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THE RELATIONSHIP OF FAMILY FUNCTIONING TO INFANT, PARENT AND FAMILY ENVIRONMENT OUTCOMES IN THE FIRST 18 MONTHS FOLLOWING THE BIRTH OF AN INFANT WITH MYELODYSPLASIA

by

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ABSTRACT

THE RELATIONSHIP OF FAMILY FUNCTIONING TO INFANT, PARENT AND FAMILY ENVIRONMENT OUTCOMES IN THE FIRST 18 MONTHS FOLLOWING THE BIRTH OF AN INFANT WITH MYELODYSPLASIA

By

Suzanne Lee Bellinger Feetham

The incidence of neural tube defects is 1.7 to 3 per 1,000 live births in the United States with 130 such births each year in Michigan. Myelodysplasia (also know as myelomeningocele and/or spina bifida) is the most common (60%) of the neural tube defects and is the one defect most frequently associated with survival. Because of improved surgical and medical techniques, increasing numbers of children with this defect are surviving and living with their families. Although myelodysplasia is a serious birth defect, persons with this defect grow to live productive adult lives. The physical manifestations of myelodysplasia include hydrocephalus in 80% of the children, handicaps of locomotion and continence in 95%, and perceptual and learning disabilities and/or intellect below their unaffected siblings in 25%.

The birth of an infant with myelodysplasia affects the family in several ways because the infant requires special care in the home and in both the health care and educational systems. The "cost" of this special care in both energy expended by the family members and in monetary terms is but one effect on the family system. The long term outcome of families raising these children in the home is documented and indicates an increased incidence of divorce, separation, maternal malaise and sibling problems over families without children with chronic health problems (Richards & McIntosh, 1973; Tew & Laurence, 1973).

The conceptual framework selected for this research is the structural-functional approach to family study. Family functioning is conceptualized holistically as the activities of everyday life or the way in which the family, as a system, operates across many dimensions. Clinical observations, the review of family theory and the review of research related to children with myeldoysplasia and their families suggest there are relationships among infant, parent and family environment variables and family functioning. Therefore, the purpose of this study was to examine the relationships among variables pertaining to the parents, to the infant with myelodysplasia and to the family environment with family functioning at five time intervals in the first 18 months following the birth of the infant.

The dependent variable, family functioning, is a composite score of family functioning across 21 indicators. The independent variables are composite scores of infant, parent and family environment variables as measured by specified items from the Parent Survey (Feetham & Perrin, 1977), Profile of Mood States (POMS) (McNair, Lorr & Droppleman, 1971), and infant hospital records and developmental testing using the Early Intervention Developmental Profile (Rogers & D'Eugenio, 1977).

The subjects for this descriptive longitudinal study were parents and their infants with myelodysplasia admitted to a Myelodysplasia Care Center in a large urban pediatric hospital. The study sample was a total of 38 infants and 66 parents (38 mothers and 28 fathers) who both met the study criteria and agreed to participate.

Three hypotheses were tested in this study. Multiple linear regression with stepwise inclusion and listwise deletion was used to test Hypotheses I and II. A simple ANOVA with repeated measures and t tests were used to test Hypothesis III.

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Hypothesis I, that the infant and parent-family environment predictor variables add significant information to the ability to predict family functioning at 3, 6, 12 and 18 months, was supported by the data. However, Hypothesis II, that the infant composite score variables would account for a greater proportion of variance than the parent variables in predicting family functioning, was not supported. Also, Hypothesis III, that the family functioning discrepant score would be higher at the one year anniversary of the infant's birth than at 3, 6 and 18 months, was not supported by the data.

Dedicated to Ann Marie Schraw who, as an infant, introduced me to the world of children with myelodysplasia and their families: for her tenacious commitment for the fullest life and for her family's unrelenting support of her commitment. To her family for sharing their family for my personal and professional growth these last 18 years.

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CHAPTER I

INTRODUCTION TO THE STUDY

Problem

The incidence of neural tube defects is 1.7 to 3 per 1,000 live births in the United States with 130 such births each year in Michigan. Myelodysplasia (also known as myelomeningocele and/or spina bifida) is the most common (60%) of the neural tube defects and is the one defect most frequently associated with survival. Because of improved surgical and medical techniques, increasing numbers of children with this defect are surviving and living with their families. Although myelodysplasia is a serious birth defect, persons with this defect grow to live productive adult lives.

The defect occurs during early embryonic life and by the twenty-eighth day the structural abnormalities are established. Myelodysplasia involves the herniation of the meninges and neural tissue through a spina bifida. A spina bifida, failure of the vertebrae to close, is found in 8% to 20% of the general population and is usually without symptoms. It is when the meninges and spinal cord also fail to develop that myelodysplasia and its associated problems of hydrocephalus (80%), lower extremity paralysis (95%), and bowel and bladder incontinence (95%), occur. In addition to the physical disabilities of lower extremity paralysis, spinal curvature and skin anesthesia, there is an increased incidence of perceptual and learning disabilities and intellect below their unaffected siblings (25%). The expected life span of these children is full adulthood if they reach adolescence without obesity and major kidney disease (McLaughlin & Shurtleff, 1979).

The birth of an infant with myelodysplasia affects the family in several ways. An initial and ongoing effect is that several professionals are introduced into the family system, far beyond the usual number introduced with the birth of a normal child. Secondly, the infant requires special care in the home and in both the health care and educational systems. This special care takes more time than the care of a normal infant and often requires the addition of special equipment in the home. The "cost" of this special care in both energy expended by the family members and in monetary terms is a third effect on the family system.

The long term outcome of families raising these children in the home is documented and indicates an increased incidence of divorce, separation, maternal malaise and sibling problems over families without children with chronic health problems (Richard & McIntosh, 1973; Tew & Laurence, 1973; Walker, Thomas & Russell, 1971). However, the data are usually obtained after the child has been in the home several years and the onset and process for the development of these problems is not documented.

In spite of the multiple effects of the birth of an infant with myelodysplasia on the family, the research reports on these families indicate the majority of studies used a univariate linear design and were conducted by members of a single discipline at one time period. The studies have included outcomes focusing primarily on single measures of 1) patterns of parental response to the birth of the infant (Freeston, 1971; Mercer, 1974); 2) sibling responses and school behavior (Richard & McIntosh, 1973; Walker, Thomas & Russell, 1971); 3) marital integrity (Tew, Laurence, Payne & Rawnsley, 1977; Trembath, 1976); 4) family life style changes (Schonell & Watts, 1956; Walker, Thomas & Russell, 1971); 5) physical and mental health of the mothers (Tew & Laurence, 1973; Walker, 1970); 6) family functions (Dunlap & Hollingsworth, 1977; Travis, 1978); or 7) family planning and sexual relations (Richards & McIntosh, 1973; Walker, Thomas, & Russell, 1971).

The basis for the traditional linear model in studying families with children with birth defects is a medical model. A medical model follows the process of delineation of symptoms, diagnosis and treatment. Etiology has been sought through the study of biology, anatomy and physiology. As researchers in other professions began to study these families they maintained the linear approach of the medical model, but examined a different set of variables resulting in univariate analyses of a very complex multivariate situation. The result of univariate research is that pieces of the family, child and/or the handicap are examined. In studying the effect of the birth of an infant with myelodysplasia on the family the need is for frameworks from which structural-functional characteristics can be examined, while also recognizing the interdependence of the many factors within the family system and between systems interacting with the family.

In addition to using univariate designs which do not illustrate interaction among the infant, parent and family environment variables, researchers studying families of children with myelodysplasia have used the mother as the primary data source, have used retrospective data collection and have conducted the studies on non-American populations. When a longitudinal design was used, data collection started 4-5 years following the birth of the child (Tew, Laurence, Payne & Rawnsley,1977). When studied in families with children with myelodysplasia, family functioning is viewed as a variable dependent upon the presence of the child. The interrelationships among parent, infant and family environment variables as they relate to family functioning have not been studied.

Purpose

In recognition of the limitations and results of previous studies of families of children with myelodysplasia, the purpose of this study is to examine the interrelationship among variables pertaining to the parents, to the infant with myelodysplasia and to the family environment as they relate to family functioning at selected time intervals in the first 18 months following the birth of the infant. Knowledge of the patterns of family functioning in the initial period following the birth of the infant and the identification of parent, infant and family environment variables accounting for the greatest variance in family functioning may be useful to the health practitioner in planning the appropriate times and modes of interventions for these families.

This study is part of a larger descriptive study examining both retrospectively and prospectively the outcomes of infants selected for non-surgical treatment of their myelodysplasia versus those infants selected for treatment. The prospective study, begun in 1977, includes parent and family environment data in addition to the infant data. This research was supported by the U. S. Public Health Service under grant #NU00632-01 awarded by the Nursing Research Branch, Division of Nursing, Bureau of Health Manpower, Health Resources Administration, Department of Health and Human Services.

Hypotheses

This research will focus on the effect selected parent, infant and family environment variables have on family functioning in the first 18 months following the birth of an infant with myelodysplasia. Family functioning is conceptualized for the purpose of this study as the parental perception of the degree that family functions are achieved in relation to how much there should be in three areas of relationship: 1) relationships with broader social units, 2) relationships with family subsystems, and 3) relationships with individual members. The difference between what parents perceive there is and should be constitutes a discrepant score, implying parental expectations of family functions are not being met.

The research hypotheses are: 1) a significant amount of variance in family functioning will be accounted for by infant outcome, parent outcome, and family environment variables at each of four time intervals (3, 6, 12 and 18 months) following the birth of an infant with myelodysplasia.

2) The composite infant scores including general status, care interventions, and central nervous system status at the neonatal and follow-up period will account for greater variance in family functioning than will parent composite scores of: general status, including marital status, education, health, parent experiences and family environment; perceptions of the perinatal periods; and Profile of Mood State Scores at 3, 6, 12 and 18 months following the birth of an infant with myelodysplasia.

3) Family functioning discrepant scores as measured by parent reports will be greater at the first year anniversary of the birth of

the infant with myelodysplasia than at 3, 6 and 18 months following the birth.

Theory

The conceptual framework selected for this research is the structural-functional approach to family study augmented by concepts from the developmental and family ecological approaches. Also referred to as functional analyses, the structural-functional frameworks for examining family functioning, the dependent variable for this study, are derived from an overall functionalist theory of society, psychology, and social anthropology (Eshelman, 1974). This theoretical origin will be reviewed followed by a discussion of characteristics, limitations and strengths of the structural-functional framework. A brief review of the current status of this framework will be followed by a review of family functions identified by family scholars. The ability to integrate the family structural-functional framework into evolving frameworks such as family development and family ecology supports the relevance of this framework for the study of the effect of the birth of an infant with myelodysplasia on the family in the first 18 months following the birth. The family ecology framework, which recognizes the interdependence of the infant, parent and family environment variables with family functioning, is the basis for the multivariate design of this study (Garbarino, 1977; Morrison, 1974).

Eshelman (1974) identifies the structural-functional framework as a dominant theoretical orientation which evolved in the 30's. This framework is commonly used in the study of families because of the overlap of concepts among the structural-functional, developmental and general systems frameworks (Aldous, Osmond & Hicks, 1979; McIntyre, 1966).

The Structural-functional framework as used in family research is attributed to several origins. The first is the functionalist branch of psychology, particularly the Gestalt position as it focuses on the relationship between a whole and its parts (Eshelman, 1974). Pitts (1964), however, identifies the origin as the biological sciences because the use of Aristotelian concepts were insufficient for explaining causality between the parts of living organisms. Two additional possible origins for the structural-functional approach are social anthropology as presented by Malinowski (1939) and Radcliffe-Brown (1935), and social system theorists such as Parsons (1949) and Merton (1968). All of these theorists recognized the interdependence of any aspect of an individual with their setting or larger system. In the application of these theories to the study of families, at times scholars have interpreted the theory out of context which results in a loss of adaptability and relevance of the theory to some research questions (Aldous, Osmond & Hicks, 1979).

Structural-Functional Framework Characteristics

Several characteristics of the structural-functional framework apply to this research. First, the task of functional analysis is to explain the parts, the relationship between the parts, the relationship between the parts and the whole, and the functions that are performed by, or result from, the relationship formed by the parts (McIntyre, 1966). Review of this relationship between and within systems guides the researcher in the identification of variables for study. By defining the whole in this study as the relationship between systems, variables identified for study include family members, the parents, the infant and siblings, and the family environment (e.g., status of the home and community and interaction with the health care system).

The structural-functional framework in recognizing system relationships allows for examination of animate (person) variables and inanimate (environmental) variables (Morrison, 1974). This characteristic is important as families with children with myelodysplasia may change their home environment to have adequate space for the special equipment used for the care of the child and/or to live within a convenient distance to specialized health care and educational facilities (Dunlap & Hollinsworth, 1977).

A third characteristic of the structural-functional framework of importance to this study is that the interrelationships and implied interdependence suggest that the groups, systems and behaviors of families and their members are not random and without regularity and predictability (Eshelman, 1974; McIntyre, 1966). Further, Pitts (1964) notes the structural-functional framework should always be capable of showing some reciprocal action. It is therefore anticipated that knowledge of parent, infant and environmental factors will show a relationship with the family's level of functioning.

In addition, a fundamental emphasis of this framework is the importance of maintaining the family system. Studies of families of children with myelodysplasia and other conditions resulting in chronic impairment indicate these families have a higher incidence of divorce, separation, and maternal malaise (Dorner, 1975; Tew & Lawrence, 1973) than families wihtout children with myelodysplasia or other chronic health problems. The system maintenance focus of the structural-functional framework implies that a variety of factors within and outside the family influence the level of functioning of the total unit. Therefore, in this author's research, variables within and outside the family will be studied. For example, one such measure, leisure activity, is examined as it contributes to the total family system including individual variables of the parents and the infant with myelodysplasia.

Family functions

The use of family functions is prevalent in research derived from the structural-functional framework. Some researchers define functions of the family by asking the question, "what does it do?", as in actual activities (McIntyre, 1966). Another definition is in terms of the consequences or results of the activities of the family or functional unit. Function is also seen as the contribution an item makes to the maintenance of the whole. Simply stated, the first definition implies the concept of process, the second implies its outcome, and the third implies its content.

The primary value of the various definitions of family functions is that they provide general direction to the researcher to consider problems of the relationships between the family system and other societal systems. A limitation of these definitions is that they have limited theoretical weight, therefore leading the researcher into statements of generalities and causalities (Pless & Satterwhite, 1973; Smilkstein, 1978). Family functions as presented by several scholars are presented in Chapter II as a basis for the specific family functions used in this study.

Assumptions

Several assumptions about the family with an infant with myelodysplasia are derived from the structural-functional framework:

1) family functioning is the sum of past experience of family members added to the present combined with future goals and expectations;
2) family functions develop and change over time;
3) family members of all ages act upon and react to their environment; and
4) family functions are influenced by and influence societal expectations, values and norms.

Limitations of the Study

The principal limitation of this study rests with the sample. It is a convenience sample of 38 families of infants from a total of 46 infants treated in one myelodysplasia care center in a large metropolitan area in Michigan. The sample is assumed to be representative of the families with infants with myelodysplasia throughout the state, but it may be biased by referral patterns from outstate hospitals into a private, urban hospital.

The total sample includes 38 families of which only a limited number supplied the data at each of the time periods. Therefore, the data are analyzed as individual sets at each time period rather than as one set of data with repeated measures over 18 months. The failure to obtain data at each time interval for each family limits the ability to generalize from the data. Incomplete data sets for each family resulted from delayed referral to the myelodysplasia care center, withdrawal from the care center during the 18 month study period and/or inconsistent parental participation in the study. It is not known if the parents volunteering to participate have the same characteristics as those who did not volunteer.

A further limitation is having the same informant provide data for both the dependent variable and some independent variables. Also, some data were collected retrospectively, thus subjecting the responses to the effects of cognitive dissonance and poor recall.

Importance

Of primary importance is that this research derives from a clinical practice question related to variables affecting the family following the birth of an infant with myelodysplasia. The researcher, through clinical experience with these children and their families, can attest to the significance, to clinical practice, of systematic documentation of family functioning following the birth of an infant with this defect.

Another significant feature is that this study provides information not currently available regarding the effect of the birth of an infant with myelodysplasia on its family. Previously researchers have not examined families prospectively from the neonatal period and longitudinally at several time periods through 18 monhts. In addition, few studies include examination of parents and infants simultaneously. This study includes the examination and discussion of the interrelationships among the infant variables, parent variables and variables in the family environment in relation to their predicting family functioning. A third significant aspect of this study is that data collection includes both the mother and father in contrast to family sociology studies which tend to be comprised only of mothers' perceptions (Safilios-Rothschild, 1970).

This is a prospective study from the time of birth of the infant. This is significant because previous studies of these children and their families have been initiated when the child is over five years of age. Although the incidence of divorce, sibling, child care and other family problems are identified in these previous studies, the process leading to these problems can only be hypothesized using retrospective methodology (Richards & McIntosh, 1973; Tew & Laurence, 1973; Walker, Thomas & Russell, 1971).

A review of the pertinent literature related to family functioning, the defect myelodysplasia including the child and outcomes, and the effects on the family is presented in Chapter II. An analysis of factors affecting the response of families to the birth of an infant with myelodysplasia including parent and family environment variables is presented to support the study design and data analysis.

CHAPTER II

LITERATURE REVIEW

Independent Variables

Through the review of the literature significant and current concepts related to the birth defect myelodysplasia, including the etiology, the effects of this birth defect on the development of the child, and the effects of a child with this birth defect on the family are identified. The concept of family functioning is analyzed to identify those functions pertinent to this research. A model for the study of the interaction of selected parent, infant and family environment variables on family functioning following the birth of an infant with myelodysplasia has resulted from this literature review.

Infant Outcomes

The effects of the birth of a child with myelodysplasia or other chronic diseases on families are frequently reported in the literature. The child with a defect is identified as affecting: employment decisions of parents; living location; vacations and leisure activity; marital relationships; family closeness; siblings, extended family interactions and economic security (Dunlap & Hollinsworth, 1977). In many studies the child with myelodysplasia is identified as the single independent variable determining the dependent variables listed above. However, in addition to specific child variables, there are variables peripheral to the child and variables within the parents and the family environment interacting to influence family functioning (Bronfenbrenner, 1974; Paolucci, Hall & Axinn, 1977; Sprey, 1973).

