, My God, what's next, no more formula?"). In some cases, these fears escalated to a more general sense of privation. For example one mother felt

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that research money went to all illnesses except PKU. Progress in genetic research in other areas brought little comfort to those who waited for progress in PKU research.

Mothers in this group sometimes did not use available support. For example, just 2 out of 7 included calling the clinic or nutritionist for advice in their descriptions of what they would do in the event of a series of blood tests showing elevated phe levels. They were also less likely to describe enlisting aid from representatives of agencies outside the family, such as teachers and school nurses.

Mothers of children with poor adherence who did rely upon familial and social supports often (in 4 out of 7 cases) found these relationships laden with conflict. One mother's spouse criticized her handling of the diet "Like I'm a bad mother." Another mother feared that her mother-in-law, upon whom she relied for baby sitting, would report her to the child protective agency if she knew the child's blood results. A teen age mother's gratitude towards her child's extended family turned bitter under the impact of a multitude of life stressors.

Conflict often occurred in several systems at once, and economic, familial, and individual personality factors interacted to reduce available resources. The teen age mother found it "hard to get along" with the PKU staff, and was reluctant to call them when another agency failed to deliver formula, risking a temporary shortage of her child's basic food. Such combined effects directly and indirectly

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affected adherence, sometimes undermining motivation. Even when maternal motivation remained high and the general attitude towards the diet was positive, conflicts in support systems sometimes interacted with individual factors to affect adherence at least moderately.

> Mrs. H. firmly believes in the diet. Her daughter's phe levels are only slightly above the "clinical intervention" range, but Mrs. H. feels they could be better. She measures everything and often works overtime at her secretarial job to pay for the special PKU foods. P., age 10, spends after school hours with her grandparents. Mrs. H. worries that P. finds it easy to cheat on the diet on these visits, because extended family members neither understand nor actively support the diet.
>
> Mrs. H. does not recall seeking family support at the time of diagnosis. She drew upon her personal faith. A cheerful, generally effective person, she takes pride in her independence. Should her child have a series of high phe levels, she would try to deal with the problem herself, at least for a time, rather than calling the doctor or clinic right away. She has given up on enlisting support for the diet from the school. Use of the cafeteria is not possible, although this means more night time bread baking and morning lunch packing on a tight schedule. Mrs. H. discounts her daughter's sense that she is perceived as different by others because of the PKU, noting there are always lots of friends around. P. is taking more responsibility for the diet, but Mrs. H. concedes that her own vigilance, rather than her child's full cooperation, still largely accounts for P.'s dietary success. She connects the dietary "cheating" with opportunity, and does not address systems-support conflicts that her child may have internalized, and that may affect motive.

Negative or ambivalent attitude towards living with PKU. The majority of mothers in this group were less able to maintain a positive attitude towards the experience of living with PKU than were mothers of more adherent children. Two of the 7 did describe the diet as a challenge rather than as a burden, and one felt that it strengthened her

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relationship with her son. One mother with a high FLQ score whose child had relatively poor adherence, and for whom some role reversal was apparent, described the diet in terms of challenge for the child, stating that the experience of living with PKU could provide a direction for the child's life. The remaining mothers in this group described the diet ambivalently or in terms of burden: "Unpleasant." "I get disgusted." "A pain in the butt." "Frustrating."

These mothers often spoke lovingly and feelingly of their children, but none offered the spontaneous panegyric that often occurred in the good adherence group. Some felt emotionally exhausted by the demands of the diet. They were concerned about their children's elevated phe levels, but felt unable to change them. Frustration and unresolved grief could cause bitterness, passivity and a sense of fatalism. Some of these mothers stated that their feelings of sadness had not changed over the years, and in some cases the feelings had intensified. Some stated that nothing short of a cure could make life easier.

For the most part, then, mothers in this group were less able to sustain adaptive hope and positive attitudes towards the diet than were mothers in the good adherence group.

In summary, mothers of children with poor adherence express a weaker diet-related sense of coherence than mothers of children with good adherence. Their patterns differ, but each sees the family's experience as less

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coherent on at least one, and sometimes on all, of the dimensions of comprehensibility, manageability, and meaningfulness.

Relations among Family Practice of Conventionally Valued Routines, Diet-Related Routines and Dietary Adherence

It was expected that frequent practice of conventionally valued routines would provide a framework for the integration of diet-related routines, and that this relationship would be revealed in mothers' comments and interview responses. In fact, except for two cases in which the FRI score was extremely low, the interview data show little relation between the FRI measure and those qualities of diet routines that were associated with dietary adherence. Among those interviewed, mothers who reported more frequent practice of conventional routines not related to the diet did not necessarily practice diet-related routines associated with good adherence.

Comparison of Specific Diet-Related Routines Across Good and Poor Adherence Groups

Practice of certain specific diet routines and behaviors, reported in Table 8, discriminated between mothers of children with good and poor adherence.

Two kinds of diet-related routines were examined: every day routines such as maintenance of the food record, and problem-specific routines. For the every day routines, mothers were asked general questions and credited if they spontaneously mentioned specific diet-related behaviors.

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For the problem specific routines, mothers were asked what they would do if a series of blood tests showed high phe levels, and credited for spontaneously given specific responses. Mothers may well have practiced routines they did not mention; however, the differences in how they spontaneously described their behavior and their children's behavior are of interest.

Table 8

Number of Mothers in Good and Poor Dietary Adherence Groups Reporting Practice of Specific Behaviors Conducive to Adherence (Total N=14)

	Good	Poor
Routine Care	Total N=7	Total N=7
M schedules formula	7	7
M maintains diet record	7	2
M measures and counts	7	4
M adjusts daily intake	7	4
High Levels Care	Total N=5*	Total N=7
M reviews food intake	5	6
M contacts nutritionist	5	2
M confirms results	5	7

M=mother

*Questions about high levels care were not asked in interviews of 2 families

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Compared with mothers in the good adherence group, mothers in the poor adherence group were less likely to state spontaneously that they maintained a diet record, measured and counted food, and adjusted daily food intake. All mothers said they would be concerned over a series of high levels, and all described steps they would take to rectify the situation, but mothers in the poor adherence group were less likely to state that they would call a clinic staff person who could help with problems, such as the nutritionist. In response to another question, several of these same mothers expressed doubt that their efforts to lower phe levels would succeed, whereas all mothers in the good adherence group believed their efforts would succeed.

Qualities of Diet-Related Routines Associated with Good Adherence

Beyond the specific behaviors that discriminated between good and poor adherence groups, what mattered was the affective valence and degree of mutual engagement associated with whatever routines the family did practice, whether they were conventional routines, diet-specific routines, family celebrations, or highly idiosyncratic traditions ("We have silly supper, it can be all dessert--get a blanket and picnic on the living room floor. ")

Mothers of children with good adherence integrated the diet with daily life in a positive manner. They performed many diet-related routines in the presence of their

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children, integrated the diet with family and social activities, engaged their children in a range of age-appropriate diet-related activities, and imbued these experiences with positive feeling.

Involvement of children in diet routines. All interviewed mothers of adherent children calculated their children's phenylalanine intake; they planned and measured the day's food, maintained a daily record of some kind, and reviewed food intake with the children in order to make dinner time and day's end adjustments. Such routines provided for much diet-related interaction. For example, a 3 1/2 year old shopped with his mother, choosing "foods that are good for me" in the produce section. A 5 year old watched her mother measure the formula, turned on the blender, and counted out grapes and chips for lunch. A 10 1/2 year old measured her own formula and planned a lunch with her mother that would permit a bigger piece of corn on the cob for dinner that night.

Broad integration of the diet across social realms. Mothers of adherent children described a diet that was integrated with home, school, and extended family activities. Favorite recipes were often made for everyone, with a portion adapted to the child's needs. Four of the 7 mothers cooked with their children, or made specially adapted treats for them, often with an ethnic flavor: Polish meatless glunkies, little English puddings, special Easter eggs. A fifth mother who described herself as a "terrible

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cook" sent measured bags of popcorn to a school party, so her daughter could be the giver who ate what, and as much of what, the other children did. All mothers in this group talked more about what their children could have rather than what the could not, and the environment seemed richer in food. Teachers knew that ice pops were permitted on trips. Grandparents made dressing of special bread, baked outside the turkey. The diet was woven into the family's culture in the context of positive social interaction.

Positive tone of interactions involving food. Mothers of adherent children offered positive descriptions of the activities in which food was included. Their children dawdled over formula sometimes, but there were very few conflicts over food. When food was of social importance to the family, mothers of adherent children found ways to help them experience its various pleasures. For example, the daughter of an Italian mother enjoyed making not only the tomato sauce she could eat but the meatballs she could not, because "It's squshy and it's fun."

The following example shows ways that mothers of adherent children integrated diet routines with daily life in a positive manner:

> Mrs. N. describes her family as "traditional"; and herself as a "stay at home mom." She enjoys cooking with R., age 8. The mother melts the special duobar chocolate, and R. dips the pretzels. Of dinner times and family favorite recipes, Mrs. N. says, "There's nothing that we have that I can't find a way of giving it to R. in a certain way," with the recipe adjusted. They pack a snack box for school, so there will be permitted foods if an unplanned party comes up. For a birthday party at home with friends, Mrs. N. is likely

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> to go all out, and make something like a multi-colored sherbet watermelon with duobar chocolate seeds. R. is well aware of all the calculating and measuring of the diet. Mother and child inspect food labels for protein content when they go shopping. When the family eats out, they always take a food scale and the PKU "equivalents" book. There is large extended family, all of whom know about the diet, and summer time is a round of cookouts. At the last one, Mrs. N. listed all ingredients of dishes for everyone, "Not just for R., everybody's on a diet of some kind."

Qualities of Diet-Related Routines Associated with Poor Adherence

Compared with mothers in the good adherence group, mothers of children with poor adherence described low integration of the diet with daily life. They were less likely to perform diet routines involving precise accounting of food, and less likely to involve their children when they did so. Qualitatively, their descriptions of food-related activities were less rich and specific, and in some cases family activities involving food had a negative affective tone.

Low child engagement in shared diet routines. Whereas all mothers of children with good adherence kept precise account of food intake, often conferring with their children about the day's food, just 2 of the 7 mothers in the poor adherence interview group maintained such a record, and 4 in 7 measured precise food amounts, usually without engaging their children in a shared activity. Only one of the mothers in this group described cooking with her child.

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Low integration of diet in social activities. Several mothers in this group baked bread, but they rarely prepared special treats adapted for PKU except for birthdays. For some parents the expense of PKU ingredients inhibited such cooking, but one well to do mother said she rarely made such treats, though she hoped her child would do so. No mother in this group spontaneously offered an anecdote of a relative's preparation of a special PKU treat of the order of Easter eggs, a PKU cake, or specially prepared Thanksgiving dressing. In general, children in these families were less involved in daily interactions involving cooking, measuring, and discussing their food.

Negative tone of food-related activities. Although the mothers in this group described many activities enjoyed by their families that did not involve food, those activities in which food was involved were sometimes negatively toned. Family trips such as going out to dinner sometimes caused conflicts. Parents hid their own trips to the refrigerator for snacks to avoid intensifying the sense of deprivation that they believed the children suffered. One mother detained her child at the dinner table for as long as it took to finish the formula, and another had her child eat alone in order to avoid conflicts over food. For most of these children, the environment was not rich in food, or in the pleasure of interactions involving food. Negative experiences were sometimes related to financial difficulties and time constraints:

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> Mrs. W. says that she rarely measures foods exactly, but "I try not to give K. [age 6] rice and potato on the same day." Keeping a written record is not a daily event. When Mrs. W discusses food with K., it is usually to explain why she cannot have something. Mr. and Mrs. W. both like to cook, but work varying shifts, and juggling the sitter schedule takes precedence over elaborate food preparation. Pizza is a family favorite, but the special crust is expensive. "I run out of that, and the diet macaroni that she likes." Mrs. W. feels sad when she has to prevent K.'s little sister from offering K a piece of her own pizza. K. is "always hungry and she feels deprived...She'll say, 'It's not fair, Mom.'"