Variables about the child affecting family functioning include the parent's perception of the neonatal period and the process of being informed of the defect, the number and duration of hospitalizations of the infant, and the infant's developmental status (Price-Bonham & Addison, 1978). In this study variables related to the infant are conceptualized as one of the independent variables interrelating with parent and family environmental variables all of which affect family functioning. The infant variables inclusive of the etiology of the defect of myelodysplasia and its related clinical mainfestations and care are presented first followed by the parent outcomes and family environment variables affecting family functioning.

Etiology of Myelodysplasia The causes of neural tube defects have not been clearly identified. Some investigators support an environmental hypothesis while others support a genetic hypothesis. The environmental etiology is supported by geographic variations and seasonal and annual fluctuations in the frequency of occurrence. Increased risk of occurrence in siblings after an affected child is born and racial differences support the genetic hypothesis (Carter, 1974; Lorber, 1965).

The highest incidence of neural tube defects occurs in the British Isles (Coffee & Jessop, 1957; Record & McKeown, 1949; 1950a; 1950b; Smithells, Chinn & Franklin, 1964). The frequency of malformation for this region is estimated to be approximately 3.5 to 10.0 per 1,000 live births (Cziezel & Revesz, 1970; Penrose, 1957). In the United States an East-West gradient has been observed with the highest incidence occurring in the New England states and decreasing west of the Rocky Mountains (Alter, 1962; 1963). Descendants of Irish immigrants in Boston have a lower incidence than in Dublin but higher than among non-Irish Bostonians (Naggan & MacMahon, 1967). Over 8,000 infants with neural tube defects are born each year in the United States (Milunsky, 1977). The incidence is higher among the poor (MacMahon, 1970) and among Caucasians. The ratio of girls to boys is about 3:2 (Brockelhurst, 1976).

The incidence of the anomaly is usually reported in relation to 1,000 live births which fails to account for a stillbirth rate estimated by Laurence (1969) to be about 25 percent. The overall incidence is computed to be about two per thousand live births. In Michigan, the incidence of neural tube defects is about three per thousand live births (130 per year), of which 1.7 to 2 (60%) are children with myelodysplasia.

The incidence in the British Isles in the last three years has declined significantly with the implementation of mass prenatal screening for neural tube defects followed by abortion of the affected fetus (Sunderland & Emery, 1979). The presence of the defect is confirmed by the finding of elevated alpha-feto-proteins in the amniotic fluid and/or maternal serum in the 14th to 17th week of pregnancy (Brockelhurst, 1976). Prenatal diagnosis is available in the United States, but mass screening has only been used for research purposes (Macri, Weiss, Tillitt, Balsam, & Elligers, 1976; Milunsky, 1979).

In addition to the regional and ethnic differences in the frequency of occurrence, there seem to be trends of secular and seasonal nature. Gradual decline in the incidence was reported over the period 1940 to 1959 in at least two locations: Scotland and New York. Yet occasional peak years were also observed (Carter, 1965; 1974). Increase in the incidence of births during the winter months, especially in December, as compared to summer, has also been observed. The incidence of myelodysplasia and hydrocephalus relative to maternal age and birth order was investigated by Ingalls, Pugh & MacMahon (1954). The greatest incidence for Caucasians was observed for birth rank 1, the lowest incidence for birth rank 2, and a subsequent gradual increase with increasing birth order. The maternal age effect was not detected when birth rank was taken into consideration.

The genetic hypothesis is supported by Polman (1950), and Lorber (1965) who suggest a single gene recessive causality. Penrose (1946; 1957) however, suggests that familial incidence is too low to be accounted for by a single recessive gene hypothesis and proposes a polygenic hypothesis. Unfortunately study of the multiple gene inheritance hypothesis lacks precision and is difficult to substantiate. Preliminary work by Mayeda and Feetham (1974) on eighty-six Michigan families supported a multiple gene hypothesis.

Suggested environmental causes for myelodysplasia are agents such as influenza (Sever, 1972); zinc deficiency (Sever & Emanuel, 1972); hormonal pregnancy test (Gal, 1972; Sever, 1973); potato blight (Renwich, 1973); and softness of local water supply (Fedrick, 1970). Canned meat and tea consumption have also been linked to the occurrence of these defects (Carter, 1974). However, none of these agents have been sufficiently substantiated to be accepted. In the final analysis, the conclusion of genetic predisposition triggered by as yet unknown environmental factors, i.e., genetic <u>and</u> environmental rather than genetic <u>or</u> environmental factors, will probably be the answer to causation.

The lack of specificity for the causation of myelodysplasia is a factor which needs to be taken into account in the study of the effect of an infant with myelodysplasia on the family. Parents identify that their sexual relations are affected by the fear that they could have another child with this defect. In addition families review their pedigrees for additional evidence of defective genes. Therefore, data on a positive family history for neural tube defects and fetal loss, birth order for the infants with myelodysplasia, and maternal age will be used in constituting the independent variables in this study.

Clinical manifestations of myelodysplasia The clinical manifestations of myelodysplasia are evident in several major body systems. These manifestations will be reviewed from the perspective of their effect both on the child and the family system.

Myelodysplasia means failure of nerve development and in this case refers specifically to the spinal cord which is a major structure in the central nervous system. This developmental failure may occur at any point from the cervical (neck) area to the sacral (lower back) area with the most common site being at or below the lumbar region (80%). Since this occurs early in fetal life, further development of the entire central nervous system is altered causing brain and/or brainstem pathology in addition to the spinal cord defect. The level of cord dysplasia and degree of brain malformation determines the degree of paralysis and loss of limb function (95%) and the occurrence of hydrocephalus in 75-80% (McLaughlin & Shurtleff, 1979).

Because the nerves affecting bowel and bladder control originate in the second through fourth sacral segments of the spinal cord, over 95% of the children have bowel and bladder incontinence. Incomplete evacuation evidenced by chronic dribbling of urine and feces is common and increases during periods of crying and activity. Urinary tract complications such as urinary tract infection, ureteral reflux, upper tract dilation and incontinence are also common manifestations (Cass, 1976). The outcomes to the family for urological management are the need to express urine manually (crêde) from the bladder or to insert a catheter into the bladder to remove the urine; to take the infant for periodic urological examinations; and to monitor the infant for complications. If urinary continence cannot be achieved with catheterization, augmented by medication, or by crêde, insertion of an artificial sphincter is now a possibility for a few children. After the age of two the child is placed on a bowel management program to achieve regular timed predictable evacuations. All of these outcomes consume time, energy and increase the costs of raising the infant. In addition, the parents may be distracted from the normal aspects of the infant and focus on the special care. Parents often report that it is hard to find babysitters who will perform these care functions in the parents' absence.

A primary question from parents of infants with myelodysplasia is the infant's potential for ambulation. The most important factors determining ambulatory status are level of the lesion, motor power within a given neurosegmental level and the extent and degree of orthopedic deformities (DeSouza & Carroll, 1976). Hip dysplasia, deformities of the feet and spinal curvature are the most common orthopedic deformities. In order to achieve ambulation, multiple surgical procedures on the hips and feet, interval casting, splinting and finally bracing are required. In addition, the infant receives physical therapy augmented by parents exercising the infant at home. Prior to braces, special equipment such as standing tables and parapodiums are used.

Each aspect of orthopedic intervention requires appointments with health professionals and hospitalization. Concerns which accompany the appointments and hospitalizations are transportation, babysitting for siblings, meals away from home and the loss of work time.

As indicated, 85% of these children develop hydrocephalus which is the accumulation of cerebral spinal fluid in the ventricles of the brain. In infants, hydrocephalus is evidenced by the head size increasing faster than normal, enlarged fontanels, developmental delay and cerebral dysfunction. The treatment of choice is the surgical insertion of a ventricularperitoneal shunt to divert the excess accumulation of cerebral spinal fluid into the abdominal cavity where it is reabsorbed into the body circulatory system (Portnoy & Croissant, 1978; Shurtleff, Foltz & Loeser, 1973). Following insertion of the shunt the children are monitored for the rest of their lives for the clinical evidence of shunt dysfunction. Ideally the original shunt and drainage tubing will function adequately and not become blocked, dislodged and/or infected.

The relationship of later cognitive development to the incidence of hydrocephalus is significant, however, reports of the relationship to incidence of shunt dysfunction and the onset of treatment for the dysfunction are inconsistent. Tew & Laurence (1974) showed children with hydrocephalus treated by shunts are significantly less intelligent than their normal siblings, whereas children with myelodsyplasia but without hydrocephalus and shunts were the same intelligence as their unaffected siblings. Children with hydrocephalus also scored lower on perceptual-motor functioning than both their siblings and children with myelodysplasia without hydrocephalus. An inverse relationship existed between level of the lesion and intelligence (Tew & Laurence, 1974). In a study of 173 children, Soare and Raimondi (1978) reported that 63% with hydrocephalus had IQs over 80 whereas 87% without hydrocephalus had IQs over 80. Similar to Tew and Laurence (1974), Soare and Raimondi (1978) found there was no difference between children with myelodysplasia without hydrocephalus and their unaffected siblings. The child's IQ was also related to family income and education. The assumption of intellectual impairment as a consequence of hydrocephalus is probably no longer valid because of the practice of early shunting. Intellectual ability is more likely related to the success of the procedure so that children with repeated episodes of shunt dysfunction and/or infection are at greater risk for intellectual impairment.

A deceiving behavior accompanying the hydrocephalus is labeled the "cocktail party syndrome" because of the high level of vacuous verbal activity seen in these children. Unfortunately the high conversational ability of these children masks the inability to conceptualize; hence the behavior is labeled the "cocktail party syndrome." A consequence of this hyperverbal behavior is parental difficulty in accepting a low normal or below normal intelligence score because they mistake verbal ability for IQ. Therefore, due to the known higher incidence of cocktail syndrome and perceptual problems among children with myelodysplasia, psychometric testing is recommended by age four with comprehensive developmental testing prior to that time (Soare & Raimondi, 1977).

However, there is no consistency in the types of tests used to test intelligence and various quotients are used to designate normal vs below normal (Hunt & Holmes, 1975; Lorber & Zachery, 1968). Some authors have used the terms normal and subnormal without reference to intelligence quotients. In addition, researchers do not attend to etiological contributions to cognitive development such as brain structure, environmental experience and lack of exposure to normal educational settings

(Diller, Swinyard & Epstein, 1978). There is general consensus that as the level of the spinal defect ascends above the sacral level, the frequency of hydrocephalus, shunting and mortality increases and cognitive function decreases (Hunt & Holmes, 1975; Lorber & Zachery, 1968; Shurtleff, Foltz & Loeser, 1973; Soare & Raimondi, 1978, Spain, 1974; Tew, 1977).

One secondary manifestation of myelodysplasia is obesity which probably is related to both immobility and overfeeding by solicitous families. Another secondary manifestation is repeated bone fractures which result from both the immobility and lack of normal nerve innervation and blood supply. Skin breakdown also occurs due to inactivity, poor circulation, sensory loss below the level of the lesion, incontinence, casts and braces.

Later outcomes In process oriented research it is important to know long term outcomes of these children in order to identify variables for study from infancy. Self care is an important milestone for children with myelodysplasia. However, clinical observations of these children suggest progress toward self care and independence is influenced by several factors. First, logistics and the time required to teach the child to remove and replace braces and then dress over braces and the time for the child actually to perform these tasks result in parents assuming care responsibility beyond the preschool years. Second, often when a child achieves a stage of independence, a hospitalization followed by casting forces the child back to a more dependent role (Steele, 1977). Third, a parent's own need to care for the child may prolong dependence. Mothers are able to identify the child's need for self care but are unable to give this responsibility to the child (Wolfensberger &

Menalascine, 1970). Fourth, some children become so obese that they cannot assume self care (Hayes-Allen & Tregg, 1973).

In a 1973 study of 1172 families Swinyard, Shakuntala & Nishimura (1978), indicated that with extensive therapy and health care, children with myelodysplasia attain independence. Of the 1172 families, whose children had a mean age of 7.8 years, the children had had an average of 2.8 surgeries for shunt revisions, 7.3 kidney x-rays and 1.3 urinary tract infections. As a result of the orthopedic surgeries and physical therapy, 25% ambulated without braces, 43% ambulated with braces, and 31% were mobile in wheelchiars. The longterm outcome of independence is a serious question in children with myelodysplasia and progress toward self care needs to be assessed from infancy.

The outcome for the children born today may be different than for those of the 50's and 60's because of early intervention educational programs, attention to perceptual development from birth and advances in health care (Bates, West & Schmerl, 1977). However, these interventions from infancy need to be measured against their effect on the family system as several professionals enter the family system with each intervention for the child (Garbarino, 1977; Garbarino & Crouter, 1978).

Parent Outcomes

The outcomes observed in families of children with myelodysplasia are as important as the specific outcomes of the defect myelodysplasia with regard to the children. In the studies that have been conducted on family response to myelodysplasia, many variables were examined including immediate effect of the birth on the parents, financial stresses placed on the family, the effect of the handicapped child on siblings, the effect on the marriage and domestic routine, the effect on social life, the physical and mental health of the parents, family planning, and assistance (non-monetary) to the family from relatives and neighbors (Hide, Williams & Ellis, 1972). In each instance, the studies were univariate with the single independent variable being existence of a child with myelodysplasia in the home and each of the above factors as the dependent variable.

Initial response to the birth With the birth of any infant the family is altered both structurally and functionally. When a child is born with a defect, providing and securing continued care for the child, the prolonged physical dependence and uncertain future may keep the family in a low state of grief and crisis (Olshansky, 1962).

The parents' immediate reactions to the birth of a child with myelodsyplasia are similar to those of other parents of children with defects (D'Arcy, 1968). Most have never heard of the defect and, of those who had heard, few knew of the ways in which the child could be affected. Explanations given by health care personnel were poorly understood by families. Freeston (1971) reports of the 85 parents interviewed, only one quarter of the fathers and mothers felt they understood what was explained to them after the birth of the child. Mothers who were separated from their infants by transfers to medical centers felt especially anxious. Fathers reported difficulties immediately after birth in getting back to work and caring for other children while also trying to visit two hospitals and be supportive to their wives (Walker, Thomas & Russell, 1971).

The initial period following the birth of a child with a defect is critical in determining the effect of the birth on the family. Families need time to decide what to do after the birth of a child with a defect

(Kallop, 1973). Apley, Barbour and Westmacott (1967) found that a delay in the treatment of the defect resulted in over-anxiety and disturbance in family patterns in over 50% of the families studied. The information provided during the neonatal period had particular significance on the outcome of the infant. A hopeless prognosis is most frequently associated with the decision not to treat a child with myelodysplasia and the placement of the child outside of the home (Hayden, Shurtleff & Broy, 1974; Slobody & Scanlan, 1959; Zachary, 1971).

Later Outcomes Contradictory findings have been reported in the literature as to the effect of a child with myelodysplasia on the integrity of the marital relationship. Walker, Thomas and Russell reported in 1971 that of the 106 mothers and fathers interviewed, 65% thought their marriage was unchanged by the birth of the myelodysplastic child; 22% felt their marriage had deteriorated. In a study of 278 parents in Michigan, Feetham (1976) reported 23% indicated the child with meylodysplasia caused added strain to the marriage, 39% said no difference and 38% indicated they were closer together. Freeston (1971) documents the breakdown of three marriages in the 85 she studied, while Hare and colleagues (1966) noted that, in a great majority of cases, parents said the event had brought them closer together (Hare, Laurence, Payne & Rawnsley, 1966).

In contrast to the findings cited above, in a later ten year longitudinal study on the quality of marital relationships, Tew, Payne and Laurence (1974) reported significant deterioration in marital relationships of families of children with major neural tube malformations in England. Fifty-nine families of children with myelodysplasia and 58 matched control families were studied. Retrospective perceptions indicated that at the time of birth 70% of the index and control families had satisfactory relationships. At the eight to nine year follow-up, only 46% of the index families had satisfactory relationships while 79% of the controls did. This was a statistically significant difference. The divorce rate of the families with affected children was almost two times the national divorce rate, while the controls were below the national average. In 1977, reporting on 56 of the same 59 families, the number of divorces cited was six (10.7%) of which five occurred in the 10 families in which the child was conceived before the marriage. The reported conclusion was that the divorce rate in families of children with myelodysplasia is "nine times higher than for the local population" (Tew, Laurence, Payne, & Rawnsley, 1977). These reports are inconsistent and interpretations questionable. One point on which all the researchers agreed was that if a marriage was vulnerable before the birth of the handicapped child, it was more likely to break down than if it were not.

MacKeith (1973) notes that the response of parents of children with handicaps is derived from many factors including cultural and social-class attitudes to children in general, but most basically the response is premised on their feelings about having a child with a handicap. Trembath (1976) reports children with myelodysplasia have little adverse effect on family stability if the major factors of medical, social, educational and economic support are present.

The effect of a child with myelodysplasia on family planning and sexual relations has also been studied (Hare, Laurence, Payne & Rawnsley, 1966; Walker, Thomas & Russell, 1971). Freeston (1971) reports that fewer than half of the families she interviewed had sought family planning advice although the majority had decided against having other children. Fifty-nine of the 86 families studied by Richards had had no more children by the time their affected child was two to six years of age. Eleven of these mothers had been sterilized while 32 were employing no active measures to insure against pregnancy. Many of these parents described adverse effects on their sex lives because they did not want "another baby like" their child with myelodysplasia (Richard & McIntosh, 1973).

The variables of mental and physical health of the parents of children with myelodysplasia have also been studied. Walker, Thomas and Russell (1971) found that only a few of 106 mothers felt themselves to be "fit and well" while most were "tired, worried and depressed" or substantially worried by their total problem. Half the fathers, in comparison, felt fit and well.

In their ten year longitudinal study, Tew and Laurence (1975) measured the mental and physical health of the mothers by using Rutter's Malaise Inventory. The mothers were asked to answer by "yes" or "no" 24 questions referring either to emotional states or to physical states known to have psychological associations. The results indicated that the mothers of children with myelodysplasia had higher stress scores than the mothers of children with psychiatric problems, brain disorder, and physical handicaps whose scores were reported in other studies. Dorner (1975) reported half of the mothers in a sample of 37 families were on medications such as antidepressants. These findings are consistent with Travis' (1976) observation that over time mothers of children with chronic illness become exhausted due to additions in care brought on by the child.

The initial responses of parents to the birth of a child with a defect include physical symptoms such as anorexia and fatigue. The parents' sense of emotional well being is labile and based on the state of adjustment to the birth (Wolfensberger & Menalascino, 1970) and the status of the infant (Travis, 1976). Therefore the emotional state of the parent is perceived as a significant variable affecting family functioning.

Sibling Outcomes The presence of a handicapped child in a family affects all the members of the family, not only the parents. Several British workers have asked parents to describe the effect the child with myelodysplasia had on other children in the family. Twenty-one percent (17) families interviewed by Richards and McIntosh (1973) felt that the attention they could give the other children was diminished and, therefore, the myelodysplastic child's presence was detrimental to the siblings. Walker, Thomas and Russell (1971) reported a "marked reaction" of a sibling to the myelodysplastic child in 20 of 85 families. In these families the response to the child with the handicap by siblings was marked by excessive negativism or over solicitude.

Tew and Laurence (1973), in their longitudinal study of children with myelodysplasia and their families, attempted objective measures of adjustment of siblings. Fifty-nine children with myelodysplasia and their 44 siblings and 59 matched control children and their 63 siblings were studied. Assessing school behavior, the siblings of myelomeningocele children showed significantly more maladjustment than the control group. The maladjustment of siblings was greater when the child with myelodysplasia was more mildly handicapped. These findings are supported by Minde, Hackett, Killou and Silver (1972) in their study of siblings.

In the Tew and Laurence sample the child with myelodysplasia was treated as the youngest by all family members irrespective of birth order.

Family Environment

The family environment can be altered following the birth of a child with myelodysplasia. Ways in which family routine is disrupted, causing stresses to develop, have been reported in the British literature. These were changes in family outings, holidays and domestic routine (Richards & McIntosh, 1973; Schonell & Watts, 1956).

Whether parents receive help from friends, neighbors and relatives is often especially important to family functioning in families with children with handicaps. Sixty percent of parents interviewed by Richards and McIntosh (1973) thought that their neighbors and friends had been understanding and helpful after the birth of the affected child. Similarly, those parents studied by Walker, Thomas and Russell (1971) reported receiving help from neighbors in 25% of cases and assistance from grandparents in 86% of cases. The type of help sought by mothers include respite from the child to facilitate the mother's mobility for shopping and social activities.

The repeated clinical visits and hospitalizations also alter the family environment. Hospitalization of the child was identified as the single most disruptive family event (Freeston, 1971; Tew & Laurence, 1976; Walker, Thomas & Russell, 1971). The combination of special care, clinic visits and hospitalizations affects type of housing, maternal employment, school selection and type of family transportation, while desire for proximity to health care services affects the geographical selection of residence (Dunlap & Hollinsworth, 1977).

It is clear from the variables studied and reviewed here that the birth of a child with myelodysplasia affects the outcomes of the children themselves, the parents and the family environment. Some variables may affect the family more than others. Marital harmony, adjustment of siblings, and the physical and mental health of the parents may be crucial outcomes affected. All researchers agree that relationships vulnerable to stresses before the birth of a child with myelodysplasia are in greater peril than those relationships more stable before the birth.

DEPENDENT VARIABLE

Family Functioning

One approach to the study of family functioning is the structural-functional framework which views the family as a social system. Also referred to as functional analysis, the structural-functional framework for examining family functioning is derived from an overall functionalist theory of society, psychology and social anthropology (Eshelman, 1974). An advantage of the structural-functional framework is that it serves to explain the parts, the relationship between the parts, the relationship between the parts and the whole and the functions that are performed by, or result from, the relationship formed by the parts (McIntyre, 1966). A second advantage of the structural-functional framework is that other frameworks, such as family development and family ecology, can be used to augment the framework and expand the parameters of study (Aldous, Osmond, & Hicks, 1979; Bell & Vogel, 1968; McIntyre, 1966). A third advantage is that the structural-functional approach has been effectively applied to the study of the family at several levels from broad macroanalysis to intensive microanalysis.

Although the structural-functional framework has only been used in a small proportion of empirical research related to the family, it has had a major effect on family studies. One reason for this major effect is that researchers refer to the importance of family functions even if they do not employ deliberate use of the structural-functional framework (McIntyre, 1966). A second reason is that researchers integrate other frameworks into the structural-functional framework (Bell & Vogel, 1968; Levy, 1949). The application of the structural-functional framework has served to make family study an integral part of the study of the larger society.

Another reason which makes the concepts of the structural-functional framework applicable to a variety of research studies is that there is no single interpretation of family functions. Function may be defined in terms of outcomes, process, and content. Using the concepts of process, outcome and content, three major areas of functions as relationships have been emphasized when applying the structural-functional approach to the study of the family. The areas of family functions when viewed as relationships are: (1) the relationship between the family and broader social units; (2) the relationships between the family and subsystems; and (3) the relationships between the family and the individual (McIntyre, 1966; Sprey, 1973). Review of scholars' definitions of family functions presents the range of functions from which the specific family functions used in this study were drawn.

Functions of the family are seen as patterns of relationships among and between the people and the environment. These functions can be thought of as activities essential to the survival of the family. These functions may be generalized to include tasks such as procreation, socialization, protection, education, and economic concern (Eshelman, 1974). In contrast, Frankena (1970) established a normative set of family functions that are 1) making the lives of each member as good as possible and at the same time, dissuading members from seeking their own good at the expense of others, 2) transmitting patterns of living from one generation to the next generation (education), 3) regulating behavior through discipline based on education not restraint, 4) assisting with achievement of identity for each member, 5) providing a center of leisure for the pursuit of goals, 6) providing a seat of love and emotional gratification, and 7) helping each member build a way of thinking about life, the world, humans, and the universe.

Some researchers (Bell and Vogel, 1968) base their descriptions of family functions on the systems model of family exchange using outcome measures. In their model, Bell and Vogel (1968) suggest that the nuclear family trades 1) labor for wages, 2) family assets for goods with the economy, 3) loyalty for leadership and compliance, 4) participation for support with the community, 5) adherence for identity with the community, and 6) values and conformity for approval. From this Bell and Vogel (1968) project the following family functions: 1) preparation of goods from the external system for family use, 2) care and maintenance of family possessions, and 3) care of dependent members which includes the socialization process that distributes tasks within the family.

On the other hand, Duvall (1971) focuses on the two adult partners and their responsibilities. They are supposed to 1) maintain marital ties, 2) maintain effective communication, 3) share in responsibilities, 4) foster the development of the members, 5) provide a safe environment, and 6) protect against the unexpected.

Smilkstein (1978), a physician, identifies five family functions from common themes in the social science literature. The five functions of adaptation, partnership, growth, affection and resolve were empirically derived to create a family function paradigm analogous to the body organ systems. The paradigm acknowledges both a unique component of each function and its interrelationship to the whole. The family functions presented by Gersten (1976) also imply interrelationships between both the individuals and the family and the individuals and the environment. Gersten (1976) identifies that family functions are achieved through a broad range of family behaviors inclusive of: the fulfillment of role functions by family members, the qualities and satisfactions with the marriage, and family communication patterns and processes. Those families which function effectively are seen by Duvall (1971) as successful in narrowing the discrepancies between what might be achieved and what is achieved.

The initial discrepancy between the expected birth of a normal child and the birth of a child with a defect precipitates a crisis which disrupts family functioning (Price-Bonham & Addison, 1978; Steele, 1977). When the child continues to be handicapped or chronically ill the family becomes chronically stressed (Martin, H., 1975; Martin, P., 1975; Olshansky, 1962; Waechter, 1977). Family functions become altered in response to this stress in the areas of 1) relationships between the family and broader social units (Tew & Laurence, 1973), 2) the relationships between the family and subsystems (Farber, 1959; Rodgers, 1973; Travis, 1976), and 3) the relationships between the family and the individual (Dorner, 1975; Klein, 1976; Tew, Payne & Laurence, 1974). Family functioning, as the dependent variable for this study, is conceptualized as a composite of the family functions reviewed previously and categorized in the three major areas of relationships.

Major Areas of Family Functioning

Relationships between the family and broader social units

Alterations in the areas of family function of relationships between the family and broader social units seem to emerge from stigma and the limited mobility of the child. Some authors suggest there is a restriction in the range of relationships and activities in families with children with handicaps in response to the stigma of the child, countered by the need to maintain appearances of normalcy to the broader social systems (Travis, 1976; Waechter, 1977). That is, relationships with the extended family, friends and neighbors are altered by the perceived acceptance of the child by the parents (Waechter, 1977). Therefore, social isolation is interpreted by these authors as self-imposed by the family to avoid a perceived stigma.

Altered activity patterns are also suggested as a result of the constraints of the handicap (Dorner, 1971; Freeston, 1971; Walker, Thomas & Russell, 1971). In 41 of 94 couples who had not been out socially since the birth of their child, the former group attributed the change in their activities to the extra demands of the care of the child with the handicap (Walker, Thomas & Russell, 1971).

The majority of mothers interviewed by Walker and colleagues felt "isolated" as a result of their affected child despite family support (Walker, Thomas & Russell, 1971). In addition, substantial anxiety over leaving the baby with someone else was expressed by two-thirds of the couples. When asked for reasons for joining a Spina Bifida parent group, 15% of 278 parents cited reduction of isolation as their motivation (Feetham, 1976). In Dorner's (1975) study the parents attributed their social isolation to their child's immobility. In a study of 400 rural Alabama families, Dunlap and Hollinsworth (1977) also reported a reduction in family activities occurring primarily when the child had a severe physical impairment. Handicapped children without mobility problems were not perceived as changing family relationships.

In contrast to the effect of limiting social contacts, the care of a child with myelodysplasia opens the family system to frequent relationships with health professionals. During the neonatal period the family may have contact with over 20 health professionals. Once discharge from the hospital occurs, after the neonatal period, the average number of clinic appointments for myelodysplasia related care is five times during the first year. While parents report satisfaction with the quality of care their child received, they do not find the average health professional supportive (Dorner, 1975; Freeston, 1971; Minde, Hacket, Killou & Silver, 1972). The number of professionals, the unfamiliar terminology and not knowing what is expected, while not feeling comfortable in asking, causes disruption for many parents. In a study by Walker and colleagues (1971), parents reported professionals knew less than they did and therefore tended to be of little help (Walker, Thomas & Russell, 1971).

The frequency of hospitalizations also affects the families' relationships to the broader social units. Parents perceive hospitalizations as the single most disruptive event (Dorner, 1975; Freeston, 1971). Walker, Thomas & Russell (1971) reported the mean number of hospitalizations by age three to be 2.9 in a British sample, In Freeston's (1971) American sample, by age four the mean number of hospital admissions was 6.

The presence of a child with a handicap may also affect where the family lives, the type of housing and accessibility to care and educational facilities for the child. All of these factors can alter the family's relationships with the broader society (Dunlap & Hollinsworth, 1977).

Relationship between the family and subsystems The second major area of family functioning, i.e., relationships between the family and subsystems, can also be altered following the birth of a child with myelodysplasia. These family functions include the completion of household tasks with or without the help of relatives and the spouse, emotional support from the extended family and overall satisfaction with the marriage.

In a study of 107 British families of children with myelodysplasia, 58% of the mothers reported their domestic routine as normal (Walker, Thomas & Russell, 1971). However, 78% of the mothers, who also had normal children, reported the child with myelodysplasia more difficult to raise. In the same study, only 31% of the fathers participated in the ongoing care of the child. However, the families with children with myelodysplasia did receive help and support from the extended family (Klein, 1976; Travis, 1976). Walker, Thomas & Russell (1971) reported the maternal grandmother as most supportive and 26% of the families also received help from the neighbors. In regard to family and child care tasks, Travis (1976) reported the mothers tended to assume the additional responsibilities while the fathers were more apt to abdicate their role. In addition, older siblings either were over solicitous to the child with myelodysplasia or showed excessive negativism (Walker, Thomas & Russell, 1971).

The overall quality and satisfaction with the marriage of parents of children with myelodysplasia has been studied through reports of marital breakdown. There are no consistent patterns reported from the studies. In some British studies, the researchers reported the divorce rate to be no higher than the national norm, while one British and two American researchers reported a significant increase in the divorce rate over matched controls (Freeston, 1971; Hare, Laurence, Payne & Rawnsley, 1966; Kolin, Scherzer, New & Garfield, 1971; Richards & McIntosh, 1973).

Relationships between the family and the individual   The third major area of family functions is relationships between the family and the individual. Specific functions within this area include discussion, disagreements and time with one's spouse, problems and time with children other than the child with myelodysplasia, and satisfaction with sexual relationships.

In their study of 400 rural Alabama families, Dunlap & Hollinsworth (1977) reported that 91% of the mothers indicated the handicapped child had no effect on either the marriage or intrafamily relationships. Of the 9% indicating the child had an effect on the marriage and family, 42% reported a positive effect. In addition, 95% of the families reported the handicapped child got along well with all immediate and extended family members (Dunlap & Hollinsworth, 1977).

The times of the greatest effect of the child with a handicap on the relationships with the family are seen as the initial period following the birth or diagnosis and the first anniversary period (Kallop, 1975; Mercer, 1974; Wolfensberger & Menalascino, 1970). Separation from the infant and the response to the birth both interfere with the normal development of parent-infant relationships. The initial period of multiple decisions and unknown outcomes of the infant add to the altered intrafamily relationships. Clinical observations of these families over time suggest that the parent's perception of the neonatal period influences their relationships with the infant, other family members and health professionals. Hayden, Shurtleff, and Broy (1974) found placement outside the home to be highly correlated with an initial hopeless prognosis.

Summary

That some families of children with myelodysplasia and other handicaps can function effectively and adapt to the care of the child while other families experience dysfunction in one or more of the three major areas of family function is evident from the literature. Travis (1976) and Minde (1978) found that the modifications in family functioning caused by the presence of a child with a handicap are stabilized by the time the child reaches age five. What is important is whether the modifications are positive in relation to expectations of the family functioning. While examples of the effect of the presence of the child with myelodysplasia on specific family functions are reported throughout the literature, data on the interrelationships among parent and infant outcomes and the family environment to the family functioning do not exist.

CHAPTER III

METHODOLOGY

Design

The purpose of this descriptive longitudinal study was to examine the interrelationships among infant, parent, and family environment variables to account for the amount of variance in family functioning. The study design is presented in Figure 1. This study was also exploratory because questions were raised regarding relationships not studied before or not studied with this population. For example, Garbarino (1977) and Sims, Paolucci & Morris (1972) used designs to study the interrelationships of many variables in the problems of child abuse and malnutrition, but this design is new in the study of children with myelodysplasia and their families.

The research hypotheses postulate relationships between 1) infant outcome, 2) parent outcome, and 3) family environment and the discrepancy between expected and actual family functioning through 18 months following the birth of an infant with myelodysplasia. The dependent variable, family functioning, is a composite score of family functioning across 21 indicators. The independent variables are composite scores of infant outcomes, parent outcomes and the family environment as measured by specified items from the Parent Survey (PS), Profile of Mood States (POMS), and infant hospital records and developmental testing.

Subjects

The subjects for this descriptive longitudinal study were parents and their infants with myelodsyplasia admitted to a Myelodysplasia Care Center in a large urban pediatric hospital. Forty-six infants with

TIME PERIOD	INFANT	MOTHER AND FATHER
Neonatal Period	Medical History Physical Examination* Echoencephalogram/ ventriculogram	Informed Consent Profile of Mood States (POMS)
Three Months	Health History and Physical Examination* Early Intervention Developmental Profile (EIDP) Shunt Evaluation**	Parent Survey #2 Parent Status Family Environment Family Functioning Prenatal - Form A Parent Status Family Environment Family Functioning Perinatal Perceptions Profile of Mood States (POMS)
Six Months	Health History and Physical Examination EIDP	Parent Survey #3, 4, 5 Parent Status Family Environment Family Functioning Profile of Mood States (POMS)
Twelve Months	Health History and Physical Examination EIDP	Parent Survey #3, 4, 5 Parent Status Family Environment Family Functioning Profile of Mood States (POMS)
Eighteen Months	Health History and Physical Examination EIDP	Parent Survey #3, 4, 5 Parent Status Family Environment Family Functioning Profile of Mood States (POMS)

*Physical examination includes complete neurological examination.

**Radiographic evaluation of shunt function is performed as necessary at any visit.

Figure 1 Study Design

myelodysplasia were admitted to the care center during the time of the study from March 1977 to July 1979. These infants represent 40% of the live born infants with myelodysplasia in Michigan in this time period. Criteria for admission of a family to the study were: (1) the infant with myelodysplasia was admitted to the myelodysplasia care center within the first six months following the birth; (2) the infant received medical and/or surgical treatment of the defect; (3) at least one parent was willing to participate in the study; (4) there was continued involvement of the biological parent(s) with the infant; and (5) survival of the infant was anticipated.

A total of 38 infants and 66 parents (38 mothers and 28 fathers) comprise the study sample. Eight families were omitted from the study for the following reasons: both of the parents of three infants refused to participate and three parents were not approached on the recommendations of the myelodysplasia center social worker. The reasons given by the social worker for not approaching these three families were because they were non-English speaking; there was severe mental illness in the mother and/or early death of an infant in an already dysfunctional family was anticipated. Two additional families were omitted as the infants were removed from the care of the biological parents.

The myelodysplasia care center receives referrals on a statewide basis. The infants and parents in this study came from six counties in southeastern and central Michigan. The comparison of characteristics of the infant sample with those of the population as reported in the literature are shown in Table 1. A review of general characteristics is shown in Table 2.

Table 1

Charactistics of Children with

Myelodysplasia Reported in the Literature*

Compared with Those of the Study Sample (N=38)

Characteristic	Previous Reported Samples	Study Sample
Majority of Conceptions	Spring-Summer	October-March (75%)
Sex ratio: Females:Males	3:2	1:1
Lesion at or below L_1	80%	91%
Presence of hydrocephalus	75-80%	81%
Pregnancy resulting in infant	1st	1st (23%)

*Dekaban, A., 1972; McLaughlin & Shurtleff, 1979

Table 2

Study Infant Characteristics (N=38)

Characteristic		Frequency	Percentage
Race:	Caucasion	28	74%
	Black	8	21%
	Hispanic	2	5%
First Hospitalization <u>M</u>		13 days	
Birth weight <u>M</u>		3231.706 gms.	
Head Circumference Percentile*			
	> 90%	16	47%
	normal	12	35%
	< 90%	6	18%+

* 81% were treated for hydrocephalus based on clinical evidence

\+ included preterm and small for date infants

All parents (mothers and fathers) of the infants admitted to the myelodysplasia care center were asked to participate in the study unless the center social worker advised against the contact. A total of 66 parents, 38 mothers and 28 fathers, participated in data collection at least once during the time periods. The demographic characteristics of the mothers are listed in Table 3. Marriage was not a criteria for participation; if both parents were available although not married, both the mother and father were asked to participate.