Other Findings

Social Skills among Children with PKU and Non-Affected Peers

Use of standardized measures permitted comparison of children in the study with their peers (i.e. children who do not have PKU) on measures of social skills and behavioral adjustment. On the Social Skills Rating System (SSRS), means and standard deviations for children with PKU (N=34) were closely comparable to those of the standardization group. The SSRS Social Skills and Behavior Problems scales each have a mean of 100 and a standard deviation of 15. For children with PKU, the Social Skills mean was 102.15 with a standard deviation of 15.93, and the Problem Behaviors mean was 97.79 with a standard deviation of 12.97. This means that, as a group and according to maternal report, children with PKU show a level of social skills and behavioral adjustment closely comparable to their peers.

School Competency

As a group, children with PKU in this study were found to be doing less well than the nonreferred group on the CBCL

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measure of school competency. The mean T score for the study children was 42.9, with a standard deviation of 9.28. This T score is two-thirds of a standard deviation below that of the standardization sample (Mean T=50, S.D.=10). This is a substantial difference. Review of parent responses for the PKU group confirmed that the children have many difficulties in school. For example, 32% of the children receive remedial services ranging from basic skills support to special class placement, and 18% have repeated at least one grade.

Summary of Findings

In summary, the findings show that:

Mothers' perceived family sense of coherence is significantly and positively correlated with mothers' report of the practice of every day family routines.

Mothers' perceived family sense of coherence is significantly and positively correlated with their report of the children's prosocial skills, and significantly and negatively correlated with their report of frequency of children's behavior problems. When a relatively even influence of social position is assumed, mothers' perceived family sense of coherence is significantly and negatively correlated with children's mean phenylalanine level, a measure of dietary adherence in which lower values indicate better adherence. When it is considered as a predictor of adherence together with other variables including social position, mothers' perceived family sense of coherence does

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not reach significance, but its effect size shows a trend in the predicted direction.

Patterns observed within the study sample suggest that, among those of middle and lower social position, low SES in conjunction with weak family sense of coherence constitutes a high risk factor for dietary adherence; and, that family sense of coherence of at least moderate strength is a prerequisite for good dietary adherence.

Mothers' report of the frequency with which the family practices every day conventionally valued routines is significantly and positively correlated with mothers' report of children's' prosocial skills, and significantly and negatively correlated with their report of frequency of children's problem behaviors, when it is considered as a single predictor. When considered with other variables, only the correlation with social skills retains influence, as a trend. With correlations at the level of chance, the evidence does not support an association between frequency of practice of conventionally valued routines and child dietary adherence.

Mothers spoke of their family experience of living with PKU in ways that were consistent with their measured perceived family sense of coherence, according to clinical interpretation of interview data. Good adherence was associated with mothers' belief in the diet as comprehensible, manageable, and meaningful. In the few instances when diet-related and general sense of coherence

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differed, diet-related sense of coherence was the better predictor of dietary adherence.

The practice of specific diet related routines, and the affective valence of such routines and activities distinguished between good and poor child dietary adherence, according to clinical interpretation of the interview data. Mothers of adherent children included the diet in daily life in a positive manner. They engaged their children in diet-related routines and activities, integrated the diet across social realms, and imbued these experiences with positive feeling.

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CHAPTER FIVE

DISCUSSION OF THE RESULTS

Discussion of Hypotheses

Hypothesis I: Relation between the Family Strengths

Hypothesis I stated: Family sense of coherence will correlate positively with family practice of regular routines. Hypothesis I was sustained. Mothers who perceived that, for the family, life and its challenges are comprehensible, manageable and meaningful were more likely than mothers who perceived family life as less coherent to report that their families regularly practiced conventionally valued interactive routines.

This relationship was explored, in part, in order to provide theoretical support for family sense of coherence as a construct. A part of the present study concerned the adaptation and development of a family version of Antonovsky's (1979, 1987) individual sense of coherence construct. One way to validate a construct is to compare it to another that is conceptually and functionally similar.

In his comparison of the individual sense of coherence construct with others, Antonovsky (1987) noted its similarity to the "sense of permanence", a construct described by Boyce and his colleagues as a deep belief in the continuity and predictability of human experience (Boyce et al., 1983; Jensen et al., 1983). These researchers argued

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that the sense of permanence is strengthened by the practice of normatively valued routines, and that the sense of permanence, indexed by the practice of such routines, predicts to better child development and health. This hypothesis was borne out in later studies (Keltner, 1990, 1992), using Jensen and Boyce's (1983) measure of the sense of permanence, the Family Routines Inventory. Thus, the two constructs, sense of coherence and sense of permanence, are considered to be both conceptually and functionally similar. In the present study, another similarity is the use of the family as referent in the respective measures of the sense of coherence and the sense of permanence, the Family Life Questionnaire and the Family Routines Inventory. The significant positive correlation between these two measures adds support to the validation of family sense of coherence construct.

A correlation of moderate strength, such as that obtained, was expected because, while the two constructs are related, the two measures address distinct realms of family experience. Family sense of coherence refers to an underlying representational/motivational orientation, while family routines refer to specific kinds of behavioral interactions. Another reason for the moderate strength could be that the sense of permanence, as described by Boyce and Jensen, is primarily concerned with the regularity of routines rather than with their affective and instrumental dimensions. In its concern with regularity and

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predictability, the sense of permanence is most closely connected to a single dimension of the sense of coherence, that of comprehensibility (Antonovsky, 1987).

Hypothesis II: Family Sense of Coherence and Child Resilience

Hypothesis II stated: Family sense of coherence will correlate with dimensions of child resilience including (a) social skills, (b) behavioral adjustment, (c) school competence, and (d) dietary adherence. Each of the sub hypotheses of Hypothesis II was sustained. When considered as a single measure, mothers' perceived family sense of coherence correlated significantly as predicted with mothers' report on measures of children's prosocial skills, freedom from behavior problems, and progress in school; and with the measure of children's' dietary adherence based on medical record of laboratory tests.

When family sense of coherence was considered along with other variables, such as social position, in regression analyses following hypothesis testing, these findings were qualified. Family sense of coherence retained significance in its associations with social skills and behavioral adjustment. The associations between family sense of coherence and the two other child variables, school competence and dietary adherence, were expressed as trends in the predicted direction.

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Family Sense of Coherence, Social Skills, and Behavioral Adjustment

Family sense of coherence may be thought to correspond, if loosely, to other family milieu constructs traditionally measured by such scales as the FACES-II and the FES. Thus, this study joins many others, such as those of children with diabetes, in which researchers have found that family cohesion, organization, and adaptability support the psychosocial function of children enduring the stress of chronic illness (Hanson et al., 1992; Hauser et al., 1985; Hauser et al., 1993; Wertlieb et al., 1986). The present study's finding that strong family sense of coherence is associated with fewer child behavior problems is consistent with findings about family function and behavioral adjustment in studies of families of children with PKU by Reber, Kazak, and Himmelberg (1987) and Shulman et al. (1991). When the focus is on absence of pathology (i.e. freedom from behavior problems) as a measure of adaptive function, the findings of the present study are similar to those of previous research.

The present study was concerned with resilience. Although it included measures of negative function, such as children's behavior problems, its focus was on measures of positive child functioning, such as prosocial skills. Here, the findings differed from those of the one study of children with PKU to consider this area, using similar methods of assessment in a similar population. In evaluating

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multiple aspects of family systems function through standardized parent report questionnaires, Reber, Kazak, and Himmelberg (1987) found no correlation between family function and children's social competence; and, in the same sample, Kazak, Reber, and Snitzer (1988) found the children to be less socially competent than a control group of children without PKU. In contrast, the present study found that family sense of coherence correlated significantly with social skills, and that, as a group, the children showed a normal range of social function.

A possible explanation of the first difference may be that the family sense of coherence construct, with its inter-related tripartite structure, is more inclusive than measures of constructs such as family cohesion and family organization. The qualities it taps may also be more closely related to the demands placed upon families of children with a disorder likely to affect social development.

One of the central challenges faced by these parents is that of ensuring that their children can move freely in the social world, participating fully in the many social interactions in which food plays a role. Free movement with full participation is a probable prerequisite for normal social development among school age children. The parent interview data attest to the sheer amount of work and dedication it takes to meet this challenge. In order to manage a diet that goes "into the world" with the child, families must build a shared knowledge base that includes

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child, teachers, friends, and friends' parents; they must use many resources and supports to integrate the diet with daily life; and they must commit themselves to the worth of this energy consuming goal. The sense of coherence components of comprehensibility, manageability, and meaningfulness relate, respectively, to each of these means of meeting the dual challenge of following the diet while promoting normal socialization. The triad of qualities measured by the sense of coherence construct corresponds closely with the qualities required of parents as agents in their children's social development, given the conditions imposed by the diet. True "goodness of fit" may account for the observed correlation.

Age differences of child subjects could account for the second difference between the finding of the present study and that of the Kazak group. In the present study, mothers reported a normal range of social skills function, whereas Kazak et al. (1988) found the children with PKU to be less socially competent than their nonaffected peers. All children in the Kazak group were 6 years old or younger, whereas children in the present study ranged up through 11 years 11 months. During the follow up parent interview, many parents in the school age group described how closely they had monitored their children's social contacts when they were young, limiting play dates, overseeing lunches with friends, prohibiting sleep overs, and supervising visits with relatives in order to prevent dietary lapses. Many

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mothers remarked that, although they were still vigilant, they were no longer restrictive of their children's social lives now that they could rely on their children's knowledge of the diet and upon their own skill in helping others to structure their children's "food environment." These children, now older and more socially active, may have overcome any social handicap resulting from their parents' earlier adaptive protectiveness.

That a long-term history of restrictive and controlling parenting style is associated with internalizing problems among patients with PKU has been shown in one study in Germany (Pietz et al., 1997), but patterns of over-control and behavior problems were not evident among the children in the present study. Although it is secondary to the hypotheses, the finding of psychosocial normality of the study sample is important, because of the questions that have been raised about the children's social competency, and because it places these children with PKU in the mainstream findings of pediatric psychologists regarding the psychosocial resilience of many children with chronic illness (LaGreca, 1990).

Family Sense of Coherence and School Competence

Family sense of coherence was the only predictor variable, including social control variables, to correlate significantly with children's school competence in bivariate correlations. Children from families in which mothers

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reported that the family sense of coherence was strong were also less likely to have repeated a grade, or to have required special educational services. The relation between sense of coherence and school competence was less robust than that between sense of coherence and other aspects of psychosocial resilience, however. It is likely that other factors play a significant role in accounting for the variation of the school performance of children with PKU. From the data, it is not clear what these factors might be. Neither mother's education nor family social position reached significance, either as single predictors or within the context of the regressions. Most surprisingly, review of data shows that child phenylalanine levels, a good predictor of IQ results among children with PKU (Brunner, Jordan, and Berry, 1983; Reber, Kazak, and Himmelberg, 1987), did not predict to school competence; correlations were at the level of chance. Clinical experience indicates that there is small set of children whose school performance is clearly adversely affected by elevated phe levels, but also a set of children on diet who perform relatively well despite moderately high phe levels. Genetic variations and social factors may account for these differences. According to the present study and that of Brunner et al. (1983), as a group, children with PKU do less well than their peers in school. It is, therefore, important that further research articulate sources of school success and failure among this population.

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Family Sense of Coherence and Dietary Adherence

When a relatively even influence of family social position was assumed, and when it was considered as a single predictor, mothers' perceived family sense of coherence correlated significantly as predicted with the children's dietary adherence. Mothers who viewed their social/familial world as comprehensible, manageable, and meaningful were more likely to have children whose phenylalanine levels indicated good dietary adherence than were mothers whose responses indicated weaker family sense of coherence. Even when family social position--itself considered by Antonovsky (1987) to be a factor of sense of coherence--was included in regression analyses, the effect of family sense of coherence retained influence as as a trend, although it did not reach significance. Within the context of small sample size, a trend with the observed effect size merits attention. The following remarks are consistent with patterns observed in the interview data for the sample, but are speculative with regard to the population.