Table 3

Mothers' Characteristics (N=38)

Characteristic	Frequency	Percentage
Marital Status:		
Married	29	75
Single	4	13
Single/living with father	3	8
Divorced/Separated	2	4
Education level:		
< High School	5	13
High School	22	58
1-3 Years Post High School	8	21
Bachelors Degree	3	8
Maternal Age at Birth:		
< 18 years	4	10
19-28 years	23	61
29-34 years	9	24
35 + years	2	5
Pregnancy Planned:		
Yes	19	50
No	19	50

Instruments

The infant data are from two primary sources: the hospital records (using chart reviews) and developmental testing, using the Early Intervention Developmental Profile (D'Eugenio & Rogers, 1975).

The selection of instruments for infant testing is a significant methodological issue. Medical technology can clarify morphological aspects of brain damage in children with hydrocephalus, but psychometric problems persist. The infant is too young for accurate psychometric evaluation. The common infant development instruments such as the Gesell or the Bayley Scales of Infant Development are based on sensorimotor development rather than verbal and reasoning skills and are recognized as poor predictors for intelligence (Illingworth, 1971). Tests measuring changes with experience and evolving with age are used reliably only after three years of age. The consequences are that there is a lag between the time of medical assessment of the possible cerebral damage resulting from the myelodysplasia and its associated problems and the ability to assess cognitive function. Also, based on a review of developmental test references, it is clear that instruments that have strong measures of reliability and validity do not exist for assessing children with developmental disabilities. In addition, existing comprehensive tests of infant development are not designed to provide parents with specific information about their child's development as it relates to the parents' activities with the infant. In light of these considerations, the Early Intervention Developmental Profile (EIDP), developed by the Institute for the Study of Mental Retardation and Related Disabilities (ISMRRD) in Ann Arbor, Michigan, was selected for this study. (See Appendix A.1 for instrument.)

Early Intervention Developmental Profile   The EIDP provides more infant data than do screening tests and does not involve as much time as the Bayley and Gesell instruments, although it comprises many items from these instruments (Honzik, 1976). The advantages of this instrument are that it is designed for the assessment of infants with developmental disabilities and is combined with a developmental program of intervention activities. As a result, following the testing procedure, parents can be given specific activities related to their infant to support their continued development rather than vague ranges of development lacking predictive ability (D'Eugenio & Rogers, 1975).

The EIDP is a compilation of major developmental milestones from birth to 36 months of age covering six areas of development. These six areas are perceptual/fine motor, cognition, language, social/emotional, self-care and gross motor. Although testing procedures are included in the EIDP manual, they are not standardized because of the need to modify testing procedures for infants with developmental disabilities. Because of the lack of standardization and age norms, the EIDP is not recommended for diagnosis but is used for obtaining a developmental age estimate.

Professionals from the ISMRDD assessed the concurrent validity of the EIDP by correlating each of the six profile scales with standardized widely used evaluation instruments on 14 children with handicaps. The correlation coefficients for all scales ranged from a low of .33 between the profile gross motor and Receptive Expressive Emergent Language (REEL) scale and a high of .96 between the profile social scale and cognitive scale and the Bayley Mental Scale. The profile gross motor scale was correlated with both the clinical motor evaluations and the Bayley motor scale with the resulting correlations of .95 and .84 respectively (Rogers & D'Eugenio, 1977). Patnales (1977) compared the EIDP to the Bayley, using a group of normal children, showing a .88 to .98 correlational coefficient between the scales.

Interrater reliability was examined by the ISMRRD team by using a tester observer paradigm. The percent of agreement between tester and observers ranged from 80% to 99% with a mean of 89%. Test retest correlation coefficients on 15 children at three month intervals ranged from .93 on language at three and six months to .98 on social-emotional measures at three months and six months with all correlations significant at the $\underline{p} < .01$ levels (Rogers & D'Eugenio, 1977).

Hospital Records All other infant data collected from a review of the hospital records were determined by a pediatrician and the researcher (a nurse clinical specialist) with a combined experience of over 40 years in the care of children with myelodysplasia. In addition, current research on the short and long term outcomes of these children were reviewed to assure inclusion of all pertinent data (see Appendix A.2 and A.3 for infant data sheets).

Physical examinations, including a complete neurological and developmental assessment, were completed on the infants in the neonatal period, and at 3, 6, 12 and 18 months. Throughout the study infants were not subjected to any procedure and/or examination other than those routinely experienced by all infants treated in the myelodysplasia care center.

The neonatal examination delineated the extent of spinal dysraphism, segmental level of motor and sensory denervation, associated musculoskeletal defects e.g., hip dislocation and club feet, bladder and anal sphincter incompetence and the presence of hydrocephalus (by appearance, head circumference and transillumination). In addition to the physical examination, echoencephalography and/or ventriculography were done in the neonatal period to quantitate ventricular size and brain mantle thickness. A formula published by Shurtleff, Foltz & Loeser (1973) was applied to calculate brain mass in grams which was compared to the brain mass expected for age. Adequate brain mass is a factor in deciding therapy, as Shurtleff's patients with less than 60% normal brain mass preoperatively were all mentally retarded (Shurtleff, Foltz & Loeser, 1973).

Follow-up physical examination at 3, 6, 12 and 18 months included assessment of vision and hearing, motor and sensory function and serial echoencephalograms or computerized tomographic (CT) scans as necessary, to determine the degree of hydrocephalus and need for shunt placement or shunt revision. An interval health history also conducted at the follow-up periods included the incidence of acute illnesses and care problems, nutritional history, infant behaviors (such as sleep and socialization), interventions received (such as primary care, immunizations and education), the family's response and coping in relation to the infant's birth, and changes in the family environment. All data were recorded on the hospital records by a pediatrician or nurse clinical specialist for later retrieval and analysis. In addition, prenatal, perinatal history and treatment data not obtained in the follow-up history were also taken from the hospital records.

For analysis the individual infant variables were combined into three major composite scores based on the clinical judgment of the investigator and correlates of infant outcomes as identified in the literature

(Diller, Swinyard & Epstein, 1978; Laurence, 1976; Lorber, 1971; Richards & McIntosh, 1973; Soare & Raimondi, 1978). The neonatal and infant follow-up composite scores included data on general health and developmental status, central nervous system status, and intervention received. The infant follow-up data were collected at 3, 6, 12 and 18 months. The infant variables used for the infant composite scores are presented in Figure 2.

Neonatal Infant Score*	Infant Followup+
General Status	
Apgar scores	care problems
associated anomalies total	developmental age++
birth weight percentile	developmental delay++
gestational age	mobility
	weight percentile
Central Nervous System	
brain mass	head circumference percentile
head circumference percentile	level of motor functioning
insertion of ventricular shunt	myelodysplasia related problems
level of motor functioning	shunt status
presence of hydrocephalus	
type of defect	
Intervention	
complications	acute illness
disposition	days in hospital
length of hospitalization	disposition
medical treatment	emergency room visits
number of referrals	hospitalizations
surgical treatment	number of clinic appointments
	number of surgeries
	supportive treatment
	well child care

Data source: *Infant neonatal records
+Infants hospital records at 3, 6, 12 and 18 months
++EIDP (Early Intervention Developmental Profile) at 3, 6, 12 and 18 months

Figure 2 Independent Variables Forming Infant Composite Scores

Parents

Parent Survey Parent data used for both the independent and dependent variables were obtained from a self-administered questionnaire, the Parent Survey. The Parent Survey (PS), developed by Feetham and Perrin (1977), gathered longitudinal data on a number of facets of parental experiences prior to and following the birth of a child with myelodysplasia (see Appendix A.4 for Parent Survey). At 3, 6, 12 and 18 months the survey gathered identical data related to (1) general parent status including education, marital status, and perception of physical and mental health, and (2) parent life experiences including the incidence of home relocations and hospitalization, illness and/or the death of friends, relatives, spouse or children. Family environment data collected at these time periods included perception of financial concerns, access to transportation, numbers of persons entering the family system because of the infant with myelodysplasia, a rating of the home environment, and a measure of how and what time was spent with the infant with myelodysplasia. The variables comprising the parent and family environment scores are listed in Figure 3.

In addition to the Parent Survey, at the 3 month time interval, the parents were asked to complete a questionnaire which included items identical to the Parent Survey but were recalled from the time prior to the infant's birth. The parents were also asked their perception of the prenatal and neonatal period. Although the limitations of recall data were recognized, it was not considered appropriate to subject the parents to a lengthy questionnaire during the neonatal period; knowing the confusion and disorientation occurring with the birth of a child with a defect (see Appendix A.5 for Form A - prenatal and neonatal parent data).

- Parent-Family Environment*
    - educational level
    - marital status
    - perception of mental health
    - perception of physical health
  - Life Experiences
    - death(s) of significant others
    - hospitalization of significant others
    - number of household moves
  - Family Environment Score
    - accessible transportation
    - distance to relatives
    - effect of infant care on finances
    - home environment rating (space, heat, food, safety, clothing)
    - how spend time away from family
    - number of persons living in home
    - number of professionals in home
    - number of professionals seen
    - time spent in activities related to infant
- Perinatal Family Score°
    - family history of neural tube anomalies
    - history of fetal loss
    - maternal age
    - number of pregnancies
  - Prenatal**
    - effect of pregnancy on finances
    - perception of pregnancy
    - prenatal perception of health
    - pregnancy planned
  - Neonatal**
    - distance traveled to see infant
    - expectations of neonatal period
    - expected contact with the infant
    - expected placement of infant
    - information on myelodysplasia
    - information on parent group
    - perception of infant's condition
    - perception of own health in neonatal period

Data Source: *Parent Survey at 3, 6, 12, 18 months
°Infant neonatal record
**Parent Survey Form A at 3 months

Figure 3 Independent Variables Forming Parent Composite Scores

The Parent Survey was reviewed for face validity by experts in either the care of children with myelodysplasia or family theory. In addition, drafts of the instrument were pretested with several parents of children with myelodysplasia whose children were beyond the age included in the study. The instruments were revised following input from all of these sources.

Family Function Survey The dependent variable, family functioning, was measured from 21 items included in the Parent Survey. The 21 family function items were developed for this study to measure the three major areas of family functions: (1) the relationship between the family and broader social units, (2) the relationships between the family and subsystems and (3) the relationships between the family and the individual (McIntyre, 1966). The specific function indicators for family function score are listed in Figure 4.

---

disagreements with spouse
discussion of concerns and problems with spouse
emotional support from friends and relatives
emotional support from spouse
help from friends
help from relatives
help from spouse
problems with other children
satisfaction with marriage
satisfaction with sexual relations
talk with friends and relatives
time in household tasks
time in leisure recreational activities
time miss housework
time other children miss school
time spent with spouse
time spouse misses work
time with children
time with health professionals
time with neighbors
time you are ill

---

*Data source: Family Functioning Survey

Figure 4 Family Functioning Indicators* for Dependent Variable

The specific function indicators were derived from review of the family functioning literature (Bell & Vogel, 1968; Duvall, 1971; Eshelman, 1975; Rodgers, 1973) and from the author's 18 years of clinical observations of family functions affected by the birth of an infant with myelodysplasia. The instrument is distinct from other family functioning instruments as it addresses each of the three areas of family functioning and can be used with one or both parents. The instrument includes interaction with friends, neighbors and relatives as this area of family functioning is known to be altered in families with children with handicaps (Tew & Laurence, 1973; 1974; Travis, 1976). Another important aspect of this instrument is that the scoring allows for the computation of a discrepant score across all items which is the measure of hypothesis III. Hypothesis III postulates that family functioning discrepant scores as measured by parents' reports will be greater at the first year anniversary of the birth of the infant with myelodysplasia than at 3, 6 and 18 months following the birth.

The measures of family function were obtained using the Porter format (Porter, 1962; 1963a; 1963b). The Porter format was designed in the early sixties to determine workers' perception of their work situation/environment. The original scale was purported to measure the magnitude, importance, and degrees of need satisfaction of managers in relation to Maslow's (1954) hierarchy of need. The scale was designed to measure not only the existing degree of need fulfillment but also the discrepancy between achieved and expected levels in addition to the relative importance of each category (Porter, 1962; 1963a; 1963b).

While some interpreted the Porter job items as measures of goal attainment (Evans, 1969; Haire, Ghiselli & Porter, 1963), later researchers using the Porter format indicate the stem questions determine the variable measured and that the format is adaptable to many conceptual frameworks (Shea, Werley, Rosen & Ager, 1973; Wernimont, Toren & Kappell, 1970). In addition to the management studies, the Porter format was used in a nationwide survey of faculty and students in health professions in relation to knowledge of family planning (Werley, Ager, Rosen & Shea, 1973), and in a survey of 278 parents regarding expectations of parent groups (Feetham, 1976a; 1976b).

The Porter format allows for the computation of a discrepant score for each item indicating perceived expectation for a particular item. Referring to the sample item in Figure 5, the deficiency score represents the difference between ratings on subscales a and b. When the importance score (part c of each item) is used with the discrepant score both direction and degree of dissatisfaction with the perceived existing situation are measured. The importance item (subscale c) can be used as an indication for intervention if the respondant has a high discrepant score (a-b) and high importance score (c).

"Amount of time you spend with your other children."

a) How much is there now? Little 1 2 3 4 5 6 7 Much

b) How much should there be? Little 1 2 3 4 5 6 7 Much

c) How important is this to me? Little 1 2 3 4 5 6 7 Much

{discrepant score calculated by (a-b)}

Figure 5 Sample Question Format Using Porter Scale

The Porter technique is an indirect measure derived from two direct measures by the respondent. According to Porter (1962) this method has two advantages. First, the tendency for a simple response set is reduced as it is more difficult for the respondent to manipulate the expectation of what is to conform to a socially desirable type of response. This multiple response set also controls for cultural and ethnic diversity as only the discrepant score between the respondent's perception of what is and should be is used. This is in contrast to single scale instruments which rely on the researchers' perceived value rating of the "what is" item, i.e., item a (Shaw & Wright, 1967). In addition, this method of measuring expectation or need is considered more conservative than a single question and is a realistic measure in comparing different groups (Porter, 1962). The second advantage is that an indirect measure of satisfaction with the activity inherent in the item is obtained (Porter, 1962).

A discrepant score is calculated for each family function item by calculating the difference between the a and b score. Each score is converted to an absolute score as the amount of difference between a and b scores is of significance rather than the direction of the difference. The discrepant score is the amount of agreement between the amount of the activity and the desired amount of the activity. That is, when the amount of the activity is the same as the desired amount, the difference is 0. The discrepancy between the amount of reported activity and the amount desired implies the degree of dissatisfaction (Porter, 1962; 1963a). Therefore, the scores closest to 0 indicate the greatest degree of satisfaction with family functioning and those farthest from 0 indicate family functioning is not what it should be as perceived by the respondents. The possible range of total discrepant scores for all the 21 family functioning items is 0 to 126.

Some content validity of the family functioning survey is evidenced by the significant Pearson correlation coefficients between the Family Functioning Index (FFI) by Pless and Satterwhite (1973) and the Family Functioning Survey used in this study. The Pearson correlations were computed on data from 103 mothers of school children with myelodysplasia throughout Michigan ($r = .54$, $p < 0.001$). The FFI was developed by Pless and colleagues (1972; 1973; 1976) to assess the functioning of families of chronically ill children for the purpose of identifying those children at risk (Pless, Roghmann & Haggerty, 1972; Pless & Satterwhite, 1973; Satterwhite, Zweig, Iker & Pless, 1976). Two types of validation studies, content and construct, have been completed across two samples. In addition, the FFI has evidence of test-retest reliability, and a correlation of .72 between the scores of husbands and wives obtained independently (Satterwhite, Zweig, Iker & Pless, 1976).

Reliability of the Family Functioning Survey (FFS) was assessed using the alpha reliability coefficient and the coefficient of stability after two weeks using the same sample of 103 mothers in Michigan. The alpha reliability coefficient is 0.72 on 103 subjects and the coefficient of stability after two weeks is 0.85 on 22 subjects.

Profile of Mood States The instrument selected to measure the independent variable of emotional response to the birth of the infant with myelodysplasia is the Profile of Mood States (POMS) developed by McNair, Lorr and Droppleman (1971). The POMS is a rapid, economical method of identifying and assessing transient, fluctuating affective states. The POMS, a 65 item factor analytically derived inventory, is a measure of six identifiable mood or affective states: tension-anxiety; depression-rejection; anger-hostility; vigor-activity; fatigue-inertia; confusion-bewilderment. Each of the 65 items is scored on a five point 0-4 scale. (See Appendix A.7 for instrument). The POMS has proved to be a sensitive measure of mood changes over time on psychiatric populations and normal subjects in both experimental and natural situations (Goldberg, 1974; Greenberg, Pillard & Pearlman, 1972; McNair & Lorr, 1964). The same six mood factors are identified, measured reliably and replicated in the research populations whether the rating period is the immediate present or spans a one week period. The POMS was selected for this study as it measures mood states identified by researchers as occurring in parents in response to the birth of a child with a defect. These mood responses include anxiety, depression, anger, fatigue and confusion (Kallop, 1973, Waechter, 1977; Wolfensberger & Menalascino, 1970). According to Mercer (1974) the parents pass through several stages of behavior and mood states in the year following the birth of an infant with a defect, therefore, an instrument measuring fluctuating affective states provides necessary data related to family functioning.

The POMS is recommended primarily as a measure of mood states in psychiatric outpatients and as a method for assessing change in such patients. However, it is also recommended for similar purposes on a research basis, for subjects 18 years and older who have had some high school education (McNair, Lorr & Droppleman, 1971). In addition to the six subscores, a total mood disturbance score is obtained by summing the six scores with vigor weighted negatively. The total mood disturbance score is used, as in this study, when a single global estimate of affective state is desired.