Two related explanations may account for the observed correlation. The first invokes the "goodness of fit" rationale proposed with regard to family sense of coherence and social skills. The qualities that are needed to help children with PKU gain social skills are also, and perhaps especially, needed to help them to follow the diet. Family members must build a consistent communication base in which knowledge about the diet and the child's response to

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treatment is reliably shared among members and with appropriate individuals outside the family. In order to build an effective treatment alliance, family members must establish trust with the clinic staff while maintaining a sense of efficacy and participatory agency in a situation in which a good deal of control must be relinquished to others. In order to meet the long term, pervasive, and high demands of the treatment, family members must be able to form and commit to goals, and draw sustenance from the meaning with which they imbue the experience. The general family qualities of comprehensibility, manageability, and meaningfulness that underlie these specific abilities are those that the family sense of coherence construct, and the Family Life Questionnaire (FLQ), attempts to measure.

The interview data suggest that, given a more accurate measure of the construct, the correlation might have been slightly stronger. Like the original, individual Sense of Coherence scale (Antonovsky, 1987), the Family Life Questionnaire may mis-classify those few subjects whose extremely high scores indicate not strong sense of coherence, but what Antonovsky called "false" or "rigid" sense of coherence (1979, 1987). This appeared to be the case with one mother of a poorly adherent child. This mother with the highest FLQ score in the study professed high commitment and claimed much family support for the diet, but her specific responses indicated role reversal and emotional distancing at odds with such avowal.

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A second, related explanation for the correlation observed in the sample between perceived family sense of coherence and children's dietary adherence concerns the concept of morale. The burden of care giving for children of PKU falls largely on mothers, and what was actually measured was mothers' perception of the degree of to which the family experiences life as comprehensible, manageable, and meaningful. Mothers who believe that relations within the family, and between the family and the social surround, are strongly coherent may feel--and be--profoundly supported as they engage in a variety of difficult endeavors, including that of overseeing this demanding diet. This, in turn, helps them to maintain a positive attitude. An example is the mother who cited multiple supports and whose extended family accompanied the couple and their newborn to the PKU clinic. She expressed a personal philosophy of commitment within this supportive world, and experienced low stress in helping her child to follow the diet. The effects of such familial support upon effective coping involving care of chronically ill children are well documented.

Hypothesis III: Family Routines and Child Resilience

Hypothesis III stated: Family practice of regular routines will correlate positively with dimensions of child resilience including (a) social skills, (b) behavioral adjustment, (c) school competence, and (d) dietary adherence.

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Hypothesis IIIa and IIIb were sustained. When considered as a single measure, mothers' report of practice of normatively valued family routines correlated significantly as predicted with mothers' report on the measures of children's prosocial skills and freedom from behavior problems.

Hypothesis IIIc received some support. There was a trend in the predicted direction between mothers' report of family routines and the measure of children's school competence.

Hypothesis IIId was not sustained. The correlation obtained between mothers' report of practice of family routines and the measure of children's dietary adherence was at the level of chance.

When family practice of routines was considered along with other variables, such as social position and family sense of coherence, in regression analyses following hypothesis testing, these findings were qualified. Family practice of routines retained influence, as a trend, only in its association with child social skills.

Family Routines and Psychosocial Resilience

According to maternal report, children in families that often engaged in shared, normatively valued routines measured by the Family Routines Inventory were more likely to be socially skilled, and less likely to have behavior problems, than were those in families that engaged in such routines less frequently. The correlation between family

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routines and social skills was relatively robust; it was the only one involving family routines to retain influence as a trend when considered with other variables in the context of regression analyses. This suggests that the patterned interactions of daily family life--bed time rituals, shared meals, family chores, customary ways of marking departures and arrivals and the like--may indeed promote stability and a sense of permanence, providing a matrix in which children acquire, practice, and develop such basic social skills as cooperation, assertion, responsibility, and self control.

Family Routines and Dietary Adherence

Contrary to expectation, the measure of the family's regular practice of family routines was not correlated with the measure of dietary adherence. This aspect of Hypothesis III was not sustained. Thus, although the matrix provided by conventionally valued family routines may foster children's general social skills, the frequent practice of such routines does not, in and of itself, foster better adherence. There may be a threshold below which the frequency of conventional routines provides insufficient framework for the integration of diet-related routines. Two interviewed mothers with exceptionally low FRI frequency scores also practiced few diet routines and integrated them poorly with daily life; their children were in very poor dietary control. The data do not, however, support the hypothesis that more frequent practice of conventional

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routines that are not related to the diet predicts better dietary adherence. Something more is needed.

The interview data suggest that families must be motivated by their positive attitude towards the diet in order to use the framework provided by daily, non-diet related routines. This framework includes not only the conventional routines measured by the FRI, but also traditions and celebrations that are culture specific or unique to the family. When families are so motivated, members imbue the diet routines with positive affect, and they weave these routines into every day life in a positive context. A general routines framework of some kind is essential, and practice of diet routines is essential, but dietary adherence is best when the two are integrated. A likely consequence of such integration is the engagement of the child, facilitating transmission of family attitudes towards the diet. These qualitative aspects of family routines, diet-related, conventional and otherwise, go beyond mere frequency of their practice, and relate most closely to the meaningfulness dimension of family sense of coherence.

Discussion of Related Findings

Family Social Position, Family Sense of Coherence, and Dietary Adherence

Following hypothesis testing, regression analyses were conducted to determine the relative strength among the

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family predictor variables with regard to child outcomes. When social position (SES, measured by Hollingshead's Two Factor Index) was considered along with sense of coherence and other variables, social position proved to be the more powerful predictor of the measure of dietary adherence, with family sense of coherence retaining influence as a trend. Even through interpretation of the regression analyses, however, it is difficult to "tease out" the relative strength of these two variables, because in salutogenesis theory they are closely intertwined. In Antonovsky's terms, wealth helps to provide the kinds of consistent experiences that lead to formation of a strong sense of coherence and that help to maintain it. Wealth and social position are also among the major generalized resistance resources available for use in avoiding and combating stress (Antonovsky, 1979, 1987). Except under extreme conditions, however, family sense of coherence and family social position overlap but are not coextensive; sense of coherence has many other sources, and there are many other generalized and specific resistance resources that facilitate coping. The relations among these variables in the present sample are complex, and merit further exploration.

The Role of Family Social Position (SES)

Considered both as a single variable, and along with other variables, family social position had a significant effect as a predictor of the dietary adherence variable. While it is a given in epidemiology that wealth secures

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better treatment for most illness, family social position might be expected to play less of a role among patients with PKU than among patients with some other chronic conditions. Many of the treatment costs are covered either directly by the state, or by legislatively mandated reimbursement of costs by insurance. Indeed, in one study of families of children with PKU from an area demographically similar to that of the present study, phenylalanine control did not correlate significantly with family income or parents' level of education (Reber et al., 1987). In the present sample, all families are entitled to the formula, the children's chief food, either free of charge from the state, or through reimbursement by mandated insurance. Formula is delivered to the homes of those receiving Medicaid. Parents themselves may do their children's blood work at home and mail it in clinic-supplied canisters to the state laboratories, where it is tested without charge. Dietary counseling is done mainly by telephone, again without charge. Clinics typically "write off" charges not covered by insurance for clinic visits. Thus, many supports to families facilitate the clinic's potential as a resource that may be reliably available to patients, at low expense.

There are, nevertheless, potential financially influenced barriers to families' use of the clinic as a resource. In the present sample, while no subject provided information meriting membership in Hollingshead social class V, the lowest social position, some in class IV were

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temporarily unemployed, had lost insurance, were at times without telephone service, or lacked transportation for formula pick up or such clinic visits as were required. One family was homeless for a time. Failure to gain weight was implicated in the high phenylalanine levels for one child from a family in class IV. Blood work was more frequently postponed (because results would have been less informative) for children in Class IV because of reported illness such as flu, colds, or asthma that affected formula consumption. Coupled with the many other life stressors associated with poverty, these treatment related stressors may certainly have had some direct effect upon phenylalanine levels, as well as both direct and indirect effects upon communication between families and the clinic, and upon dietary adherence.

The Role of Family Sense of Coherence in Relation to Social Position

A description of the sample, and inferences from interview and clinical data, permitted identification of families most likely to be at risk for poor adherence, and of the conditions under which family sense of coherence is most likely to affect treatment outcome.

In the sample, doubly advantaged families--those with strong perceived sense of coherence and high social position--were highly likely to have children in good dietary control. Conversely, doubly disadvantaged families--those with weak perceived sense of coherence and low social

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position--were highly likely to have children in poor dietary control. These are families at highest risk.

Among relatively well-to-do families, the anecdotal evidence indicated that high social position (membership in class I or II) could over ride weak family sense of coherence as a predictor of dietary adherence, but only if the mother herself demonstrated high personal commitment to the diet. For example, a mother in Class II (above middle class) had a low (<170) FLQ score, confirmed by her expressed feelings of isolation and reportedly low level of familial support. Nevertheless, despite feeling stressed by the work the diet entailed, she was strongly dedicated to the diet, and her child was in good adherence for the duration of the study. The child's adherence has since been erratic, however, which calls into question the long-term stability of high SES as a buffer against the stress of weak family sense of coherence. Anecdotal evidence suggested that wealth provided no buffer against weak family sense of coherence without strong maternal commitment. In another social class II family, the mother with a very low FLQ score expressed a laissez-faire attitude towards the diet. ("You take it or leave it, just do the best you can.") Her child's adherence was among the poorest of those in the study.

In the sample of families from the middle and lower classes (III and IV), none of the 8 families with a measured family sense of coherence FLQ score below 180 had a child with phenylalanine level indicating good dietary adherence,

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regardless of class membership. In contrast, 60% of the remaining 15 families in Class III and IV with FLQ scores >180 had a child with good phenylalanine control. This suggests that, for those who do not enjoy the benefits of wealth, a minimum belief in the coherence of familial and social world (in this case represented by an FLQ score that is no further than 1/3 standard deviation below the group mean) may be a necessary though not sufficient condition for good treatment outcome.

The model emerging from these data and inferences indicates that, for the less wealthy families (middle and lower classes), the primary caregiver's having a strong belief in the family's ability to make cognitive and emotional sense out of the world and to manage its challenges may indeed be one of the things that, as Rutter (1979) said, "goes right" in the promotion of desirable or resilient outcomes. The sample figures suggest that, in these less wealthy classes, the benefit from perceived family sense of coherence proceeds along class lines. That is, among the set of families with a sense of coherence that is "strong enough" (FLQ >180) to "permit" good adherence, a greater proportion (75%) of those families in Class III are helped by its presence than the proportion (42%) of those in Class IV that are helped by its presence.

Accounting for the negative case, that is, for those families in Class III and IV in which strong family sense of coherence does <u>not</u> predict better dietary adherence, is

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beyond the scope of this study, but some speculations may be based on the qualitative data. Mis-classification may account for a few cases. For example, interview responses suggested that the moderately high FLQ score of a teen age mother in Class IV reflected her distanced assessment of the functioning of a family from which she herself felt estranged. Barring such mis-classifications, the question remains, what are the particular, and differing, aspects of social position that make it a risk factor for some families but not others among those who have strong sense of coherence? Among those Class III and IV subjects in the poor adherence group who showed genuinely strong family sense of coherence, which deficits in resistance resources specific to the diet may have prevented its expression?