Use of the POMS showed significant difference in anxiety scores in the same groups over time, when the subjects were exposed to an anxiety provoking experience (Greenberg, Pillard & Pearlman, 1972). There were also significant differences in a small sample of 14 subjects associated with sleep deprivation (Hord, Tracy, Lubin & Johnson, 1975). The POMS was used in several drug related studies on psychiatric patients to test the effects of placebos, nothing and prescribed drugs (Landauer, 1975; Lorr, McNair & Weinsteen, 1964; Raskin & Crook, 1976). The POMS was also used on 36 subjects as a measure of diurinal variations in mood as asserted by self report and verbal content (Taub & Berber, 1974). When a short form of the POMS was used in a study of personality differences between inflammatory bowel disease patients and their healthy siblings, there was no significant difference between the two groups. However, in the same study there was a significant difference between the two groups using a longer test such as the MMPI (McMahon, Schmitt, Patterson & Rothman, 1973).

The POMS has two particular advantages: it is designed to be self-administered to persons with at least a seventh grade education, and it includes a time reference in contrast to many standard personality inventories. By being time referenced the researcher is able to determine whether an enduring personality trait is being measured or the desired mood state (McNair, Lorr & Droppleman, 1971).

Extensive reliability and validity testing have been completed on the POMS. For internal consistency the homogeneity of the six replicated POMS factor scores are .87 and above on the 350 male and 650 female psychiatric outpatients. However, the test-retest reliability coefficients are lower (.65 to .70) on 250 psychiatric outpatients. The lower coefficients are considered to be a result of the construct validity of the instrument and the ability to measure mood changes.

Factorial validity is evidenced by the six factor analytic replications completed in the development of the POMS as the results were congruent for the different patient and normal samples (McNair, Lorr & Droppleman, 1971). Studies with psychiatric patients receiving therapy versus those not on treatment suggest the scores are not a function of repeated testing but are related to drug and psychiatric treatment (Lorr, McNair & Weinsteen, 1964; McNair, Fisher, Sussman, Droppleman & Kahn, 1970).

For concurrent validity the POMS scores were correlated with the Hopkins Symptom Distress Scale (Parloff, Kelman & Frank, 1954), the Taylor Manifest Anxiety Scale (MAS) and the Interpersonal Behavior Inventory (IBI) using samples of 200 to 1,000 subjects. All correlations were significant at the $p \leq .05$ or $p \leq .01$ levels. On the symptom distress scales the variance shared by the mood and symptom measures ranged from 5% to 73%. There is a limited effect of demographic data such as sex, education, age, and race on the POMS scores as no more than 5% of the variance is accounted for by these variables (McNair, Lorr & Droppleman, 1971). Also measures of social desirability were found to have low to moderate correlations with the POMS scores of 150 patients. With the exception of the anger subscore ($r = .52$) the mood scores are independent of role playing and measures of defensiveness or lying (Wiggins, 1964).

The parent instruments were organized in the following sequence: demographic, Parent Survey, and at three months, Form A of the Parent Survey (prenatal and neonatal data), and the POMS. The rationale for the sequencing of the instruments was to present material apt to be perceived as most pertinent prior to that probably perceived as least relevant to the respondent.

Data Collection Procedures

Data were collected from infants and their parents at five time intervals: the neonatal period, 3, 6, 12 and 18 months postpartum. Because of the known perceptual distortions, confusion and shock experienced by the parents following the birth of an infant with a defect, only the POMS was administered during the neonatal period (Wolfensberger & Menalascino, 1970). The time intervals for data collection were selected to provide time for family adaptation on the resolution process in response to the birth and for measurable differences in the health and developmental status of the infant to occur. In addition, the first eighteen months is the peak time for the occurrence of morbidity complications and mortality (McLaughlin & Shurtleff, 1979).

The data collectors consisted of the principal investigator and one other nurse. Both data collectors were white females between the ages of 32 and 38, with experience in interviewing techniques. In addition, both nurses had experience in working with children with myelodysplasia and their families. The second data collector received an intensive orientation to the data collection protocol and instruments from the principal investigator.

Infants

The physical examinations in the neonatal period were all completed by a board certified pediatrician specializing in the care of children with neurological problems. The follow up history and physicals (3, 6, 12 and 18 months) were completed by the same pediatrician and/or one of two clinical nurse specialists with master's degrees, one of whom was the researcher. The examinations were conducted during a regularly scheduled appointment at the myelodysplasia care center. During each center visit, the infants were also examined by a urologist, orthopedist, neurosurgeon and physical or occupational therapist. Data from each examination were recorded on the medical records and later transferred during chart review to the study infant data sheets for coding and keypunching. Also, data from other clinic visits and hospitalizations were collected during chart reviews.

Developmental testing was completed by the two data collectors. The second data collector, who completed the developmental testing on 90% of the subjects, received reliability training on the EIDP by its developers at the Institute for the Study of Mental Retardation and Related Disabilities (ISMRRD) in Ann Arbor, Michigan. Subsequently, interrater reliability between the testers was monitored by using a tester observer paradigm and discussions of findings and procedures at periodic intervals. The testing was administered during the regularly scheduled myelodysplasia clinic visit or during home visits. The home visits were conducted if there was insufficient time during clinic, if the infant was not amenable to testing in the clinic, or if the clinic appointment was not kept. All home visits were prearranged at the parent's convenience.

Parents

Initial Contact Because the research site is a myelodysplasia care center, the practice is for the local hospitals to transfer the infant within hours of birth. As soon as possible after admission of the infant, the parents met with members of the myelodysplasia care team for discussion of their infant's condition and prognosis. After this discussion, the nurse clinical specialist or the social worker on the myelodysplasia team informed the parents they would be contacted in person by one of the nurse data collectors during a visit to their infant. No parent was ever asked to come in just for data collection purposes.

The parent(s) were greeted by the data collector, escorted to a waiting room within the patient area and informed of the study (see Appendix B for informed consent). If the parent(s) agreed to participate, written informed consent was obtained and the Profile of Mood States (POMS) administered. If the parent(s) desired, the materials, including the statement explaining the study, were given to the parents with a stamped return envelope. Because of their familiarity with the infant and knowledge of myelodysplasia, the data collectors responded to parent(s) questions about the defect and available resources.

Follow-up Contacts The collection of parent data and infant data at 3, 6, 12 and 18 months was completed during a regularly scheduled visit to the myelodysplasia clinic or in the home. With parental agreement, some infants were scheduled one hour before the regularly scheduled afternoon clinic to provide a more relaxed environment for developmental testing and time for parental completion of the questionnaire.

If the parents did not complete the questionnaires during the clinic visit and/or both parents were not in clinic, the questionnaires were taken home with a stamped envelope for return. If the clinic appointment was not kept or the developmental testing was not completed during the clinic visit, a home visit was made. In additton, three parents appeared to have difficulty with reading the questionnaires, therefore, home visits were scheduled to permit the data collector to read the questionnaire to the parent.

The variety of approaches for data collection were used to limit the amount of missing data and facilitate the parents' participation. An earlier study with parents of children with myelodysplasia in Michigan showed no significant difference between data collected by mail and by home interview (Feetham, 1976). In addition, Bohrnstedt (1967) cites no difference between questionnaire and interview collection of the same data on 200 subjects.

Data Analysis

All variables from both the infant and parent data were coded such that a low score corresponded to none or a low incidence of a problem or indicated a normal finding and a higher score indicated dysfunction. The coding of variables was ordered so that a correlation would be positive if that was expected.

Six composite scores were derived from the infant, parent and family environment data for the independent and dependent variables. The process of selection of a variable as part of a composite score was based first on the clinical judgment of the researcher, a nurse clinical specialist, in collaboration with a board certified pediatrician. A second step in developing the composite score was to include variables from research which suggested a correlation between the manifestation of the defect and infant, parent, and/or family environment outcomes. A third step in the process was review of the descriptive statistics for each variable at each time period to assure sufficient data were available for continued analysis. Finally, intercorrelation matrices _within_ each of the six variables were computed. The matrices were examined in order to delete those variables not providing independent information within the composite score for each independent variable. The composite scores used for each independent variable are: (1) the neonatal infant score; (2) the follow-up infant score; (3) the parent and environment score; (4) the parent perinatal score; and (5) the Profile of Mood States (POMS) score. In contrast to the composite scores, the POMS score was computed on the 65 items as directed in the test manual.

Multiple linear regression was used for hypotheses I and II to determine the relationships between the dependent variable of family functioning and the independent variables of infant, parent and family environment outcomes.

Multiple regression is used when analysis of the relationship between a dependent and a set of independent variables is required as in this study design. In addition, the decision to use multiple regression was based on the facts that regression is the procedure of choice when multiple correlations are desired and that this correlation procedure can tolerate violations of parametric assumptions such as a nonrandom sample. Also, the total data sets at each time period, as listed in Appendix C, indicated that a minimum 4:1 or 5:1 subject to variable ratio could be maintained. Because of the complex process in developing the composite scores and the sample size an alpha of $\underline{p} < .05$ was set as the criterion for rejecting the null hypotheses.

That a significant amount of variance in family function will be accounted for by infant outcome, parent outcome, and family environment variables at each of the four time intervals (3, 6, 12 and 18 months) following the birth of an infant with myelodysplasia was tested in Hypothesis I.

While this hypothesis suggests the importance of the relationships of all the variable scores to family functioning, Hypothesis II postulates the greatest amount of information is provided by the infant scores.

One independent variable composite score, the infant neonatal score, remained constant in the regression equation for Hypothesis I at each time. The infant neonatal score was derived from the neonatal period but was retained in the equation to examine data collected at 3, 6, 12 and 18 months because both clinical observation of these families and researchers' reports suggest a carryover effect of the perinatal period on both the infant outcomes and parent outcomes (Farber, 1959; Feetham, 1976; Hayden, Shurtleff & Broy, 1974). Multiple regression analyses on the infant, parent and family environment measures were used to examine the predictive validity of the measures and the amount of variance in family functioning accounted for by each independent variable (composite score).

A simple repeated measures ANOVA with control for missing data and t tests were completed to test Hypothesis III: that the family functioning score will be greater at the first year anniversary of the birth of the infant than at the other time periods. All of the analyses of the data in this study were computed using programs from the Statistical Package for the Social Sciences (SPSS) and the multiple regression equations were computed using standardized regression coefficients.

A total of 38 infants and 66 parents comprised the study sample. Of the 66 parents, 38 mothers and 28 fathers participated in data collection at least once during the five time periods. Because the sample did not include mother-father pairs for each infant, t tests for independent samples were computed for both single variables and composite scores to determine if there were significant differences between the responses of the mothers and fathers.

CHAPTER IV

RESULTS OF DATA ANALYSIS

The relationship among the infant, parent and family environment variables with family functioning in families of infants with myelodysplasia is tested in this research. The dependent variable, family functioning, is a composite score of 21 measures of the difference between what the parent perceives a family function should be and what it is. The independent variables of infant, parent and family environment are composite scores derived from review of the infants' hospital records, developmental testing of the infant and the parents' responses on specified items from the Parent Survey. To determine the relationships among the infant, parent and family environment variables and family functioning, multiple linear regression, t tests, Pearson correlations, and a simple repeated measures ANOVA were used.

Comparison of Mothers and Fathers

Because the sample did not include mother-father pairs for each infant, t tests for independent samples were computed for both single variables and composite scores to determine if there were significant differences between the responses of the mothers and fathers. There was a total of 66 parents for the 38 infants comprising the study sample. Of these 66 parents, 38 mothers and 28 fathers participated in data collection at least once during the five time periods. There were no significant differences at the .05 level between the mothers' and fathers' responses. The summary of mothers' versus fathers' scores is presented in Table 4 and 5. Because there is no significant difference between the mothers' and fathers' responses, to avoid having two parent scores for some infants, the fathers' data were not used in further analysis.

Table 4

Composite Variables Comparison of Mothers' versus Fathers' Responses

Composite Variable	Mothers		Fathers			Signi-ficance
	M	SD	M	SD	t Value	
Family function score						
3 months	15.550	15.350	11.000	9.426	1.08	NS
18 months	20.315	21.990	12.000	7.000	1.35	NS
Parent-Family Environment						
3 months	65.144	4.252	58.053	9.872	0.61	NS
18 months	66.890	9.572	56.240	6.191	2.02	NS
POMS						
3 months	0.371	0.574	0.339	0.679	0.15	NS
18 months	0.185	0.542	0.264	0.630	-0.30	NS
Perinatal Score	28.213	7.908	25.994	7.495	0.83	NS

Sample Size	3 months	18 months*
Mothers	27	16
Fathers	19	8

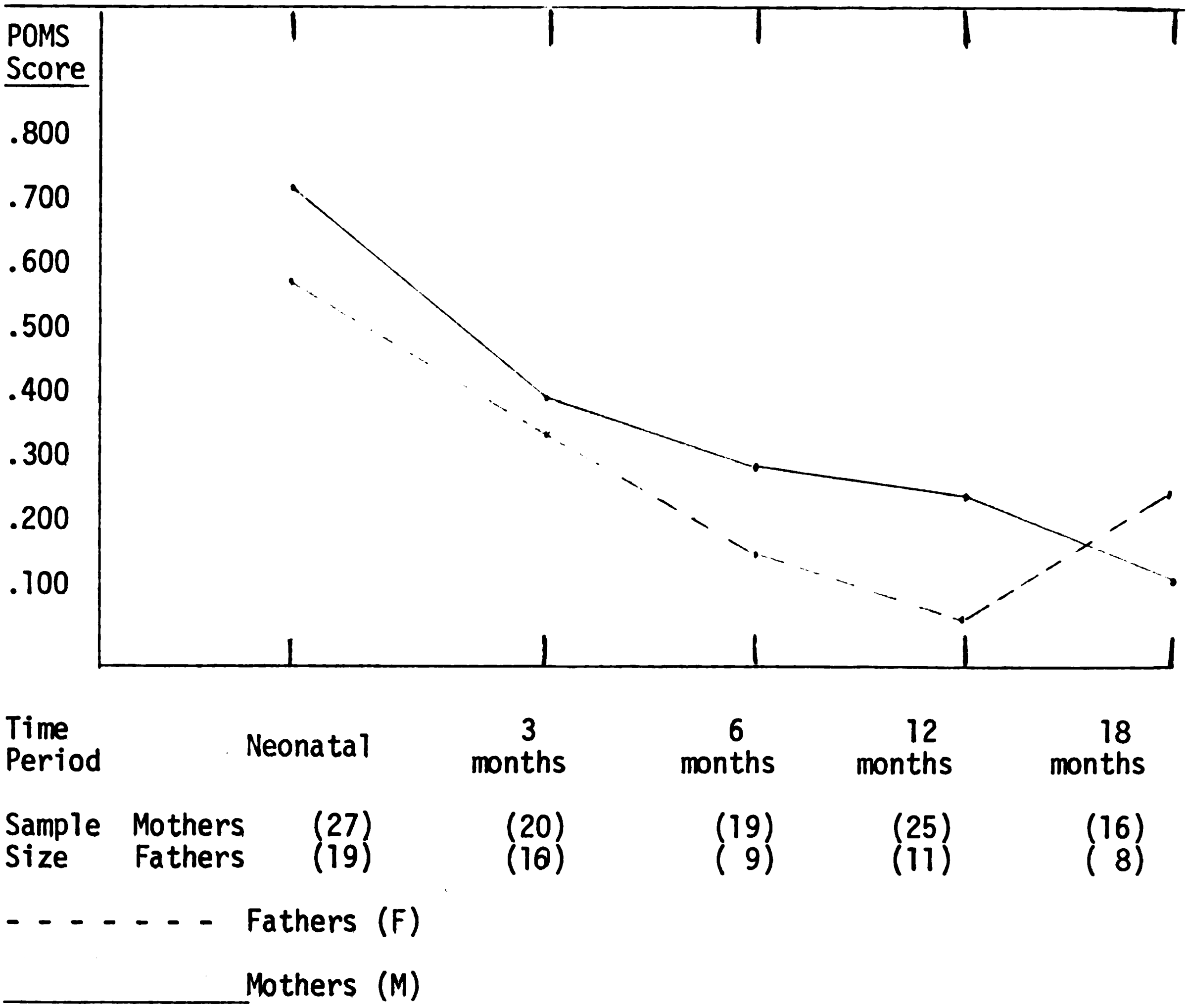
*The N at 18 months is affected by the number of infants not yet reaching 18 months of age by the end of the study.

Table 5

Profile of Mood States (POMS) Comparison of Mothers' and Father's Scores

Time Period	Mothers			Fathers			Combined			Mothers vs Fathers t values	Signi-ficance
	M	SD	(N)	M	SD	(N)	M	SD	(N)		
1 (Neonatal)	0.709	0.683	(27)	0.591	0.696	(19)	0.660	0.683	(46)	0.57	NS
2 (3 months)	0.371	0.574	(20)	0.339	0.679	(16)	0.345	0.561	(36)	0.15	NS
3 (6 months)	0.207	0.609	(19)	0.163	0.514	(9)	0.193	0.596	(28)	0.66	NS
4 (12 months)	0.299	0.558	(25)	0.055	0.407	(11)	0.245	0.516	(36)	1.47	NS
5 (18 months)	0.186	0.542	(16)	0.265	0.630	(8)	0.212	0.560	(24)	-0.30	NS

Although there is no significant difference between the mothers' and father's scores on both the dependent and independent variables, there are differences in the pattern of the scores. The pattern of responses to the POMS is presented in Figure 6 indicating the fathers' scores are lower than the mothers' until 18 months following the birth when their scores increase over the mothers' scores. In addition, the difference between the parent-family enviornment scores of the mothers and fathers also increased at each time period. The variable



		Neonatal	3 months	6 months	12 months	18 months
Sample Size	Mothers	(27)	(20)	(19)	(25)	(16)
	Fathers	(19)	(10)	(9)	(11)	(8)

Figure 6 Patterns of Responses of Mothers and Fathers on Profile of Mood States (POMS) at 3, 6, 12 and 18 months

scores contributing to this increasing difference were the infant care score and home environment rating score. The mother's perception of their time in caring for the infant increased over the fathers' at each time period while their rating of their home environment decreased. A third area of increasing difference in the mothers' and fathers' scores were the family functioning items related to activities and communication with the person assuming the role of spouse with the mothers' discrepant score higher than the fathers'.

Between time groups analysis

Because the number of mothers completing the Parent Survey over all five time periods was small (N = 4), t tests were computed to determine differences between the groups at each time period. The responses of mothers responding at time two (3 months) but not time three (6 months) were compared; (1) with mothers responding at time four (12 months) and not time two (3 months); as well as (2) compared with mothers responding at both times two (3 months) and four (12 months). The three parent composite scores, parent-family environment, POMS and family functioning score were used in the t tests. None of the Fs for the t tests were significant at the .05 level. In addition, the scores for the infants of these same sets of mothers were examined. Again, there were no significant differences between groups on the infant neonatal and follow-up scores at each time period. The lack of significance on the predictor and criterion variables suggests that subjects with complete data, also subsequently used in the multiple regression, are similar to the subjects not used in the regressions because of incomplete data for all scores.

Hypothesis I

To test Hypothesis I, that a significant proportion of variance in family functioning will be accounted for by infant, parent and family environment variables at each of four time intervals (3, 6, 12 and 18 months) following the birth of an infant with myelodysplasia, multiple regression with listwise deletion of variables was used. The stepwise regressions using the four predictor variables at Time 2 (three months) are reported in Table 6. The data fail to show that knowledge of infant, parent and family environment variables adds to the ability to predict family functioning over and above no knowledge.

The regression summary for Time 3 (six months) is presented in Table 7. The POMS score was the only variable that contributed significant information to the prediction of family functioning at this time period. The proportion of variance accounted for by the POMS is 60% supporting the research hypothesis. For the regression at Time 4 (twelve months) both infant and parent variables are significant predictors. The variable accounting for the greatest variance (25%) is the neonatal infant variable, a score which is unchanged at each time period. The second variable contributing significant information (accounting for 22% of the variance) is again the POMS. The amount of independent variance contributed to the prediction of family functioning by each of these variables was about the same. The analysis summary for Time 4 is presented in Table 8.

The POMS variable also is a significant predictor at Time 5 (eighteen months) and accounts for 43% of the variance. The neonatal infant score enters the regression equation on the second step but is not significant. The analysis summary for Time 5 is presented in Table 9.

Table 6

Hypothesis I

Summary of Stepwise Multiple Regression
Analysis to Predict Family Functioning
at Time 2 (three months)
(N=13)

Step	Predictor Variables	R	R^2	Simple r	F	Significance
1	Parent-Family Environment	0.182	0.033	-0.182	0.375	NS
2	Neonatal infant	0.296	0.088	0.138	0.602	NS
3	POMS	0.304	0.092	0.007	0.028	NS
4	Followup Infant	0.311	0.096	-0.034	0.037	NS

Table 7

Hypothesis I

Summary of Stepwise Multiple Regression
Analysis to Predict Family Functioning
at Time 3 (six months)
(N=11)

Step	Predictor Variables	R	R^2	Simple r	F	Significance
1	POMS	0.772	0.597	0.772	13.30	$< .05$
2	Followup Infant	0.777	0.603	0.036	0.134	NS
3	Neonatal Infant	0.799	0.638	0.421	0.668	NS
4	Parent-Family Environment	0.799	0.638	0.104	0.016	NS

Table 8

Hypothesis I

Summary of Stepwise Multiple Regression Analysis to
Predict Family Functioning at Time 4 (Twelve Months)
(N=16)

Step	Predictor Variables	R	R^2	Simple r	F	Significance
1	Neonatal Infant	0.498	0.248	-0.498	4.629	< .05
2	POMS	0.681	0.463	0.442	5.213	< .05
3	Parent-Family Environment	0.702	0.493	0.491	0.702	NS
4	Followup Infant	0.705	0.498	-0.102	0.100	NS

Table 9

Hypothesis I

Summary of Stepwise Multiple Regression Analysis to
Predict Family Functioning at Time 5 (Eighteen Months)
(N=12)

Step	Predictor Variables	R	R^2	Simple r	F	Significance
1	POMS	0.654	0.428	0.654	7.471	< .05
2	Neonatal Infant	0.699	0.489	-0.258	1.087	NS
3	Followup Infant	0.753	0.567	0.414	1.425	NS
4	Parent-Family Environment	0.771	0.594	0.638	0.473	NS

Although interpretation of the data must be limited because of the sample size available for each regression, it can be noted that the POMS is the major significant predictor of family functioning at three of the four time periods. Although the neonatal infant score is a significant predictor at one time period, neither the parent-family environment score nor the score representing the current status of the infant provide a significant amount of information at any of the four time periods.

The original study design called for inclusion of a fifth predictor variable, the perinatal parent score. However, the number of complete data sets at each time period did not support the inclusion of this fifth variable as the sample having complete data on all variables dropped to nine at six months and four at 18 months. At Time 2 (three months) Pearson correlations of the perinatal variable with the criterion variable, family functioning, suggest that with a larger sample, addition of the parent perinatal variable may add significant information to family functioning. The correlations with the parent-perinatal variable is presented in Table 10.

In summary, the data at 6, 12 and 18 months support the research hypothesis that a significant proportion of variance in family functioning is accounted for by infant, parent and family environment variables.

Hypothesis II

To test Hypothesis II, that the composite infant scores will account for greater variance in family functioning than will the parent-family environment composite score and POMS at 3, 6, 12 and 18 months following the birth of an infant with myelodysplasia, two multiple regressions were computed at each time period. The first multiple regression entered the infant scores into the equation prior to the parent scores. The second mutliple regression reversed the order of entry.

Table 10

Correlations of Predictor Variables with Family Functioning Including the Parent Perinatal Score

	FF2	FF3	FF4	FF5
Sample Size	N=13	N=9	N=11	N=4
Predictor Variables				
Parent Perinatal	0.5499 p=0.026*	0.7052 p=0.017*	0.4667 p=o.074	0.9956 p=0.002**
Neonatal Infant	0.1380 p=0.327	0.7189 p=0.015*	-0.2857 p=0.197	0.9825 p=0.009**
Parent-Family Environment	-0.1815 p=0.276	0.1500 p=0.350	0.4047 p=0.108	0.3550 p=0.322
Followup Infant	-0.0336 p=0.457	0.0487 p=0.451	-0.2379 p=0.241	0.8516 p=0.074
POMS	0.0071 p=0.491	0.8623 p=0.001**	0.3168 p=0.171	0.9845 p=0.008**

* $p \leq .05$ ** $p \leq .01$

The results of the first regression, infant information entered ahead of parent information, varied by time period. At Time 2 there was no significant change with infant or parent variables. At Time 3 the infant data did not contribute significantly while entry of the first parent variable (POMS) added significant information. At Time 4 both the infant and parent variables contributed significantly with each accounting for 25% of the variance. At Time 5 the parent data did not contribute significant information.

The results of the second regression, parent information entered ahead of infant information, also varied by time period. Although parent information was significant at Time 3, Time 4 and Time 5, the infant information did not add significant independent variance at any time. In summary, analysis revealed that the infant variables did not account for significant information over and above the parent information or no information in predicting family functioning. The research hypothesis is not supported from these data.

Hypothesis III

There were only four families with complete data at each time period. This negated the use of a repeated measures ANOVA covering all four time periods. Therefore, to test Hypothesis III, that family functioning discrepant scores will be greater at the first year anniversary of the birth of the infant with myelodysplasia than at 3, 6, and 18 months following the birth, a repeated measures ANOVA at 3, 6, and 12 months (N=7) and at 6, 12 and 18 months (N=6) was used. The mean scores for these small sample sizes correlated significantly with the means for the total sample. There was no significant difference between the family functioning scores for each time period for the mothers with complete data from three to 12 months and mothers with complete data from six to 18 months. In addition, repeated measure t tests were computed between pairs of the time periods on the dependent variable. The dependent variable, family functioning discrepant scores, at 3, 6, and 18 months were each compared with the 12 month family functioning scores. The results of these analyses are presented in Table 11. In summary, the family functioning scores are not significantly different at the twelve month anniversary period from the 3, 6 and 18 month periods following the birth of the infant. Hypothesis III was not supported by the data.

Table 11

Hypothesis III

Change in Mothers' Family Functioning Discrepant Score Between Time Periods

Mothers' Family Functioning Score Time	Sample Size	M	SD	t/F value	Significance
2 (three months)	13	18.769	17.801	-0.22	NS
4 (twelve months)		19.301	14.688		
3 (six months)	11	15.909	11.049	-1.54	NS
4 (twelve months)		20.818	9.806		
4 (twelve months)	12	18.333	13.473	-0.48	NS
5 (eighteen months)		20.083	23.083		
Mothers at 2	9	14.556	19.749	-0.24	NS
Mothers at 2 & 4	11	16.364	11.578		
Mothers at 4	9	25.778	17.057	1.41	NS
Mothers at 2 & 4	13	16.846	9.940		
2 (three months)		17.714	13.363		
3 (six months)	7	14.714	12.244	0.479	NS
4 (twelve months)		18.571	9.360		
3 (six months)		15.833	13.512		
4 (twelve months)	6	18.500	11.743	0.381	NS
5 (eighteen months)		16.167	12.529		

The family functioning discrepant scores were most stable over time for those mothers with complete data over three or four time periods. Mothers with data for one or two periods had higher family functioning discrepant scores, although there was no significant difference in the scores. (When the individual items comprising the family functioning score are examined the items contributing the greatest increase in the [illegible] discrepant score are those items related to the relationship with the person assuming the role of spouse, e.g., emotional support from the spouse; amount of time with the spouse; and satisfaction with sexual relations.

Summary

Three hypotheses were tested in this study. The results of the tests of each hypothesis are presented in Table 12. Multiple regression with stepwise inclusion and listwise deletion was used to test Hypotheses I and II. ANOVA with repeated measures and t tests were used to test Hypothesis III. The incomplete subject data for all four predictor variables and the one criterion variable resulted in a reduction from a potential sample at each time period of 24 to 30 to a sample range of 11 to 16 for the regressions.

Table 12

Summary of Hypotheses

Hypothesis		Research Hypothesis	Significance level
I	Time 2	not supported	NS
	Time 3	supported	$p < .05$
	Time 4	supported	$p < .05$
	Time 5	supported	$p < .05$
II		not supported	NS
III		not supported	NS

Hypothesis I, that parent-family environment and infant data add significant information to the knowledge of family functioning, is supported by the data. However, Hypothesis II, that the infant variables would account for a greater proportion of variance than the parent variables in predicting family functioning, is not supported. Also, analysis revealed that Hypothesis III, that the family functioning discrepant score would be higher at the one year anniversary of the infant's birth, is not supported.

CHAPTER V

SUMMARY AND CONCLUSIONS

Summary

The incidence of neural tube defects is 1.7 to 3 per 1,000 live births in the United States with 130 such births each year in Michigan. Myelodysplasia (also known as myelomeningocele and/or spina bifida) is the most common (60%) of the neural tube defects and is the one defect most frequently associated with survival. Because of improved surgical and medical techniques, increasing numbers of children with this defect are surviving and living with their families. Although myelodysplasia is a serious birth defect, persons with this defect grow to live productive adult lives. The physical manifestations of myelodysplasia include hydrocephalus in 80% of the children, handicaps of locomotion and continence in 95%, and perceptual and learning disabilities and/or intellect below their unaffected siblings in 25%.

The birth of an infant with myelodysplasia affects the family in several ways. An initial and ongoing effect is that several professionals are introduced into the family system far beyond the usual number introduced with the birth of a normal child. Secondly, the infant requires special care in the home and in both the health care and educational systems. This special care takes more time than the care of a normal infant and often requires the addition of special equipment in the home. The "cost" of this special care in both energy expended by the family members and in monetary terms is a third effect on the family system. The long term outcome of families raising these children in the home is documented and indicates an increased incidence of divorce, separation, maternal malaise and sibling problems over families without children with chronic health problems (Richard & McIntosh, 1973; Tew & Laurence, 1973; Walker, Thomas & Russell, 1971).

The conceptual framework selected for this research is the structural-functional approach to family study (Eshelman, 1974). Family functioning is conceptualized holistically as the activities of everyday life or the way in which the family, as a system, operates across many dimensions. Clinical observations, the review of family theory and the review of research related to children with myelodysplasia and their families suggest there are relationships among infant, parent and family environment variables and family functioning (Garbarino, 1977; Travis, 1976). Therefore, the purpose of this study was to examine the relationships among variables pertaining to the parents, to the infant with myelodysplasia and to the family environment with family functioning at selected time intervals in the first 18 months following the birth of the infant.

The dependent variable, family functioning, is a composite score of family functioning across 21 indicators. The independent variables are composite scores of infant, parent and family environment variables as measured by specified items from the Parent Survey (Feetham & Perrin, 1977), Profile of Mood States (POMS) (McNair, Lorr & Droppleman, 1971), and infant hospital records and developmental testing using the Early Intervention Developmental Profile (EIDP) (Rogers & D'Eugenio, 1977).

Three hypotheses were tested in this study, using multiple linear regression with stepwise inclusion and listwise deletion to test Hypotheses I and II. A simple ANOVA with repeated measures and t tests were used to test Hypothesis III.

The subjects for this descriptive longitudinal study were parents and their infants with myelodysplasia admitted to a Myelodysplasia Care Center in a large urban pediatric hospital. Forty-six infants with myelodysplasia were admitted to the care center during the time of the study from March 1977 to July 1979. These infants represent 40% of the live born infants with myelodysplasia in Michigan in this time period. The study sample was a total of 38 infants and 66 parents (38 mothers and 28 fathers) who both met the study criteria and agreed to participate. There were no significant differences between the mothers' and fathers' responses, therefore, because of unequal N's between mothers and fathers, the fathers' responses were not used in the multiple regression, ANOVA, and t tests to test the three hypotheses.

Hypothesis I, that the infant and parent-family environment predictor variables add significant information to the ability to predict family functioning, was supported by the data. However, Hypothesis II, that the infant composite score variables would account for a greater proportion of variance than the parent variables in predicting family functioning, was not supported. Also, Hypothesis III, that the family functioning discrepant score would be higher at the one year anniversary of the infant's birth than at 3, 6 and 18 months, was not supported by the data.

Conclusions

Study Design Several considerations for research, clinical practice and teaching derive from this study. The use of a multivariate design to examine family functioning in families with children with myelodysplasia is supported, while raising a challenge to the univariate designs implying single causation between the birth of the child with a defect and the outcomes of the individual or family (Freeston, 1971; Richards & McIntosh, 1973; Walker, Thomas & Russell, 1971). The multivariate design is also supported by the work of Garbarino (1977; 1978) and Sims, Paolucci & Morris (1972). If the results of this study can be replicated with other families with children with myelodysplasia or children with other chronic health problems, the applicability of this multivariate design to research, practice, and teaching is strengthened. In addition, as indicated previously, this study is unique as it is prospective from the time of birth of the infant and simultaneously examines parent, infant and family environment variables. The value of the prospective design is reinforced by the parent scores which indicate a high level of parental distress in the neonatal period.

Structural-functional framework In addition to the use of the multivariate design being supported, characteristics of the structural-functional framework are also supported in this research. The purpose of functional analysis is to explain: 1) the parts, 2) the relationship between the parts and the whole, and 3) the functions that are performed by the parts. The importance of examining the relationships among the parent, infant and family environment variables is reinforced by the fact that different independent variables provided significant information in predicting family functioning at 6, 12 and 18 month time periods. In future studies, other researchers could test other independent variables within the infant, parent and family environment composite scores to determine the amount of information provided by different variables in relation to family functioning.

Another characteristic of the structural-functional framework supported by this research is the issue of system maintenance. It is implied in the issue of system maintenance that a variety of factors within and outside the family influence the level of functioning of the total unit. The family environment composite score included environmental variables such as availability of transportation, distance to health care services, and the number of persons entering the family system. In future research, a researcher could identify what they perceive to be pertinent infant, parent and family environment variables and test the relationship of these variables to family functioning whether it be with a normal child within a family or a child with a chronic health problem. The ability to identify the significant variables affecting family functioning could provide the basis for the development of clinical assessment tools to be used to provide interventions to prevent family dysfunction.

Parent data   Other clinical and research implications can be derived from this study. It is important to note that the Profile of Mood States (POMS), a parent measure, entered the regression equation at three of the four time periods at a level providing significant information in predicting family functioning. The POMS scores were highest for the parents at the neonatal period and at the one year anniversary of the birth, supporting clinical observations and researchers' reports of the stress of these periods (Feetham, 1976; Mercer, 1974; Wolfensberger & Menalascino, 1970). For further interpretation of the POMS data, studies using larger samples are needed to compare families with normal infants and families with infants with myelodysplasia. If additional studies support the predictive ability of the POMS with family functioning, the POMS could be a useful clinical tool which could augment clinical judgment when planning intervention with families.

Although not significant, by 18 months following the birth of the infant, the increasing differences between the mothers' and fathers' POMS scores support the need for continued study and suggest the need for preventive intervention. The trend toward differences between the mothers and fathers supports the importance of studying both parents. This beginning discrepancy between the mothers and fathers may be the foundation for later family dysfunction. Further data analysis also needs to be completed comparing the POMS scores of the single mother living with the father, the single mother with no one assuming the spouse role, and the married mother living with her spouse.

Neonatal infant data The clinical issue of the extent of the carryover effect of the neonatal period, following the birth of a child with a defect, to family functioning, is also raised in this study. The independent variable, of the neonatal infant score, presented in the regression equation as being most predictive of family functioning at one year following the birth of the infant. By the carryover effect it is implied that the parent's perception of the neonatal period and the actual experiences and outcomes of the infant have an ongoing effect on family functioning. The concept of the carryover effect of the stress of the neonatal period following a birth of a child with a defect is also supported by the clinical observations and research of others (Mercer, 1974; Travis, 1976). Some carryover effect is assumed with the brith of a normal child. However, if the findings of this study were to be replicated with a larger sample of families with infants with myelodysplasia or infants with other defects, yet not replicated with families with normal infants, the extent of carryover of the neonatal period following the birth of an infant with a defect, to later functioning of the family would be supported. If the presence of the carryover effect is supported in future research, the development of instruments to assess parent and infant outcomes that would be predictive of later family functioning should follow the research.

Using these data, a clinical approach by the professional, when working with families of children with birth defects, would be to counsel the parents that the one year anniversary of the birth may reactivate thoughts and feelings of the newborn period. Professionals can then provide the family with an opportunity to discuss their feelings and perhaps enable them to progress through another stage in the resolution of the brith of an infant with a defect.