Interview data implicated money, time, and breakdown in client-clinic communication. A family might manage tight funds well, draw emotional support from church and relatives, care about the diet, but have no insurance coverage for the supplementary foods. A single mother might have little time to cook such foods, regardless of her dedication to the diet and confident outlook on life. Several Class III and IV mothers among those interviewed, including some with strong measured family sense of coherence, cited such constraints. The supplementary foods are not essential to the diet, but they make it much easier, and more enjoyable, to follow. Regular clinic-client communication is essential to diet management. Clinical

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experience shows that loss of telephone service, language difference, breakdown of trust, failure in clinic outreach, misunderstanding of cultural differences, family pride preventing acceptance of agency-provided help all have a disproportionate effect on diet management and adherence, even among those families who may feel strong sense of coherence with regard to many other aspects of their lives. Given a working assumption that strong family coherence will support dietary adherence unless it is prevented from doing so, it is clinically important for further research to identify and such address barriers, in order to better engage the family's strengths.

Family Sense of Coherence, Diet-Related Sense of Coherence, and Dietary Adherence

Mothers spoke about their family experience with the diet in ways that usually reflected their measured family sense of coherence, and that predicted to dietary adherence.

Among mothers who were interviewed, there was "goodness of fit" between their perceived general family sense of coherence as measured by the FLQ and their diet-related conversation in 12 out of 14 cases. In the two cases in which there was a difference, clinical assessment called the original FLQ scores into some question. Even including these two cases, however, there was high consonance between general family sense of coherence and diet-related sense of coherence. Mothers with higher FLQ scores described the diet

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in ways that showed they experienced it as comprehensible, manageable, and meaningful.

Consonance between a mothers' perceived family sense of coherence and her own expressed sense of coherence with regard to the diet as revealed in her speech is consistent with constructivist representational models of internalization. In the terms of this thesis, Antonovsky notes that if a collectivity such as a family can have (or be perceived to have) a common way of seeing the world, this then becomes an independent variable shaping the sense of coherence of individuals within that collectivity (Antonovsky, 1987). The individual's internalized global dispositional orientation towards the social world then further shapes and influences particular attitudes and responses to particular problems. Reiss (1981) also holds that the individual's representations and behavior partake of, and contribute to, the family's construction of the social world, and that larger representations shape individual responses, in this case those that involve coping with the particular stressor of managing the child's diet.

Sense of coherence is a global and relatively stable orientation. The primary strength and direction of influence is assumed to be from global orientation to particular behavior, but the influence is also bi-directional. Successful coping with sets of particular problems may gradually enhance global sense of coherence. As articulated by Antonovsky (1979, 1987), salutogenesis refers not only

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to the movement towards health facilitated by the sense of coherence, but also to the potential incremental changes in the sense of coherence itself. At any point in this process, close consonance between perceived family sense of coherence, diet-related sense of coherence, and dietary adherence is what would be expected. The observed consonance is therefore consistent the salutogenesis model.

Comparison of two cases with disparate (difference of 1 1/3 S.D.) perceived family sense of coherence scores illustrates this consonance as it is manifest in all three dimensions of the sense of coherence construct. The comparison also provides a case illustration of the statistical finding that, when a relatively even influence of social position is assumed, family sense of coherence correlates significantly with dietary adherence.

The families of both mothers are in Social Class III. The first mother has an FLQ score >185, or perceived family sense of coherence in the modal "higher" group of those interviewed. Her child is in the good (M phe <10 mg/dL) range of dietary adherence. Her expressed attitudes show diet-related sense of coherence consonant with her general sense of coherence. On the diet-related comprehensibility dimension, she and her husband see the diet as predictable; they can find a reason for high and low blood levels. On the manageability dimension, there are reliable resources available and problems are solvable. On the meaningfulness dimension, a struggle is contexted within an ascribed

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meaning ("it made us stronger..."), a goal is stated, and there is speculation about what might be getting in the way of its achievement:

> Mother A
>
> (Solving a high blood level mystery) "We were doing everything right. We kept saying, why is it high? Why, why, why? And then we said, Hm, that turkey on G.'s (the sister's) plate...It was a little sneaky thing, you know? ... But H. (the child with PKU) really has been very good. And you can see it in the blood levels, too."
>
> (Help from relatives as resources) "My mother is wonderful. I could say, O.K., this much equivalents, she knows...she likes to make little things for H. ...that's a grandmother thing..."
>
> (The doctor as a resource) "I mean listening to the doctor and doing what they say, you know you're going to feel better, and if you don't there's a reason for it."
>
> (On struggling with formula during the child's infancy): "When we were trying to get [the baby] to drink the [formula], I'd try, then he (her husband) would try, then if H. still wouldn't do it, then it's my turn ... actually, it probably made us stronger."
>
> (On goals) "Drinking the milk (formula) I want to be better. I don't know if it's, 'Mom wants me to and I'm not going to...'"

The second mother is also from a family in Social Class III. Perceived family sense of coherence is in the modal "lower" group of those interviewed (FLQ in <170 range). Phenylalanine levels are in the poor range (M phe >10). On the comprehensibility dimension, the mother describes the information she receives about blood levels as inconsistent and confusing. On the manageability dimension, daily dietary management is experienced as difficult, and a frequently encountered problem, coping with the diet on a trip, is unsolved. On the meaningfulness dimension, she expresses

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uncertainty as to the level of commitment to a stated goal, and includes no means for its achievement.

> Mother B
>
> (Describing waiting for blood results) "Actually sometimes when I think it's going to be good it comes back with a 16 or 18, and the last time, I don't know why, it was a 10 ...I feel frustrated. ... and L. (her son with PKU) says, 'It doesn't make any sense.' And I don't know how to answer because I'm puzzled myself."
>
> (Reacting to suggestions for managing a dietary problem) If you don't live it day to day you don't know how hard [it is]. ... I'll try [the suggestions] but I know ... that there's always going to be something, it's still going to be hard, it's not ever going to be easy."
>
> (Describing a trip) "If we stop and there's nothing [that L likes that he can eat, and we get something] he says, 'I'm starving.' It hurts."
>
> (Asked about goals for the future) "I hope the levels start to change." (Asked how this might come about and how committed she is to this goal) "I don't know."

For the first family, the environment and the experience of living with PKU are sources of information and meaning; for the second, the environment and the experience are sources of confusion and frustration. Both families work very hard at the diet, but for the second family, all aspects of care are effortful. This mother reports high stress.

Mothers' report of stress was frequent among those in the poor adherence group. Often, mothers spoke of stress in connection with conflicts with those upon whom they had to rely for help. When several systems were involved--insurance companies, the clinic, a mother in law--the

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potential for both conflict and confusion increased. Ongoing, inescapable conflicts with unreliable resources could certainly, eventually, erode both individual and family sense of coherence. For those with a sense of coherence already towards the low end of the continuum, any added stressors would have disproportionate effect.

According to the systems salutogenic model here proposed (Antonovsky, 1979, 1987; Fiese and Sameroff, 1992), when perceived sense of coherence is towards the low end of the continuum, it is harder for family members to bring order and meaning in to the system when they must cope with a stressor, such as the child's illness, even when reliable resources are seemingly available. Tension therefore cannot be resolved, and so causes stress. Stress further inhibits function, and affects parent-child interactions. In the process of these interactions, conflicts are internalized by the child. For the child, the outcome is likely to be an increase in stress, and antipathy towards the diet. A frequent reaction to this is cheating on the diet, often by eating excessive amounts of otherwise permitted foods, thereby elevating phe levels. These processes were observed in the interview data, and have been observed repeatedly in clinical experience.

General Family Routines, Diet-Related Routines and Activities, and Dietary Adherence

In this study, the frequency with which families practiced conventional routines not related to the diet did

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not correlate with adherence. Practice of certain diet-related routines, the engagement of children in their practice, the integration of these routines in daily life and culture, and the affective quality of diet-related interactions, however, were all strongly associated with good dietary adherence, according to interpretation of qualitative data. The PKU interviews were rich in examples of such behaviors. Reber et al. (1987) have suggested that experience near measures may be better than general scales in the prediction of outcomes among chronically ill children, and the PKU Parent Interview is an experience near measure.

Addressing specific behaviors is not meant to imply a simple behavioral model. Any present diet-related routine is contexted, and has meaning. Depending on many contextual factors, a child watching his mother measure his food may infer that she cares enough to do so, that she believes it is important, and will make a difference, that it is one of her ways of caring for him. The behavior is motivated and representational.

The intent of the open ended questions, even those designed to elicit descriptions of particular behaviors, was to tap the representation, to learn whether some routines were so much a way of life that they were naturally and spontaneously expressed with little prompting. That some mothers did not mention routines did not mean that they did not practice them, but that, perhaps, they were less a part

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of life, less fully internalized as part of a guiding care-giving orientation.

Practice of Diet-Related Routines and Behaviors

Two kinds of diet related behaviors, one set from the routine care (recording, measuring, and adjusting food intake), and one behavior from the high levels care (calling for clinic consultation), discriminated between mothers' inclusion in the good or poor adherence groups, according to the qualitative interpretation of the interviews.

Mothers in the good and poor adherence groups did not differ as to whether they spoke about routines they might perform alone, such as making formula at a regular time. All mothers mentioned this. Rather, they differed as to whether they spoke about diet related routines likely to be practiced throughout the day. Mothers in the good adherence group were more likely to describe routines such as maintaining a food record, measuring food, and adjusting food intake based on what the child ate during the day. Like formula preparation, such routines are essential to the diet, but what distinguishes them as a group is that, especially for the school age child, they are potentially highly interactive, and likely to involve ongoing dialogue and negotiation between parent and child. They are also likely to involve the web of relations among parent and child, child and family, school and child, school and parent. In order for the mother to perform them she must usually involve the child and others in dialogue ("Did you

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eat all the grapes at lunch?" "Is Jessica running low on the frozen cup cakes for the school parties?" "What would you like to bring along in case there's no fries at the fair?") Mothers of children in good adherence were not only likely to mention such routines of measuring and adjusting, but to give detailed accounts, often including dialogue: "I'll say, 'We're having corn on the cob,' Like if I can plan [dinner] ahead, I'll say, 'Make sure you don't have that much at lunch so you can have a bigger piece of corn on the cob.'"

These kinds of repeated enacted exchanges involve the child in active participation as well as compliance, embodying the participatory agency that makes experience meaningful, the shared information that makes it comprehensible, and the behavioral steps to what is often a mutually or reciprocally desired outcome, making experience manageable. In short, these are the kinds of exchanges through which the child both affects the system and internalizes the mother's and family's sense of coherence, and the family's attitudes towards the diet and towards the child herself. The affective quality of these exchanges is a crucial aspect of the transactional systems model(Fiese and Sameroff, 1992). This model is compatible with, and complements, the salutogenic model. If the transactions between parent and child are negative, according to Fiese and Sameroff's theory, a negative representation of self, other, and the relationship, including the negative

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connotations associated with the child's chronic condition, results.

The set of diet behaviors that distinguish between the good and poor adherence groups requires planning, projecting, and reminiscing about self, other, and the food between, over time. Thus these simple behaviors have representational and transactional implications for the dyad, and for the outcome variable that is most truly a systems variable, dietary adherence.

A second behavior that distinguished mothers of children with good adherence from those with poor adherence was their reference to maintaining ongoing contact with the clinic staff, usually the nutritionist, in the case of a series of high blood levels, rather than continuing to try to handle the problem without clinic assistance, or simply waiting until the next clinic visit. Keeping in close contact is very important in such an event, in order to share information required for making any necessary dietary adjustments. All mothers of children in good control to whom this problem was posed mentioned initiating or participating in telephone contact with the clinic, whereas just two of the seven with children in poor control did so. Of the five who made no mention of telephone contact, four had low FLQ scores. Three stated that they were uncertain that the steps they took to rectify the situation would be effective, yet they did not mention a step that would

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utilize an available resource and bring maximal information into the system.

Antonovsky's salutogenesis model would predict this pattern. When there is a dynamic orientation within the collective towards seeking order, a motivating belief in the essential coherency of experience likely to be provided by existing resources, individuals within the group are likely to partake of that orientation and likewise to seek out available order. In Werner and Smith's words, some of these parents may not reach out for support because they do not "expect well" (Werner and Smith, 1982). They do not "expect well" because the individual and the family as a whole has experienced little of the world's coherency and have, in consequence, learned there is little to expect (Antonovsky, 1987).

The pattern of being unable to rely on others, even when help is available and when it would be adaptive to do so, is also consistent with attachment theory. Belief in the reliable availability of the other, and in the self as a competent actor and a deserving recipient of support, is a tenet of this theory. The construct of "healthy self-other reliance" is, according to Bowlby, a concomitant of secure attachment (Bowlby, 1980), and reflects a flexible balance between reliance on self and others as the situation demands. The inability to seek needed support suggests disturbance in the attachment system. Attachment theory is quite consistent with Antonovsky's salutogenesis model, and

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he explicitly includes it as part of the foundation for the formation of the sense of coherence, which develops in the context of parent-child interactions and child rearing practices (Antonovsky, 1987). He emphasizes, however, that these interactions are always contexted in a socio-cultural and historical surround.

The interview data suggest that differences in self-other reliance may indeed influence some mothers' behavior in coping with the serious problem of prolonged high blood levels. One mother who had earlier spoken of how hard it is for her to trust stated, "I don't talk to the nutritionist...it's like a mental block. ...I never call unless it's a dire emergency, like M's (her child) hospitalized, God forbid." In contrast, a mother who stated that she would freely call for needed assistance had earlier stated, "I realized growing up, too, that so many people want to help if they are close to you or if they want to."

Affective Quality of Routines Including the Diet

It is not surprising that mothers whose children complied with the diet reported, in general, more pleasure in their everyday interactions, such as trips and dinner time, than did mothers of children who were noncompliant. It is not easy to enjoy eating with a resistant child. Yet the overall positive attitude towards the diet probably was a main source of influence, not only on the specific interactions themselves, but on how a mother framed the nature of the interaction. For example, one mother in the

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good adherence group, with a measured family sense of coherence towards the strong end of the continuum, was able to define her child's dinner time resistance as "picky eating" not related to the PKU. She could then decide "not to be so hard on myself" and found a way to adjust the formula so dinner was not affected, and became enjoyable. This instance of the breaking of a transactional cycle that could otherwise have escalated into true dietary noncompliance illustrates how the transactional model may operate within a salutogenesis framework.

Integration of the Diet with Daily Life

The integration of the diet across different realms of daily life--in the family, at school, in extended family contexts--was clearly one of the things that "went right" for a subset of children in the good adherence group. In general, these mothers' descriptions of living with PKU were positive. It is speculated that the mother's liking to make something the child liked, and sending it out into the world with the child, or encouraging its appearance in another form, for the environment to give and the child to find, is a powerful predictor of good treatment outcome. There are grounds for interpreting this in terms of Winnicottian object relations theory; the food may be thought of as a form of transitional object, a gift from the culture that is funded with interpersonal meaning. That food and food activities with the child represented a kind of gift from the culture to the mother, or mother's positive connection

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to her ethnic identity, was clear from some of the interview statements. For example, it was an Italian mother whose child enjoyed making "squshy" meatballs she could not eat, and stirring the tomato sauce that she could.

As a source of whole sets of potentially coherent experiences, a positive ethnic identity may be one of the major sources contributing to a strong sense of coherence. The integration of homely cooking and eating routines including the diet is a way of transmitting this sense of coherence, weaving the diet into the culture, the culture into the diet, and bringing the child into the family. The Italian mother (her spontaneous addition near the interview's close): "She has beautiful hair. So I think maybe that's the PKU, but I don't know, M's mother has exactly the same hair color so maybe her hair would have been that. All I know is she is what she is because of what she is, and however she is I wouldn't want her any other way....we're our family with her."

Implications for Clinical Practice

The finding that social position was an important predictor of treatment outcome among these families, even when many costs of treatment are covered, highlights the continuing need for outreach and support. Use of local supports is especially important, because families of children with rare disorders often live far from one another and far from the clinic.

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A problem, however, is that those families whose function reflects weak sense of coherence are also those who have difficulty using resources, even when they are available. Addressing a family's "global, motivational orientation to life," even when it can be reliably identified, may well be beyond the scope of what can be done in the context of outpatient clinical care. Coherency can, however, be a clinic goal for all transactions with families. The doctor, or the health care clinician, Antonovsky notes, is a potential generalized resistance resource (1979). It is simply good (or salutogenic) practice to engage the family's strengths, including the degree of strength of family sense of coherence, which may be suppressed by environmental barriers or breakdown of coherence in other systems.

To the extent that the shared health care encounter increases understanding, makes a specific problem more manageable, and meaningfully engages the family as participant, the encounter is a coherent experience. This research highlights the need for such experience.

This research also highlights the importance of supporting the integration of the diet with the family's preferred routines, especially those centered on family sense of identity. Childhood chronic illness connects family and clinic strongly, engaging many levels of structure and feeling. Childhood chronic illness involving regulation of something so basic as food, a necessity that

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is deeply and daily encultured, and close to the heart of any theory of human psychology, may forge a particularly close connection. Parents of children with PKU rely on the clinic continually for instruction in the feeding of their children. Many transactions occur. Many coherent transactions may help to maintain and reinforce family sense of coherence, with improvement in treatment outcome for the child.

Limitations of the Study

Findings of this study should be interpreted with the following limitations of the study in mind:

1. Sample size was small. This was a function of the rarity of this chronic condition. The small sample size was a handicap in the multivariate analyses that were conducted, and prevented further statistical investigation of some interesting relations among the measures of family sense coherence, family social position, and dietary adherence.

2. Maternal completion of questionnaires was used for several measures. Mothers' tendency to respond with a directional bias across measures may, therefore, have inflated some correlations; however, the consistency of the results of these correlations with findings not directly based on maternal report, such as direction of findings regarding dietary adherence and interpretation of interview data, argues against such directional bias.

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3. The sample included a relatively high proportion of families in Hollingshead Class IV, and some of these families experienced periods of marked financial stress during the study. This should be taken into account in generalizing findings, especially those involving effect of social position, to demographically different populations.

Suggestions for Further Research

The family sense of coherence construct shows promise as a correlate and predictor of psychosocial resilience and behavioral treatment adherence among chronically ill children. Further research could include:

1. Expansion of the study to other PKU sites, in order to confirm findings and to explore relations among family sense of coherence, family social position, and dietary adherence in a larger normative sample.

2. Refinement and development of the sense of coherence measure, including (a) further traditional piloting of the FLQ (Family Life Questionnaire) in other populations, with subsequent adjustment and revision of items; and (b) exploration of other means of measurement of the family sense of coherence construct, such as obtaining responses from all family members, developing an interview form of the FLQ, or observing and scoring family members' interactions in response to construct-based vignettes; and (c) development of objective scoring techniques for the construct based semi-structured PKU Parent Interview.

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3. Testing of the salutogenesis hypothesis among families of children with chronic illness other than PKU, such as children with juvenile diabetes.

Summary and Conclusions

This study explored a family systems version of Antonovsky's (1979, 1987) "salutogenesis hypothesis" in a sample of 34 families with pre-school and school age children with the chronic metabolic disorder phenylketonuria (PKU). It proposed that children's psychosocial resilience and treatment adherence are facilitated when family members have a strong belief that their shared experience is coherent; that is, when they believe that, for the family, the social and familial world is comprehensible, manageable, and meaningful. It further proposed that, because family attitudes are transmitted to children through interactions, family routines that reflect the family's sense of coherence are also related to resilient child outcomes.

Results generally supported the salutogenesis hypothesis. As a single predictor, mothers' perceived family sense of coherence correlated significantly as predicted with measures of child psychosocial resilience and dietary adherence. Considered along with family social position, a strong predictor of adherence, family sense of coherence retained significant association with measures of psychosocial resilience, and showed a trend in the predicted direction in its association with dietary adherence. The second measure of family function, family practice of

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interactive routines not specifically related to the diet, correlated significantly with measures of psychosocial resilience, but did not correlate with dietary adherence.

Interpretation of interviews with mothers suggested that sense of coherence is most closely associated with good adherence when it is expressed in terms related to the family's experience of living with PKU. Similarly, family routines that reflect the valuing of the diet are closely related to adherence.

Mothers spoke about their family experience with the diet in ways that reflected their measured family sense of coherence, and that predicted to dietary adherence. Compared with mothers of children with poor adherence, mothers of children with good adherence shared a consistent set of attitudes and behaviors. Mothers of children with good adherence believed that the experience of living with PKU was comprehensible, manageable, and meaningful; they believed in the diet, made effective use of resources, and maintained a positive attitude towards the experience of living with PKU. Their practice of diet related routines reflected commitment and positive affect consistent with the meaningfulness dimension of the sense of coherence; they engaged their children in shared and enjoyable diet-related activities and integrated the diet with daily life across many social realms.

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These findings are consistent with the salutogenesis hypothesis, which holds that strong sense of coherence permits the effective management of stress and promotion of healthful outcomes; and with attachment theory, which holds that the healthy self-other reliance of secure individuals permits effective use of resources. Extrapolated as a process, results are consistent with a transactional systems version of the salutogenesis model, in which family members, through participation in shared interactions, internalize group values and enact these values in the terms of a particular situation, in this case, supporting the child with the diet. Through further parent-child interactions, these positive diet-related values and behaviors are actively internalized by the child, resulting in psychosocial resilience and good treatment outcomes.

Based upon the study results, and considering the study limitations, the following conclusions are warranted for the population of families of children with PKU:

1. Mothers' perception that family life experience is coherent is positively associated with the psychosocial resilience of children with PKU.

2. Family social position is a factor affecting dietary adherence of children with PKU, with children in families with low socio-economic status at risk for poor adherence and elevated phenylalanine levels.

The following conclusions are warranted for the study sample:

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1. Mothers' perception that family life experience is coherent is positively associated with children's dietary adherence.

2. Mothers' attitudes and behaviors that promote dietary adherence and that relate to general sense of coherence include (a) positive attitude towards the diet, (b) integration of the diet with daily life, and (c) engagement of the child in shared, positive diet-related activities.

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APPENDICES

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APPENDIX A

THE FAMILY LIFE QUESTIONNAIRE

Development

The Family Life Questionnaire (FLQ) is designed to measure the construct family sense of coherence, or the extent to which the responding individual family members have internalized a global, "dynamic feeling of confidence" (Antonovsky, 1979, p. 10) that for the family and its members the social world within the family and between the family and the larger social world is comprehensible, manageable, and meaningful, and that "there is a high probability that things will work out as well as can reasonably be expected." (Antonovsky, 1979 p. 10, 1987)

The scale has 36 items, of which 21 refer to the respondent's assumptions about "within family" relations and 15 to "between family and social environment" relations. In each of these two categories, items addressing the comprehensibility, manageability, and meaningfulness dimensions are included. At this time, there is some emphasis on manageability items. Items are on a Likert type scale, from 1 to 7; higher scores represent higher family sense of coherence. A single summary score is used. Scoring for approximately half the items is reversed.

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The scale is conceived as being unifactorial, following Antonovsky's design. It is based in part upon his sense of coherence scale for individuals, in which, while each item primarily addresses one of the central components (comprehensibility, manageability, meaningfulness) in a statement that places the respondent in a life situation, each item stem and response also contains a set of systematically varied "facets" such as time reference (past, present, future), source of stimulus (internal, external environment, both) etc. such that variety is achieved. (Antonovsky, 1987, p. 77). The effect is to intertwine the sense of coherence components of comprehensibility, manageability, and meaningfulness, as when a comprehensibility item is couched in affective terms, giving it a meaningfulness tone, etc. To the extent possible, this "facet" approach has been followed in the new scale.

Approximately 1/3 of the items were newly written for the scale, based on the theoretical work of Antonovsky (1987) and Reiss (1981). Also consulted for construction of new items were Snyder's construct of "hope", or an individual's conviction of effective agency and available pathways for action (Snyder et al., 1991), and Olson, Larsen, and McCubbin's work (1982) on "family strengths" or family sense of loyalty and competency. Remaining items were adapted either from Antonovsky's (1987) individual scale, rewritten in family terms, or

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from the family sense of coherence scale in Sourani (1983) and Antonovsky and Sourani (1988).