Family Environment The family environment items, within the parent-family environment score, were conceptualized as a quantified measure of entry into the family system. Other studies using multivariate models to examine child abuse and nutritional problems in children have also attempted measures of system entry (Garbarino, 1977; Sims, Paolucci, & Morris, 1972). Although the family environment data did not contribute significant information in the testing of Hypothesis I, some patterns of the responses are interesting to note and suggest areas for future research with both families with normal infants and families with infants with problems. The data are useful from a descriptive standpoint because the extent of system entry in the first 18 months following the birth of an infant with myelodysplasia is documented for this sample of 38 families.

Indicators of family system entry, in this study, included the number of professionals entering the home system because of the infant with myelodysplasia. This home entry measure included arranging with friends, relatives and/or neighbors for transportation to appointments and documented the number of professionals seen either in the health care system and/or in the home. The home entry score, the parents' perception of their home environment, and time spent in the care of the infant with myelodysplasia were all conceptualized as measures of the family environment.

There was no significant difference over time in the home environment score which included an assessment of home: space, heat, food, safety, and clothing. The family functioning discrepant score increased as the home score decreased suggesting a relationship between family dysfunction and the perception of the home environment. There were also patterns of response noted in the mothers' perceptions of their care of the infant with myelodysplasia. The patterns in the mothers' data indicated that, as time perceived to be required for special infant care activities increased, the perceived time for worrying also increased, while time for enjoying and playing with the infant decreased. In order to interpret these data fully, data on both the home environment and infant care scores need to be obtained from parents of normal infants and from a larger sample of parents of infants with myelodysplasia.

To document entry into the family system, parents reported seeing four to six professionals on each visit to the myelodysplasia care center and averaged 2.4 clinic visits in each time period. In addition, at least one professional entered the home during each time period. These professionals enter the family system because of the infant with myelodysplasia. An area for further study and consideration in planning care intervention is the family's ability to maintain their system boundaries in light of the entry of numerous health professionals (Kantor

& Lehr, 1975). An interesting trend in the data was that parents who remained in the myelodysplasia care center from the time of birth of the infant through 18 months or for at least 12 of the 18 months, had lower family functioning discrepant scores and lower POMS scores than parents who were in the care center for only three to six months. Further study is needed to determine if this trend is sustained. The research question becomes: do families experiencing ongoing comprehensive interdisciplinary care function better than families who do not receive this type of care or is it that these families who continue with interdisciplinary care have a higher level of family functioning to begin with and therefore tolerate multidisciplinary contacts?

Future Research Related to Families The state of the art for the development of valid and reliable measures of family functioning as a holistic concept is limited. There are several reasons for the limitations in this area of research. First, the concept of family functioning is still vague and poorly defined (Pless & Satterwhite, 1973). As cited in the theoretical discussion, family scholars are inconsistent in both the definitions and measures of family functioning (McIntyre, 1966). Family functioning may be defined as process, outcome and/or content. The primary value of the various definitions of family functions is that they provide general direction to the researcher to consider problems of the relationships between the family system and other societal systems. One limitation of these definitions is that they have limited theoretical weight and therefore, lead the researcher into statements of generalities and causalities (Smilkstein, 1978). In addition, in reocgnition of the complexity of the many conceptual frameworks for family analysis, it would be misleading to imply that family functioning can be assessed adequately by a simple index. Therefore, an objective for future research related to family functioning should be development of a measure which is of pragmatic value in assisting professionals working with families to identify the potentially dysfunctional family, and also to identify appropriate intervention.

A second objective for research related to families is the accurate, systematic documentation of processes used by families to maintain an adequate level of functioning. However, several methodological issues are raised when considering the second objective. The maintenance of both the integrity and boundaries of a family is of primary importance during research. For families responding to unexpected, potentially devastating experiences, such as the subjects of this study, family maintenance must take precedence over data collection. Therefore, the timing and methods of data collection are major issues. Researchers must be sensitive to the context of the family and to evaluate the appropriateness of extensive interviews, video taping and/or participant observation during the time a family is responding to an unexpected event such as the birth of a child with a defect. Since research related to families with normal children is relatively limited, research on normal families is needed prior to and/or concurrent with research examining family functioning in families responding to unexpected experiences.

Another research question that needs to be addressed in future studies is focusing on the dyadic interaction of the mother and father, over time, in response to the birth of an infant with a defect. In addition, outcomes of families receiving care in a multidiscipline comprehensive care center need to be compared with families receiving, what is perceived by some health professionals as, fragmented care from medical specialists throughout the community. Once the process of response of families to the birth of a child with health problems is documented the ensuing research should test care interventions in a variety of contexts. In addition to examining the process of response of the family unit to the birth of an infant with health problems, researchers need to study the relationships among and between family members, particularly among siblings.

In the preceding conclusions, several implications for both research and clinical practice derived from this study have been discussed. The need for additional process oriented multivariate research to examine the effect of complex health problems on families is clearly supported. In addition to presenting future research directions in this chapter, the value of this unique study is reinforced as it simultaneously examines parent, infant and family environment variables over the first 18 months following the birth of an infant with myelodysplasia. A difference exists in how families respond to the birth of an infant with a severe defect. This study is a beginning in examining the process of how these families respond. This study provides a model for professionals to understand the process and therefore to assist families to sustain and grow through this experience.

APPENDIX A

Appendix A.1

Chart review-Neonatal

1 -5 Family Code____________

Infant Neonatal Assessment

Card 45

(6) Repair___Non-Repair___

(7) Sex M___F___

(8 & 9) Race:____________

(13) Age at Admission_________

(39) Maternal contact visual___touch___

none____not incidated____

(14-17) Birthdate mo____year____

(18-22) Birthweight grams______%______

(23-25) Length cm____%_____

(26-27) Gestational Age ____weeks

(41-42) Apgar 1 minute___

(43-44) 5 minute

(28) SGA__AGA__LGA__

Head circumference

(31-34) at birth___%____

(35-38)at two weeks____%____

(45) Type of defect
Meningocele _____
Myelomeningocele____
Rhichesis____
Other____

Level of functioning

(46-47)
Motor
____T 10 or
____T 11
____T 12
____L 1
____L 2
____L 3
____L 4
____L 5
____S 1&2
____Not clearly specified
____Intact cord

(48-49)
Sensory
____T 10 or
____T 11
____T 12
____L 1
____L 2
____L 3
____L 4
____L 5
____S 1&2
____Not clearly specified
____intact cord

Associated anomalies
(50) Vertebral (specify) ____
(51) Arnold chiare ____
(52) GI ____
(53) Rectal prolapse ____
(54) Club foot ____
(55) Hip dysplasia ____
(56) Cranial/cerebral defects ____
(57) Lacunae skull ____
(58) GU:uncles, testicle, kid ____
(59) Neurogenec bladder ____
(60) Flexion contractures ____
(61) Orthopedic anomalies ____
(62) desmorphism, FLK, micrognathia ____
(63) Cardiac ____
(64) Other neurology (specify)____

(67)Medical Treatment
____None
____Topical ____
____Systemic____
____Restarted
(68) Surgical Treatment
____None
____Surgical Back
____Surgical Head
(69) Other Surgical Anomalies?
___yes___no

Card 46

(13) Hydrocephalus yes___no___
(14) Age of onset_________
(15-17) Cerebral mantle____cm
(18-19) Brain mass: 60% 60%
(20-23) Brain mass_______gm
(24-25) Ventricular size___ ___cm
(26) Ventriculogram/pneumogram ___yes___no
(27) Enchoencephalogram ___yes___no
(28) Cut Scan ___yes___no
(31) Age of first shunt_________

Complications:
(32) None Rx____________
(33) Infection: CNS Rx____________
(34) Other: Non-CNS Rx____________
(35) Post-op Rx____________
(36) CNS (non-infection) Rx____________
(37) Fractures Rx____________
(38) Meningocele ruptured sac Rx____________
(39) Other Rx____________

Referalls and consultations
(41) Nursing/clinician____
(42) Neurosurgery ____
(43) Urology ____
(44) Pediatrics ____
(45) VNA ____
(46) Social Service ____
(47) ENT ____
(48) Neurology ____
(49) Genetics ____
(50) Cardiology ____
(51) Orthopedics ____

(52) PT/OT____
(53) Opthamology____
(54) Other (unchenalology____

(57)length of first hospitalization ____

(62) Anticipated Primary Care Person
____
____

(61) Desposition
Family care____
Foster care____
Nursing home____
Death ____
Other ____

(58-60)Age at discharge ______

(67) ~~Breech~~ yes___no___
(68) Polyhydramnusus yes___no___
(69- Maternal G___P___AB___AGE___
History or fetal loss________

Family History
(63) Mother________
(64) Father________

Appendix A.2

Chart review-Infant Follow-Up

Family Code____________
2 3 4 5

Infant Follow-up

Repair:
____Repaired ____Non-repair
____Delay Repair

Age____months____days
Height____%____
Weight____%____
Shunt No____Yes____
dysfunction No____Yes____
repaired No____Yes____

Echo No____Yes____
Cerebral mantle____Brain mass____
Head Circumference____%____

Radiation Exposure
skull ____
chest ____
cat skan ____
abdomen ____
hips ____
legs ____
ventriculogram____
Other (specify)____________
Total number ____
IVP____
VCUG____

Diagnosis of Additional Anomalies:
No____Yes ____System:
CNS ____ Specify__________
Genetourinary ____ Specify__________
Musculo-skeletal ____ Specify__________
Sensory visual,
hearing ____ Specify__________
Other ____ Specify__________
Anomaly treated
Yes____No____Not required____
1. Myelo related____
2. Non-myelo related ____

Acute Illnesses:
None____
None noted____
Type (URI, GI, Fractures, etc.)____________
Treated: by parents ____
by physician____
Other____________
none____
1. Myelo-related____
2. Non-myelo related ____

Other Care Problems:
None____
None needed____
Type (diaper rash, feeding, sleeping, etc.)____

Emergency or Physician Visits
None____
None noted____
Number____
Reason____
Well child____
1.. Myelo related____
2. non- myelo related____

Hospitalizations:
None____
Other hospital____
CHM____
Reason____________
1. Myelo related____
2. Non- myelo related ____

Surgical Procedures:
None____
Number____
Reason____________
1. Myelo related__________
2. Non-myelo related__________

Development:
DDST - To Age
Delay____Specify areas__________
ISMRDD - To Age
Delay____Specify areas__________

Myelodysplasia Clinic Visits:
Number scheduled____
Number kept________
Number disciplines seen/visit____
Specify discipline(s)__________

Other appointments:
None____
None noted____
Specify service(s)
PT ____
OT ____
Opthy ____
Nutritionist____
Neurosurg ____
Ortho ____
Other ____
Psych Test ____

Supportive Treatment:
Early intervention____
Newington splint____
Other (specify)____________

Primary Care Physician:
No____
Yes____
None noted____

Disposition:
Family____
Foster care____
Nursing home____
Death____
Other (specify)____________

Physical Findings
Neurological function level of lesion ________
Lower extremity function____
Urinary tract function (regulation) ____________
Problems (specify)____________
Bowel anal wink____
prolapse____
treated(specify)__________
regulation __________
Other pertinent findings (specify) ____________

Comments:

Appendix A.2

Chart review-Infant Follow-Up

Developmental Summary

ISMRDD

11. Language _____
12. Social _______
13. Eating _______
14. Toileting ____
15. Dressing _____
16. Cognitive ____
17. Perceptual Fine Motor _____
18. Gross Motor ____

0 - non testable
1. 0-2 mos.
2. 3-5 mos.
3. 6-8 mos.
4. 9-11 mos.
5. 12-15 mos.
6. 16-19 mos.
7. 20-23 mos.
8. 24-27 mos.
9. Above 27 or other

31. Denver:
    1. normal
    2. questionable
    3. abnormal
32. Personal Social ______
33. Fine Motor _____
34. Language ______
36. Gross Motor ____

Is there a delay?
1. Yes
2. No
3. Untestable

Blank: not applicable

Scoring: (1) On age line for F is indicated by testing.

(2) If child was unable to perform according to chronological age milestones he was tested at a low age milestone until able to pass. All milestone falling between age line and unindicated p. were considered failures.

(3) Did not test for failure if passed all milestones on age line.

Appendix A.3

Early Intervention Developmental Profile *

Developmental Programming for Infants and Young Children

D. Sue Schafer and Martha S. Moersch, Editors

Early Intervention Developmental Profile

by Sally J. Rogers, Diane B. D'Eugenio, Sara L. Brown, Carol M. Donovan, and Eleanor W. Lynch

The University of Michigan Press
Ann Arbor

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Introduction

Early Intervention Developmental Profile is a compilation of major developmental milestones for use with children whose cognitive, motor, social, self-care, and/or language skills fall within the zero to thirty-six-month developmental range. The profile provides a systematic means of evaluating a child's skills, selecting appropriate objectives for treatment of developmental delays, and designing an appropriate, individualized curriculum based on a developmental model.

The profile evaluates the child's functioning in six areas: perceptual/fine motor, cognition, language, social/emotional, self-care, and gross motor. The profile is designed to be adminstered up to four times a year for one child by an interdisciplinary team which includes a psychologist or special educator, physical and/or occupational therapist, and a speech therapist. Each section of the profile assumes a certain degree of disciplinary knowledge and skill on the part of the evaluator, skills which can be taught to other members of the team.

Scoring procedures for each item are described in volume 1, *Assessment and Application*, of the three-volume set *Developmental Programming for Infants and Young Children*. Briefly, an item *passed* by the child is marked with a P if the criteria are met. When there is a question as to whether the child has fully met the criteria for an item, a *pass-fail* (PF) should be used to indicate the emergence of the skill measured by that particular item. Clear *failures* are marked by an F. A final scoring category, O, is used to signify that an item has been *omitted* by the evaluator because of intervening variables which should be described in the scoring box.

The child's performance can be plotted on the profile graph (inside back cover) by marking the highest number of consistently passed items in each of the six areas and then connecting the marks. The resulting graph provides a visual representation of the child's relative strengths, weaknesses, and general developmental level. Objectives for the child in each developmental area can be designed to aid the acquisition of skills in the appropriate developmental level, supporting the strong (highest) skills as well as facilitating the development of the weak (lowest) area(s). The process of translating evaluation data into individualized programs is fully discussed in volume 1.

The materials necessary to administer the entire profile are listed below:

Rattle
Bell
Small doll
Black and white pictures of common objects
Bottle for doll
Doll's chair
Saucer
Mirror
Balls, small (4-inch diameter)
    medium (8-inch diameter)
    large (12-inch diameter)
Paper-wrapped candy
Toy car
Cloth or diaper
Sponge
Baby bottle
Three identical coffee cups

Ten 1-inch cubes—two each of red, blue, yellow, green, black
1/2-inch cube
Small clear vial
12-inch stick
Small square box
Two sets of 1-inch cards with four geometric shapes—circle, square, star, cross
Penny
Ring on string
Raisins or sugar pellets
Six pegs (3/8-inch to 1/2-inch diameter)
Six-holed pegboard
Crayon
Paper
Picture book with cardboard pages
Three-piece formboard
Blunt-end scissors
Push toy
Stairs

Child-size chair
Adult-size chair
Balance beam
Tricycle

Name ______________________________

Perceptual/Fine Motor

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
	0-2 Months				
1	Follows moving object past midline				
	3-5 Months				
2	Integration of grasp reflex				
3	Reaches for dangling object				
4	Moves head to track moving object				
5	Fingers own hands in play at midline				
6	Uses ulnar palmar prehension				
7	Looks at hands				
8	Reaches for cube and touches it				
9	Uses radial palmar prehension (uses thumb and two fingers)				
10	Transfers toy from hand to hand				
	6-8 Months				
11	Retains two cubes after third is offered				
12	Rakes or scoops up raisin and attains it				
13	Has complete thumb opposition on cube				
14	Pulls one peg out of pegboard				
15	Uses inferior pincer grasp with raisin				
	9-11 Months				
16	Pokes with isolated index finger				
17	Drops blocks imitatively with no pause before release				
18	Uses neat pincer grasp with raisin				
19	Attempts to scribble (holds crayon to paper)				
20	Holds crayon adaptively (crayon projects out of radial aspect of the hand, one end up and one end down)				

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Volume 2. Early Intervention Developmental Profile

Perceptual/Fine Motor

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
	12-15 Months				
21	Turns page of cardboard book				
22	Removes cover from small square box				
23	Places one or two pegs in pegboard				
24	Builds two-cube tower				
25	Scribbles spontaneously (no demonstration)				
	16-19 Months				
26	Places six pegs in pegboard without help				
27	Builds three-cube tower				
28	Places round form in formboard (three forms presented)				
29	Imitates crayon stroke (crayon gripped with butt end firmly in palm)				
	20-23 Months				
30	Places six pegs in pegboard in 34 seconds				
31	Makes vertical and circular scribble after demonstrations				
32	Completes three-piece formboard				
33	Builds six-cube tower				
34	Begins to manipulate crayon with fingers				
35	Folds paper imitatively				
	24-27 Months				
36	Draws vertical and horizontal strokes imitatively				
37	Aligns two or more cubes for train, no smokestack				
	28-31 Months				
38	Builds eight-cube tower				
	32-35 Months				
39	Copies a circle already drawn				
40	Cuts with scissors				

Developmental Programming for Infants and Young Children
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Name ______________________________

Cognition

Item Number	DEVELOPMENTAL LEVELS AND ITEMS	Date	Date	Date	Date
	0-2 Months				
41	Uses adaptive movements rather than reflexive reactions				
	3-5 Months				
42	Demonstrates vocal contagion				
43	Repeats random movements (primary circular reactions)				
44	Watches place where moving object disappeared				
45	Coordinates two actions in play				
	6-8 Months				
46	Attains partially hidden object				
47	Looks to the floor when something falls				
48	Uncovers face				
49	Rotates a bottle inverted less than 180° to drink				
50	Acts to have pleasurable interaction repeated				
51	Imitates sounds or hand movements already in his repertoire				
	9-12 Months				
52	Pulls string to secure ring and succeeds				
53	Imitates facial movements inexactly				
54	Attains completely hidden object (single visible displacements)				
55	Imperfectly imitates sounds and movements never performed before				
56	Shows knowledge of toy hidden behind a screen				
57	Rotates a bottle inverted 180° to drink				
	13-18 Months				
58	Repeatedly finds toy when hidden under one of several covers (multiple visible displacements)				
59	Balances eight 1-inch cubes in a coffee cup				