Reliability and Validity

Content validity of the FLQ was reviewed by two reviewers. The scale was then piloted with 26 mothers from a broad SES, from urban and suburban areas similar to those of the study subjects. In recruiting subjects for the pilot, the only requirement was that the family have children of at least preschool age; it is not known whether some children in the families had chronic illness. Mothers completed the FLQ and the Family Environment Scale (FES, Moos, 1994). Obtained range of scores for the FLQ on this pilot was 171-231, with a mean score of 205, and a standard deviation of 20.1. Split half reliability (r11) was .87. As predicted, in a validity check, scores obtained on the FLQ correlated positively ($r=.40$, $p<.05$) with a factor on the FES identified by Kronnenberger and Thompson (1990) as relevant to care of children with chronic illness, in which family conflict is mediated by family organization and support. A moderately strong correlation was expected, because the FES measures primarily "within family" factors, while the FLQ also includes items that refer to the family's relations with the larger social world.

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Construct and Functional Definitions of Components

A copy of the FLQ with items categorized by component (comprehensibility, manageability, and meaningfulness items) and by category (within the family; between family and environment) is printed on pages 165-175. Construct definitions for each of the components are given as headings for groups of items, followed by functional definitions used in the construction of items. The consecutively numbered scale used in the study is given on pages 176-184. Symbols indicate the sources of items. Because the Antonovsky and Sourani (1988) scale is itself an adaptation of the individual Antonovsky (1987) scale, some items have two sources, and are so credited. Items not preceded by symbols were newly written for the scale. Sources are cited by symbol (*,†) on page 184.

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FAMILY LIFE QUESTIONNAIRE ITEMS CATEGORIZED BY COMPONENT

COMPREHENSIBILITY ITEMS

Construct definition. Comprehensibility refers to the "understandability" of one's internal and external world, the belief that the world has a lawfulness and structure that makes sense--and that the individual and the family "makes sense to others; the family is definable and its relationship to the larger society is understandable. For the family, the predictability of the world can include lawful change; the order of the world may include sad and traumatic events, but families can make sense of these events, discern their structure, place them in perspective.

Functional definition for items. These items refer to the extent to which family members understand one another, cognitively and affectively, the exchange of information that permits such understanding and predictability, the relative consistency of the social world and the extent to which family members believe there is understandability, predictability and consistency between the family and the social environment.

Within family comprehensibility items:

*1. In your family, is there a feeling that you all understand one another very well?

1	2	3	4	5	6	7
complete understanding among family members						lack of understanding among family members

†9. In the past two years, in your family there have been

1	2	3	4	5	6	7
many major changes						very few major changes

*† Please see p. 184 for sources of items.

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*11. In your family you feel that the rules and roles of family life are

1	2	3	4	5	6	7
very clear						very unclear

*20. To what extent do individuals share with other family members the events that cause them sadness?

1	2	3	4	5	6	7
full sharing						no sharing

23. When family members strongly disagree about an important issue, talking about it together usually leads to

1	2	3	4	5	6	7
less understanding						more understanding

*26. Does it seem to you that other family members can empathize with the way that you are feeling?

1	2	3	4	5	6	7
no one can "feel my feelings"						everyone can "feel my feelings"

*36. To what extent do family members share with one another the events that cause them joy?

1	2	3	4	5	6	7
no sharing						full sharing

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Between family & environment comprehensibility items:

5. When you discuss your child's well being and progress with concerned people such as teachers, activity leaders, doctors, the meetings usually are

1	2	3	4	5	6	7
predictable; they go as you expected						unpredictable; full of unexpected information and reactions

15. Think of the people with whom your family spends time socially. These people

1	2	3	4	5	6	7
really know what matters most to your family						don't really know what matters most to your family

28. In the past, when sad events occurred in the family it seemed to you that

1	2	3	4	5	6	7
"no one else can understand how we feel"						"most other people can understand how we feel"

33. For your family, knowing just what others in the community expect of you has been

1	2	3	4	5	6	7
perfectly clear						completely confusing

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MANAGEABILITY ITEMS

Construct definition. Manageability refers to family members' belief that resources are available to the family to meet the challenges of life. Such resources may be of many types--money, flexible coping strategies, family cohesiveness, ego strengths, social support, religious beliefs, trustworthy professionals, to name a few. This component may include but is not limited to internal locus of control; in many cases the family may feel a strong sense of agency and effectancy; but in other cases events are not within the family's control, yet are under control of sources the family considers legitimate, and the family has a sense of participation. Members are confident that resources can be accessed, and that, within reason, their use will be effective. Conversely, members acknowledge and act upon the legitimate expectations of others. Antonovsky notes that "to the extent that one has a high sense of manageability, one will not feel victimized by events or feel that life treats one unfairly. Untoward things do happen in life, but when they do occur, one will be able to cope and not grieve endlessly." (Antonovsky, 1987, p. 18.)

Functional definition for items. These items refer to problem solving, the efficacious or inefficacious enactment of goals, and family members' belief that resources are available to facilitate such enactment, both within the family, and between the family and environment.

Within family manageability items:

4. Suppose your family comes up against a problem and the solution isn't clear. You expect the family attitude will be

1	2	3	4	5	6	7
"there are lots of ways around any problem"						"I guess we're stuck with this"

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*13. In the past when hard to bear, difficult events have occurred in the family, the feeling was

1	2	3	4	5	6	7
"this is overwhelming us"						"this is a challenge fir us"

*†17. When you think of the major difficulties and painful events your family faces, the attitude in your family is

1	2	3	4	5	6	7
there are some things we just can't handle						we can always find a way to cope

*19. In the past, when a decision has been made on important family matters, the final decision has always been

1	2	3	4	5	6	7
satisfactory and good for all family members						good for only some family members; you can't satisfy everyone at once

†22. In the past, when unfortunate events that could not be changed have happened, the tendency was for the family to

1	2	3	4	5	6	7
hang onto what happened, going over it and over it						say "OK, that's that we have to live with it

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*†27. Do you feel that in your family, it will always be possible to receive help from one another when there is a problem?

1	2	3	4	5	6	7
you're certain family members will help						you doubt family members will help

*†29. Many families--even those with a lot of strength--sometimes feel ineffective, like "losers" in certain situations. In your opinion, how often has this been true of your family in the past?

1	2	3	4	5	6	7
never						very often

*30. When major family problems have arisen in the past, was it possible, in your opinion, to find out together how the problem happened?

1	2	3	4	5	6	7
to a small extent						to a great extent

†31. When you think of the major difficulties that are likely to get in the way of important family goals, you feel that

1	2	3	4	5	6	7
you'll surely reach your goals						you surely won't reach your goals

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Between family & environment manageability items:

*†2. In the past, when your family has had to do something that depended upon help form people outside the family, did you have the feeling that it

1	2	3	4	5	6	7
surely wouldn't get done						surely would get done

†6. Think of the interactions your family has with people and institutions outside the family. Do you have the feeling that others treat family members unfairly?

1	2	3	4	5	6	7
very often treated unfairly						very seldom treated unfairly

*8. Suppose that your family has to move, and there will be job changes for adults, school changes for children. You expect that the family's adjustment to the new situation will be

1	2	3	4	5	6	7
very easy						very difficult

18. When your family has needed reliable information from others--friends, community members, professionals--to help with an important problem, your experience was

1	2	3	4	5	6	7
it's impossible to find out what you need when you need it						we can always find what we need to know

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*21. Suppose that, in the future an important person or agency outside the family criticizes the family. In dealing with this, the family members will

1	2	3	4	5	6	7
unite to deal with the criticism						each deal with it separately

25. In the future, if a difficult problem arises that the family can't solve on its own, you expect that finding the right outside people to help will be

1	2	3	4	5	6	7
very easy						nearly impossible

†35. Has it happened that people whom you and your family counted on for support disappointed you?

1	2	3	4	5	6	7
often happened						never happened

MEANINGFULNESS ITEMS

Construct definition. Meaningfulness refers to the deep emotional investment, engagement, and commitment--the valuing--that family members and the family as a whole bring to every day life and to life's challenges. As the motivational element it is a central aspect of the sense of coherence, and has a strongly Existentialist quality, in that the meaning of an event is in part "made" by the family's active engagement. Life's problems are seen as worthy of the investment of emotional energy. Conversely, the family feel that the investment it makes is valued by the groups in which it participates. Family members with a strong family sense of meaningfulness will have faith in the family's ability to rise to a challenge, to actively commit to goals and find shared meaning even in times of trouble. They will trust that the family can make emotional sense of the world.

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Functional definition for items. These items refer to the emotional investment of the family in their interactions within the family and between the family, the sense of meaning they derive from these interactions, and their capacity to form goals and purposes and be committed to them.

Within family meaningfulness items:

3. It seems to you that, once the family sets a goal

1	2	3	4	5	6	7
some members care a lot less about it than others						everyone's behind it 100 percent

*†7. Until now, life in your family has had

1	2	3	4	5	6	7
very unclear goals						very clear goals

*†12. You anticipate that life in your family in the future will be

1	2	3	4	5	6	7
basically boring						full of interest

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*†16. When you think about life in your family, you very often

1	2	3	4	5	6	7
feel how good it is to be alive						ask yourself why you go on living like this

†32. How often do you have the feeling that there is very little meaning in the things that you do in daily family life?

1	2	3	4	5	6	7
very often						never

Between family and environment meaningfulness items:

10. Think about the groups you've been involved with as a family, or in support of a family member. The people in these groups

1	2	3	4	5	6	7
did not much value your participation						really valued your participation

14. When you think of the ties that your family has with the people in the community around you, these relationships seem

1	2	3	4	5	6	7
unfulfilling and routine						deeply meaningful and rewarding

24. In the next year or so, individual family member's involvement with people, and groups "outside" the family will probably make family life seem

1	2	3	4	5	6	7

more rewarding less rewarding

34. Your family finds that the "business of life"--working, daily interactions with community, participating at school--is

1	2	3	4	5	6	7
a source of interest and pleasure						dull and unrewarding

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FAMILY LIFE QUESTIONNAIRE*

In order to help us learn more about your family for the PKU Family Study, please answer each of these questions by circling the number from 1 to 7 that is closest to how you feel. There are no right or wrong answers; simply pick the number that you think best describes how things are for your family. Please answer all the questions, giving only one answer for each question.

*1. In your family, is there a feeling that you all understand one another very well?

1	2	3	4	5	6	7
complete understanding among family members						lack of understanding among family members

*†2. In the past, when your family has had to do something that depended upon help from people outside the family, did you have the feeling that it

1	2	3	4	5	6	7
surely wouldn't get done						surely would get done

3. It seems to you that, once the family sets a goal

1	2	3	4	5	6	7
some members care a lot less about it than others						everyone's behind it 100 percent

*†For item sources, please see page 184

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4. Suppose your family comes up against a problem and the solution isn't clear. You expect the family attitude will be

1	2	3	4	5	6	7
"there are lots of ways around any problem"						"I guess we're stuck with this"

5. When you discuss your child's well being and progress with concerned people such as teachers, activity leaders, doctors, the meetings usually are

1	2	3	4	5	6	7
predictable; they go as you expected						unpredictable; full of unexpected information and reactions

†6. Think of the interactions your family has with people and institutions outside the family. Do you have the feeling that others treat family members unfairly?

1	2	3	4	5	6	7
very often treated unfairly						very seldom treated unfairly

*†7. Until now, life in your family has had

1	2	3	4	5	6	7
very unclear goals						very clear goals

*8. Suppose that your family has to move, and there will be job changes for adults, school changes for children. You expect that the family's adjustment to the new situation will be

1	2	3	4	5	6	7
very easy						very difficult

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†9. In the past two years, in your family life there have been

1	2	3	4	5	6	7
many major changes						very few major changes

10. Think about the groups you've been involved with as a family, or in support of a family member. The people in these groups

1	2	3	4	5	6	7
did not much value your participation						really valued your participation

*11. In your family you feel that the rules and roles of family life are

1	2	3	4	5	6	7
very clear						very unclear

*†12. You expect that life in your family in the future will be

1	2	3	4	5	6	7
basically boring						full of interest

*13. In the past when very hard to bear, difficult events have occurred in the family, the feeling was

1	2	3	4	5	6	7

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"this is overwhelming us" "this is a challenge for us"

14. When you think of the ties that your family has with the people in the community around you, these relationships seem

1	2	3	4	5	6	7
unfulfilling and routine						deeply meaningful and rewarding

15. Think of the people with whom your family spends time socially. These people

1	2	3	4	5	6	7
really know what matters most to your family						don't really know what matters most to your family

*†16. When you think about life in your family, you very often

1	2	3	4	5	6	7
feel how good it is to be alive						ask yourself why you go on living like this

*†17. When you think of the major difficulties and painful events your family faces, the attitude in your family is

1	2	3	4	5	6	7
there are some things we just can't handle						we can always find a way to cope

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18. When your family has needed reliable information from others--friends, community members, professionals--to help with an important problem, your experience was

1	2	3	4	5	6	7
it's impossible to find out what you need when you need it						we can always find what we need to know

*19. In the past, when a decision has been made on important family matters, the final decision has always been

1	2	3	4	5	6	7
satisfactory and good for all family members						good for only some family members; you can't satisfy everyone at once

*20. To what extent do individuals share with other family members the events that cause them sadness?

1	2	3	4	5	6	7
full sharing						no sharing

*21. Suppose that, in the future an important person or agency outside the family criticizes the family. In dealing with this, the family members will

1	2	3	4	5	6	7
unite to deal with the criticism						each deal with it separately

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†22. In the past, when unfortunate events that could not be changed have happened, the tendency was for the family to

1	2	3	4	5	6	7
hang on to what happened, going over it and over it						say "OK, that's that, we have to live with it"

23. When family members strongly disagree about an important issue, talking about it together usually leads to

1	2	3	4	5	6	7
less understanding						more understanding

24. In the next year or so, individual family member's involvement with people and groups "outside" the family will probably make family life seem

1	2	3	4	5	6	7
more rewarding						less rewarding

25. In the future, if a difficult problem arises that the family can't solve on its own, you expect that finding the right outside people to help will be

1	2	3	4	5	6	7
very easy						nearly impossible

*26. Does it seem to you that other family members can empathize with the way that you are feeling?

1	2	3	4	5	6	7
no one can "feel my feelings"						everyone can "feel my feelings"

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*†27. Do you feel that in your family, it will always be possible to receive help from one another when there is a problem?

1	2	3	4	5	6	7
you're certain family members will help						you doubt family members will help

28. In the past, when very sad events have occurred in the family it seemed to you that

1	2	3	4	5	6	7
"no one else can understand how we feel"						"most other people can understand how we feel"

*†29. Many families--even those with a lot of strength--sometimes feel ineffective, like "losers" in certain situations. In your opinion, how often has this been true of your family in the past?

1	2	3	4	5	6	7
never						very often

*30. When major family problems have arisen in the past, was it possible, in your opinion, to find out together how the problem happened?

1	2	3	4	5	6	7
to a small extent						to a great extent

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†31. When you think of the major difficulties that are likely to get in the way of important family goals, you feel that

1	2	3	4	5	6	7
you'll surely reach your goals						you surely won't reach your goals

†32. How often do you have the feeling that there is very little meaning in the things that you do in daily family life?

1	2	3	4	5	6	7
very often						never

33. For your family, knowing just what others in the community expect of you has been

1	2	3	4	5	6	7
perfectly clear						completely confusing

34. Your family finds that the "business of life"--working, daily interactions with community, participating at school--is

1	2	3	4	5	6	7
a source of interest and pleasure						dull and unrewarding

†35. Has it happened that people whom you and your family counted on for support disappointed you?

1	2	3	4	5	6	7
often happened						never happened

*36. To what extent do family members share with one another the events that cause them joy?

1	2	3	4	5	6	7
no sharing						full sharing

Item sources:

* Adapted from Journal of Marriage and the Family, "*FAMILY SENSE OF COHERENCE AND FAMILY ADAPTATION*," Aaron Antonovsky and Talma Sourani; 50:1, 91-92. Copyrighted 1988 by the National Council on Family Relations, 3989 Central Ave. NE, Suite 550, Minneapolis, MN 55421. Reprinted by permission.

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APPENDIX B

PKU PARENT INTERVIEW

We'd like to learn more about your experience as a family in living with PKU, and how you help (name of child) **with the diet and** (his/her) **treatment.**

A. Everyday life at home

Let's start with the present, with what it's like for your family handling the diet now, on a day to day basis.

A1. Could you describe a typical weekday for (name of child), **and how you do things in your family to make the diet work?** (If the parent hesitates, say, **Just start with the morning of a typical school day.**

(Try to get a sense of the whole day. Note if parent covers getting ready for school, preparation of lunch, midday meal, after school activities & homework, meal time, bed time. If at least three general routines are not mentioned, ask specific questions such as **How does your family handle the diet at breakfast? Is there an after school routine? How has it been to fit the diet in to family dinner time?** until three are covered. Note if the parent at least mentions formula preparation, storage, food preparation, maintenance of food record, and daily adjustment of intake. If at least two are not mentioned, ask questions until two are covered such as **How is preparing the formula usually done? How do you keep track of what** (name of child) **has eaten?) And how does drinking the formula fit in?**

A2. On a daily basis at home, what aspects of following the diet are most difficult? (cover both food and formula; if parent does not bring it up, ask about child's degree of cooperation or resistance to the diet). Of one of the difficulties, ask, **Can you give me an example of that**? if the parent has not spontaneously done so. Follow up questions may be :

What do you think caused the difficulties that time?
How did you feel about the situation?
And how did (others involved, including the child) **feel?**
Was the problem resolved? How?

A3. Are there some routines connected with the diet that (name of child) **regularly handles?**

A4. Are there some special rituals that (name of child) **uses to help (himself/herself) to follow the diet?** (if needed, prompt) **For example, some parents say their child always uses the same cup and crazy straw, or always watches special video right after school when he drinks his formula...**

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A5. If you had to pick one or two words to describe how the diet fits into your family life, what would they be? Can you tell me why you chose those words? (Follow up prompt, only if needed): **Can you give me an example that shows why you chose those words?**

Traditions and activities within the family

B1. Now I'd like to ask you about events that are a little less frequent, some of the special things your family might do together regularly but not every day, maybe once a week, or on weekends Could you describe some of those activities? (If the parent can't think of any, prompt, **Like a special dinner every week, or a sport you watch together, or trips to the park)**

B2. How does (name of child's) **having to follow the diet affect these activities for your family?**

B3. May substitute: **How do you usually handle celebrations like birthdays?** Instead of all the following: **How about celebrations in your immediate family, such as anniversaries or birthdays? Do you make changes to accommodate the diet when it's another family member's celebration? When it's** (name of child's)**?** (If the parent does not do so, ask, **Can you give me an example?)**

C. Ethnic/extended family identity and celebrations

C1. Now we'd like to know more about your extended family, grandparents, aunts, uncles, cousins . Is there someone special in your extended family that you could turn to for help about (child's) **treatment, or is that something that stays pretty much in your immediate family?** (If yes, ask **Has this person actually been helpful? In what way?)**

C2. Are there ever conflicts with extended family members over the diet, or do things go fairly smoothly? (whichever the parent says, if the parent has not spontaneously given an example, ask **Can you give me an example of that?)**

C3. Your ethnic background is...? (If two parent home) **And your husband's...? Do your holiday celebrations include ethnic traditions? How important is food in your family traditions and celebrations? How about American celebrations-- Thanksgiving, Christmas season...**

(If the parent indicates that food & especially ethnic or particular foods are especially important, ask **Has that affected how involved and included** (name of child) **can be in the festivities? Have you been able to find ways to include the PKU diet in the holiday celebrations, or has that been hard to do?**

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C5. (optional) Are there some special recipes and traditions that you've developed around the diet that you've been able to contribute to these more extended family gatherings?

D. In the world

D1. May substitute **How do you usually handle things like school lunches, parties at school, visits to friends?** Instead of **How have you handled school lunches? Parties at school where food might be a problem? Visiting friends and participating in activities where food is involved?**

D2. In general, how do you feel others such as family friends, parents of your child's friends, teachers accept the situation? (If the parent says there have been difficulties, ask whether the family has been able to find way to cope with the situation).

D3. Does (name of child) **have a "standard way" he/she explains about the diet to others when that is necessary?**

D4. Do you feel being on this diet has affected your child's social life? In what way? Do you think it affects how s/he feels about himself or herself? In what way?

E. Professionals and procedures

E1. (Omit if this has been made obvious) **In your family, who usually makes contact with the PKU staff--arranges appointments, talks with the nutritionist, sees the blood work is done, and so on? Is that arrangement working well, or would you like to see some changes in that?**

E2. How would you describe the communication between your family and the PKU staff?

E3. What are some ways the clinic could be more helpful to you, your family and your child in treatment?

F General (Additions coded G)

G1. Now, PKU is something you can't see, and some parents have told me that, even now, even though they know it intellectually, deep down its hard for them to really believe their child has PKU. How has that been for you? Has it been hard for you to believe your child has PKU? What about what other family members believe? (If parent does not offer spontaneously, prompt):

What are the things that helped you to believe or doubt?

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G2. And the diet--do you believe it really works? Do other family members believe it works? (If parent does not offer spontaneously, prompt):

What helps you to believe or doubt it works?

G3. This is complicated treatment--so many adjustments. What motivates you to keep working at it? Do other family members feel the same?

G4. When you get your child's blood work results, are they usually about what you expected? (If results are a surprise, ask how often. Always follow up with):

When they're not what you expected, can a reason usually be found?

Do other family members talk the results over with one another, or does that stay pretty much with one family member?

G5. Suppose you got a single blood work that was high? How concerned would you be? What if you got a series of results that were high. How concerned?

Would other family members be equally concerned?

How would you find out what is causing the problem?

What steps would you take to deal with the problem?

Do you think the steps you described would be successful?

How would you know?

G6. At this time, just now, what are some of your family's goals for your child's treatment? And another goal? Any others?

How would you rate your commitment to each of these goals? (somewhat committed, committed, very committed)

Your husband's?

Your child's?

G7. Would you describe the diet as a burden or a challenge for the family, or both? (Tailor follow up questions in light of response):

In what major way has it been a burden for the family?

In what major way has it been a challenge for the family?

Now, I'd like you to think back

F1. When you first learned about (name of child's) **having PKU, what were your thoughts and feelings about what this would mean for your family?**

F1A. Was it difficult for you to understand why this had happened to a child in your family? Was it difficult for other family members? (If it is not given spontaneously, at least touch on the believability of the clinic's explanation, then if the "why us" gist isn't spontaneously given, ask):

Did you go through a "Why me, why us, why my kid" stage?

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Has that changed or stayed pretty much the same? (If the parent says it has passed, ask):
What do you think helped you through it?

F2. And how did your family find support during that time? Was there support? (Regardless of whether parent says others were or were not supportive, ask **Did that change or is it still the same?)**

F3. Now, looking back and reflecting, what are your thoughts and feelings about what living with PKU has meant for you and your family. (With care, and if appropriate, ask what it has meant to family relationships including spousal and siblings, family life style, and the family's relation to extended family. Who, apart from the child, does the parent see as most affected? How has the family as a whole been affected? Did this change much over time?)

F4. Looking forward, what are your hopes, concerns, and expectations about how (name of child) **will handle his/her treatment as s/he grows older? (If the parent does not spontaneously say s/he hopes for diet for life, add):**
There have been so many changes in thinking over the years about how long a child should stay on diet. What do you hope for your child? (If parent says stay on diet for life ask):
What would that involve in terms of what s/he does?

Thank you. We could take a few more minutes . . . . are there questions that you would like to ask me?

Or something you would like to tell me that we haven't covered?

Note: Questions about this interview should be addressed to Shaune Bornholdt, 501 West 121st Street, Apt. #52, NY, NY 10027.

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APPENDIX C

CHILD BEHAVIOR CHECKLIST FOR AGES 4–18

For office use only
ID #

Please Print

CHILD'S FULL NAME: FIRST MIDDLE LAST

SEX: ☐ Boy ☐ Girl

AGE

ETHNIC GROUP OR RACE

TODAY'S DATE: Mo. _____ Date _____ Yr. _____

CHILD'S BIRTHDATE: Mo. _____ Date _____ Yr. _____

GRADE IN SCHOOL _____

NOT ATTENDING SCHOOL ☐

Please fill out this form to reflect *your* view of the child's behavior even if other people might not agree. Feel free to print additional comments beside each item and in the spaces provided on page 2.

PARENTS' USUAL TYPE OF WORK, even if not working now. *(Please be specific—for example, auto mechanic, high school teacher, homemaker, laborer, lathe operator, shoe salesman, army sergeant.)*

FATHER'S TYPE OF WORK: _____

MOTHER'S TYPE OF WORK: _____

THIS FORM FILLED OUT BY:

☐ Mother (full name) _____

☐ Father (full name) _____

☐ Other—full name & relationship to child: _____

I. Please list the sports your child most likes to take part in. For example: swimming, baseball, skating, skate boarding, bike nding, fishing, etc.

☐ None

Compared to others of the same age, about how much time does he/she spend in each?

	Don't Know	Less Than Average	Average	More Than Average
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

Compared to others of the same age, how well does he/she do each one?

	Don't Know	Below Average	Average	Above Average
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

II. Please list your child's favorite hobbies, activities, and games, other than sports. For example: stamps, dolls, books, piano, crafts, cars, singing, etc. (Do *not* include listening to radio or TV.)

☐ None

Compared to others of the same age, about how much time does he/she spend in each?

	Don't Know	Less Than Average	Average	More Than Average
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

Compared to others of the same age, how well does he/she do each one?

	Don't Know	Below Average	Average	Above Average
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

III. Please list any organizations, clubs, teams, or groups your child belongs to.

☐ None

Compared to others of the same age, how active is he/she in each?

	Don't Know	Less Active	Average	More Active
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

IV. Please list any jobs or chores your child has. For example: paper route, babysitting, making bed, working in store, etc. (Include *both* paid and unpaid jobs and chores.)

☐ None

Compared to others of the same age, how well does he/she carry them out?

	Don't Know	Below Average	Average	Above Average
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

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Child Behavior Checklist/4-18, p. 2

Please Print

V. 1. About how many close friends does your child have? ☐ None ☐ 1 ☐ 2 or 3 ☐ 4 or more
(Do *not* include brothers & sisters)

2. About how many times a week does your child do things with any friends outside of regular school hours?
(Do *not* include brothers & sisters) ☐ Less than 1 ☐ 1 or 2 ☐ 3 or more

VI. Compared to others of his/her age, how well does your child:

	Worse	About Average	Better	
a. Get along with his/her brothers & sisters?	☐	☐	☐	☐ Has no brothers or sisters
b. Get along with other kids?	☐	☐	☐	
c. Behave with his/her parents?	☐	☐	☐	
d. Play and work alone?	☐	☐	☐	

VII. 1. For ages 6 and older—performance in academic subjects. ☐ Does not attend school because _____

Check a box for each subject that child takes	Failing	Below Average	Average	Above Average
a. Reading, English, or Language Arts	☐	☐	☐	☐
b. History or Social Studies	☐	☐	☐	☐
c. Arithmetic or Math	☐	☐	☐	☐
d. Science	☐	☐	☐	☐
Other academic subjects—for example: computer courses, foreign language, business. Do *not* include gym, shop, driver's ed., etc. e. _____	☐	☐	☐	☐
f. _____	☐	☐	☐	☐
g. _____	☐	☐	☐	☐

2. Does your child receive special remedial services or attend a special class or special school? ☐ No ☐ Yes—kind of services, class, or school:

3. Has your child repeated any grades? ☐ No ☐ Yes—grades and reasons:

4. Has your child had any academic or other problems in school? ☐ No ☐ Yes—please describe:

When did these problems start?

Have these problems ended? ☐ No ☐ Yes—when?

Does your child have any illness or disability (either physical or mental)? ☐ No ☐ Yes—please describe:

What concerns you most about your child?

Please describe the best things about your child:

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APPENDIX D

THE CITY COLLEGE
OF
THE CITY UNIVERSITY OF NEW YORK
NEW YORK, N.Y. 10031

THE PSYCHOLOGICAL CENTER
DEPARTMENT OF PSYCHOLOGY
(212) 690-6602, 3, 4

Consent Form for PKU Study

Shaune Bornholdt, M.A., Principal Investigator

Thank you for your willingness to participate in this study of children with PKU and their families.

1. You will be asked to complete several questionnaires, two about family life, two about your child's social skills and behavior. You will also be asked to answer a few questions about your child's progress at school, and to complete a brief demographic sheet.

2. By signing this form, you will indicate your consent to the researcher's obtaining information about your child's treatment and phenylalanine levels over the past year from the medical director at your child's PKU treatment center.

3. Following your response to the questionnaires, we would like to interview some parents to learn more about your family's experience in dealing with PKU. If you agree to be interviewed, please let us know by checking the box at the end of this form.

4. There are no anticipated risks in participating in the study. You are free to decline to complete the study. Your decision to participate or not to participate will not affect your child's medical treatment in any way.

5. You and your child have the right to privacy. Completed questionnaires will be kept confidential by the researcher, and will not be made available to the treatment center or to anyone else. Data will be reported as group correlations. In any discussion of findings, no names of participating individuals will be used.

I have read the above consent form and I understand it. All questions regarding this form or the study have been answered to my satisfaction. I am the parent or legal guardian of _____(name of child with PKU).

Name:_____ Signature:_____

☐ I also agree to be interviewed.

Date:_____ Witness: _____

This research is being conducted under the auspices of the Program in Clinical Psychology at the City University of New York. Questions may be addressed to the investigator, Shaune Bornholdt, M.A., Counselor, PKU Program of Northern N.J., 501 West 121st Street, Apt. 52, N.Y., N.Y. 10027, telephone (212) 662-0137.

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APPENDIX E

PKU FAMILY STUDY

To help us with the study, we need to know a little more about your family.

How many people live together in your home? _____
Are there one or two parents living in your home? _____

If grandparents of the child or other related adults are living at home, please list them and their relationship to your child.

What is the age of your child with PKU? ____ Date of birth _____ _Boy _girl
Child's ethnic group (optional)
__Asian __Black __Hispanic __Indian (Native American) __White
__Other
Please list the ages and sex of other children living at home:

Mother's highest grade or year in school completed?

__ elementary school
__ 1-3 yrs, high school
__ high school graduate
__ technical school
__ 1-3 yrs. college
__ college degree
__ advanced degree

Father's highest grade or year school completed?

__ elementary school
__ 1-3 yrs. high school
__ high school graduate
__ technical school
__ 1-3 yrs. college
__ college degree
__ advanced degree

Parents' type of work (please be specific, for example, auto mechanic, computer systems analyst, army sergeant, homemaker, nurse (RN), nurse (LPN), delicatessen clerk, elementary school teacher)
Mother's work_____
Father's work _____

Thank you very much for your help.

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IMAGE EVALUATION
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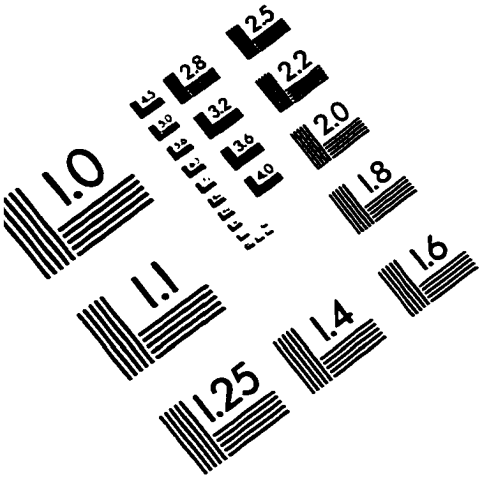

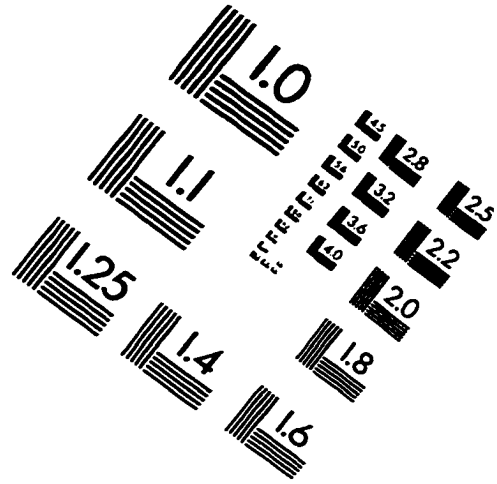

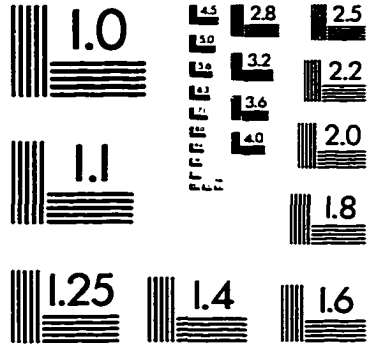

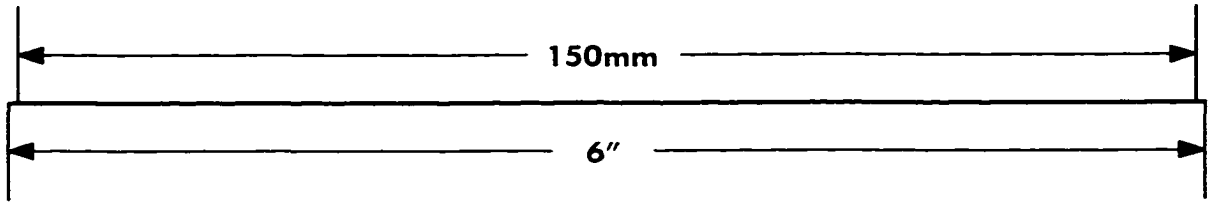

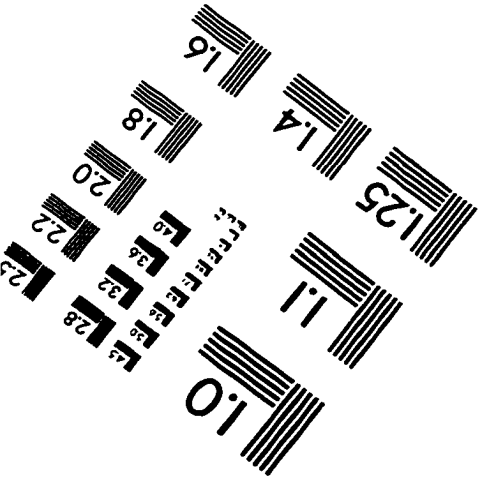

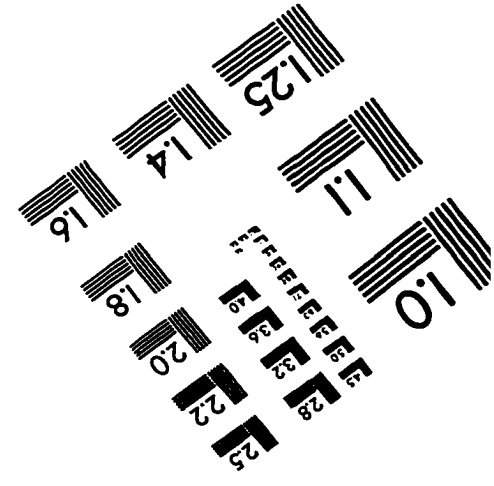

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