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Cognition

Item Number	DEVELOPMENTAL LEVELS AND ITEMS	Date	Date	Date	Date
60	Lifts a 1/2-inch cube off a 1-inch cube cleanly, with pincer grasp				
61	Inverts a small vial in order to retrieve raisin				
62	Uses a stick to try to attain an object out of reach				
63	Uses trial-and-error approach to precisely imitate new sounds, words, or movements				

19-23 Months

64	Deduces location of object from indirect visual cues (invisible displacements)				
65	Anticipates trajectory by detouring around object				
66	Imitates sounds, words, or body movements immediately and exactly without practicing				
67	Indicates knowledge of cause-effect relationships				

24-29 Months

68	Matches colored blocks (red, yellow, blue, green, black)				
69	Pretends to be engaged in familiar activities (being asleep, telephoning)				
70	Understands concept of one				

30-36 Months

71	Repeats two digits				
72	Matches four shapes (circle, square, star, cross)				
73	Identifies objects by their use (car, penny, bottle)				

Name ______________________

Language

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
	0-2 Months				
74	Moves limbs, head, eyes in response to voice, noise				
	3-5 Months				
75	Vocalizes when talked to or sung to				
76	Turns head in direction of voices and sounds				
77	Vocalizes emotions, intonation patterns				
78	Exhibits differentiated crying				
	6-8 Months				
79	Imitates speech sounds				
80	Forms bisyllabic repetitions (ma-ma, ba-ba)				
	9-11 Months				
81	Waves or claps when only verbal cue is given				
82	Imitates nonspeech sounds (click, cough)				
83	Inhibits activity in response to *no*				
	12-15 Months				
84	Uses appropriate intonation patterns in jargon speech				
85	Imitates words inexactly				
86	Uses two words meaningfully				
87	Uses gestures and other movements to communicate				
	16-19 Months				
88	Shows body parts, clothing items, or toys on verbal request				
89	Labels one object				
90	Follows two simple directions				
91	Uses single words to express wants				
92	Points to several body parts (on self or doll)				

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Language

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
93	Names one black and white picture				
94	Selects two of three common objects				

20-23 Months

95	Begins using simple two-word sentences				

24-27 Months

96	Uses first name when referring to self				
97	Uses three-word sentences				
98	Uses negation, *no*				
99	Uses simple pronouns (I, me, you, mine)				
100	Labels at least three common objects or pictures				

28-31 Months

101	Demonstrates an understanding of two prepositions				
102	Understands body part functions				

32-35 Months

103	Says first and last names				
104	Demonstrates an understanding of three prepositions				
105	Forms questions spontaneously using a verb				
106	Follows two-step commands				
107	Forms or uses plurals				

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Name ______________________

Social/Emotional

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*

0-2 Months

108	Quiets when picked up				
109	Quiets to face and voice				
110	Maintains brief periods of eye contact during feeding				
111	Smiles or vocalizes to talk and touch				

3-5 Months

112	Reflects adult's smile without verbalizing				
113	Laughs				
114	Cries when left alone or put down				
115	Shows awareness of strange environments				
116	Reaches to familiar people (discriminates strangers)				
117	Likes physical play				
118	Smiles spontaneously				
119	Smiles at image in mirror				
120	Watches adult walk across room				

6-8 Months

121	Laughs at pat-a-cake and peek-a-boo games				
122	Withdraws or cries when stranger approaches				
123	Shows dislike when familiar toy is removed				
124	Pats and touches mirror image				

9-11 Months

125	Shows discomfort when separated from mother in strange environment				
126	Participates in pat-a-cake and peek-a-boo games				
127	Repeats vocalizations or activity when laughed at				
128	Offers toy but does not release				

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Social/Emotional

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
	12-15 Months				
129	Offers and releases toy to adult				
130	Initiates ball play or social games				
	16-19 Months				
131	Uses mother as secure base, checking back with her frequently				
	20-23 Months				
132	Occasionally plays near other children				
133	Often clings to or pushes away adult				
134	Cries when preferred activity is blocked				
135	Picks up and puts away toys on request				
	24-27 Months				
136	Independently chooses toy and begins to play				
137	Prefers to play near, but not with, other children				
138	Mimics domestic activities				
	28-31 Months				
139	Stares at or points to sexual differences				
	32-35 Months				
140	Separates from mother easily in strange environment				
141	Identifies own sex				
142	Begins to understand taking turns				

Self-care

Name ____________________

Feeding Skills

Item Number	DEVELOPMENTAL LEVELS AND ITEMS	Date	Date	Date	Date
	3-5 Months				
143	Sucks and swallows pureed foods from spoon				
144	Integration of rooting reflex				
145	Coordinates sucking, swallowing, and breathing				
146	Anticipates feeding with increased activity				
147	Gums or mouths pureed food				
148	Integration of bite reflex				
	6-8 Months				
149	Gums and swallows textured food				
150	Closes lips on spoon to remove food				
151	Drinks from cup with help				
152	Begins to pick up spoon				
153	Begins chewing movements with appropriate tongue motion				
154	Holds bottle to drink				
	9-11 Months				
155	Finger feeds small pieces of food				
156	Holds cookie				
157	Bites cookie				
158	Chews cookie				
159	Licks food off spoon				
160	Eats mashed table foods				
161	Ceases drooling				
	12-15 Months				
162	Feeds self with spoon (many spills)				
163	Picks up and drinks from cup (some spilling)				
164	Chews well				
	16-19 Months				
165	Drinks from cup without assistance				
166	Eats with spoon independently (entire meal)				
167	Discriminates edibles				
	20-23 Months				
168	Unwraps candy; peels or pits fruit				

Developmental Programming for Infants and Young Children
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Self-care

Item Number	DEVELOPMENTAL LEVELS AND ITEMS	Date	Date	Date	Date
	24-35 Months				
169	Begins to use fork				
170	Gets drink without help				
171	Spoon feeds (no spilling)				
Toileting Skills	**12-15 Months**				
172	Remains dry for 1 to 2 hour periods				
173	Fusses to be changed				
	16-23 Months				
174	Bowel movements are regular				
175	Toilet training begins				
	24-35 Months				
176	Uses gestures or words to indicate need to toilet				
177	Toilets independently except for wiping				
178	Seldom has bowel accidents				
Dressing/Hygiene Skills	**12-15 Months**				
179	Pulls off hat, socks, or mittens on request				
180	Cooperates in diapering and dressing by moving limbs				
181	Attempts to brush hair				
	16-19 Months				
182	Imitates simple grooming actions with various objects, i.e., toothbrush, comb, washcloth, with little attempt to groom				
	20-23 Months				
183	Undresses completely except for fastenings				
184	Attempts to put shoes on				
185	Unzips and zips large zipper				
	24-31 Months				
186	Puts on simple clothes without assistance (hat, pants, shoes, etc.)				
187	Washes and dries hands with assistance				
	32-35 Months				
188	Dries hands independently				
189	Puts on coat, dress, T-shirt except for buttoning				
190	Undoes large buttons, snaps, shoelaces deliberately				

Name ______________________

Gross Motor

Item Number	DEVELOPMENTAL LEVELS AND ITEMS	Date	Date	Date	Date
	0-2 Months				
191	Prone: turns head to either side				
192	Neck righting				
193	Upright: head bobs but stays erect				
194	Upright: negative support reaction (integration of stepping reflex)				
195	Prone: labyrinthine righting				
196	Prone: optical righting				
197	Prone: raises and maintains head at 45°				
198	Supine: kicks feet alternately				
	3-5 Months				
199	Integration of Moro reflex				
200	Prone: head and chest are raised to 90° with forearm support				
201	Upright: bears small fraction of weight on feet				
202	Prone: props with extended arms				
203	Pulled to sitting with no head lag				
204	Pulls self to sitting				
205	Prone: rolls to supine				
206	Prone: integration of TLR				
207	Supine: integration of TLR				
208	Prone: integration of STNR				
209	Supine: integration of ATNR				
	6-8 Months				
210	Sitting: trunk erect in chair				
211	Upright: extends legs and takes large fraction of weight				
212	Sits alone for at least 5 seconds				
213	Supine: lifts head spontaneously				
214	Integration of neck righting				

Gross Motor

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
215	Body on body righting begins				
216	Supine: labyrinthine righting				
217	Supine: optical righting				
218	Prone: Landau reflex				
219	Sitting: protective extension to the front				
220	Parachute reaction				
221	Prone and supine: equilibrium reactions				
222	Sitting: labyrinthine righting when tipped to sides				
223	Sitting: optical righting when tipped to sides				
224	Supine: rolls to prone				
225	Prone: pivots				
226	Prone: crawls				
227	Sitting: protective extension to the sides				
228	Standing: moves body up and down				
229	Sits unsupported for 60 seconds				
230	Prone or sitting: assumes quadruped position				
231	Quadruped: equilibrium reactions				
232	Sitting: assists in pulling upright				
233	Prone or quadruped: assumes sitting position				
234	Standing: raises one foot (attempts to step)				

9-11 Months

235	Quadruped: creeps				
236	Sitting: protective extension to the rear				
237	Sits alone and steady 10 minutes				
238	Sitting: pulls to standing using furniture				
239	Standing: lowers self to floor				
240	Standing: cruises by holding onto furniture				
241	Walks with one hand held				
242	Sitting: equilibrium reactions				
243	Stands alone				

Item Number	DEVELOPMENTAL LEVELS AND ITEMS	Date	Date	Date	Date
	12-15 Months				
244	Walks by himself				
245	Creeps up stairs				
246	Standing: throws ball with some cast				
247	Walks well (stops, starts, turns)				
248	Supine: raises self to standing position independently				
249	Walks backward				
	16-19 Months				
250	"Runs" stiffly				
251	Walks sideways				
252	Walks up stairs held by one hand				
253	Creeps backward down stairs				
254	Standing: seats self in small chair				
255	Climbs into adult-size chair				
256	Standing: balances on one foot with help				
257	Standing: equilibrium reactions				
	20-23 Months				
258	Walks down stairs with one hand held				
259	Squats in play; resumes standing position				
260	Jumps in place				
	24-27 Months				
261	Goes up and down stairs alone nonreciprocally				
262	Stands on balance beam with both feet; attempts to step				
263	Kicks ball				
264	Jumps from bottom step (both feet together)				
	28-31 Months				
265	Walks on tiptoes				
266	Throws ball 5 to 7 feet in a vertical pattern				

Gross Motor

Item Number	*DEVELOPMENTAL LEVELS AND ITEMS*	*Date*	*Date*	*Date*	*Date*
267	Takes a few alternate steps on balance beam				
268	Supine: rises to standing with mature pattern				

32-35 Months

269	Rides tricycle using pedals				
270	Goes up stairs alternating feet				
271	Stands on one foot and balances				
272	Walks with heel-toe gait				
273	Walks with reciprocal arm swing				
274	Runs				

Profile Graph

Name ____________________ Birth Date ____________

Evaluation Dates ______________________________

Developmental Level in Months	*Perceptual/ Fine Motor*	*Cognition*	*Language*	*Social/ Emotional*	*Self-care*			*Gross Motor*
					Feeding	Toileting	Dressing	
32-35	40* 39	73	107 ↑ 103	142 ↑ 140	171		190 ↑ 188	274 ↑ 269
28-31	38	72 71	102 101	139	170	178	187	268 ↑ 265
24-27	37 36	70 ↑ 68	100 ↑ 96	138 ↑ 136	169	177 176	186	264 ↑ 261
20-23	35 ↑ 30	67 ↑ 64	95	135 ↑ 132	168	175	185 ↑ 183	260 ↑ 258
16-19	29 ↑ 26	63 ↑	94 ↑ 88	131	167 ↑ 165	174	182	257 ↑ 250
12-15	25 ↑ 21	↑ 58	87 ↑ 84	130 129	164 ↑ 162	173 172	181 ↑ 179	249 ↑ 244
9-11	20 ↑ 16	57 ↑ 52	83 ↑ 81	128 ↑ 125	161 ↑ 155			243 ↑ 235
6-8	15 ↑ 11	51 ↑ 46	80 79	124 ↑ 121	154 ↑ 149			234 ↑ 210
3-5	10 ↑ 2	45 ↑ 42	78 ↑ 75	120 ↑ 112	148 ↑ 143			209 ↑ 199
0-2	1	41	74	111 ↑ 108				198 ↑ 191

*Profile item numbers

Developmental Programming for Infants and Young Children
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Appendix A.4

Parent Survey # 2

3 Months

FAMILY CODE NUMBER _________

DATE _______________________

2

PARENT SURVEY

THE PURPOSE OF THIS QUESTIONNAIRE IS TO IDENTIFY THE EFFECTS THE BIRTH OF AN INFANT WITH SPINA BIFIDA (MYELODYSPLASIA) HAS ON A FAMILY AND TO IDENTIFY WHAT FAMILIES EXPECT OF PROFESSIONALS IN THE CARE OF THEIR CHILD. YOUR COMPLETION OF THE QUESTIONNAIRE MAY GIVE US SOME SUGGESTIONS ABOUT PROVIDING THE TYPE OF ASSISTANCE THAT MAY BE HELPFUL TO YOU AS A PARENT OF A CHILD WITH MYELODYSPLASIA.

PART 1

THE FIRST SERIES OF QUESTIONS ASKS SOME BACKGROUND QUESTIONS ABOUT YOU. PLEASE CHECK THE APPROPRIATE RESPONSE OR FILL IN THE EXACT NUMBERS AS INDICATED.

1. What is your relationship to the child with myelodysplasia?

_____ Mother
_____ Father

2. What is the year of <u>your</u> birth? Give exact numbers. _________ year

3. What category most closely describes your occupation? (Check only <u>ONE</u>)(13)

_____ Unskilled worker (1)
_____ Semi-skilled worker (2)
_____ Skilled worker (3)
_____ Professional (4)
_____ Owner of business (5)
_____ Work in business (specify)(6) _______________
_____ Home and family (7)
_____ Other (specify) (8)

4. Are you employed now? (14)

_____ Yes (1)
_____ No (2)

5. If yes: _____ Full time
_____ Part time
_____ Other (specify)

6. What is the highest educational level you have completed? (Check only <u>ONE</u>) (16)

_____ 8th grade or less (1)
_____ 9-11 grade (2)
_____ High school(3)
_____ 1-3 years post high school(4)
_____ Bachelor's degree (5)
_____ Degree(s) beyond Bachelors (6)
_____ Other (specify)(7) __________

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7. What is your present marital status? (Check only <u>ONE</u>) (17)

_____ Married to the father/mother of child with myelodysplasia (1)
_____ Married but not to father/mother of of child with myelodysplasia (2)
_____ Single (3)
_____ Single but living with father/mother of child with myelodysplasia(4)
_____ Divorced (5)
_____ Separated (6)

8. What is the total number of <u>pregnancies</u> you or your spouse have had? (18-19)

Exact number _________

9. What is the total number of living children you have now? (20-21)

Exact number _________

10. How many children do you have living <u>at home</u> at the present time? (22-23)

Exact number _________

11. Are there persons other than your spouse and children living with you in your home? (24)

_____ Yes (1)
_____ No (2)
Exact Number _____ (25-26)

12. If ves, state their relationship to you. (Check as many as necessary)

_____ Your relatives (27)
_____ Spouse's relatives 28)
_____ Non-related adult(s) (over 18 years of age) (29)
_____ Non-related child(ren) (under 18 years of age) (30)

13. For each item circle the number indicating how you rate your home environment now.

	Low				High	
Room (space)	1	2	3	4	5	(33)
Heat/cooling	1	2	3	4	5	(34)
Food	1	2	3	4	5	(35)
Safety	1	2	3	4	5	(36)
Distance to stores	1	2	3	4	5	(37)
Clothing	1	2	3	4	5	(38)

14. How many of your adult relatives (children, parents, aunts, uncles, first cousins, sisters, brothers, grandparents) live within 50 miles of your home? (Check only one) (39)

____ No relatives (1)
____ 1-9 relatives (2)
____ 10 or more (3)
____ Don't know how many (4)

15. Have you moved in the months since the birth of the child with myelodysplasia? (Check only one) (40)

____ Yes (1)
____ No (2)

16. Have you or your spouse been in the hospital in the months since the birth of the child with myelodysplasia? (Check only one) (41)

____ Yes (1)
____ No (2)

17. Have any of your other children been in the hospital in the months since the birth of the child with myelodysplasia? (Check only one) (42)

____ Not applicable (no other children)(8)
____ Yes (1)
____ No (2)

18. Have any relatives (other than your children or spouse) and/or close friends been in the hospital in the months since the birth of the child with myelodysplasia? (Check only one) (43)

____ Yes (1)
____ No (2)

19. Have you experienced the death of a close friend or close relative in the months since the birth of your child with myelodysplasia? (Check only one) (44)

____ Yes (1)
____ No (2)

20. Since the birth of your child, when you spend time away from your immediate family (spouse and children), was this time spent with (Check only one most common). (45)

____ Other family and/or friends (1)
____ Work and/or school associates (2)
____ Alone (3)
____ Do not spend time away from (4)
immediate family (5)

21. How would you rate your physical health now? (Check only one) (46)

____ Poor (1)
____ Fair (2)
____ Good (3)
____ Excellent (4)

22. How would you rate your emotional health now? (Check only one) (47)

____ Poor (1)
____ Fair (2)
____ Good (3)
____ Excellent (4)

PART II

THE FOLLOWING QUESTIONS ARE RELATED TO YOUR CHILD WITH MYELODYSPLASIA.

1. What is the situation of your child with myelodysplasia? (49)

_____ cared for in our home (1)
_____ cared for in nursing home (or state home) (2)
_____ cared for in own home and nursing home (3)
_____ cared for in other relative's home (4)
_____ cared for in foster home (5)
_____ cared for by adoptive home (6)
_____ deceased (7)
_____ other (specify)(8)_______________

2. To take your child to appointments and/or to visit your child do you (Check the one most common) (50)

_____ drive the family car (1)
_____ your spouse drives the family car (2)
_____ ride with friends and/or relatives (3)
_____ ride city bus (4)
_____ ride taxi (5)
_____ ride special medical transport van (6)
_____ do not visit or take to appointments(7)
_____ Other (please specify)(8)__________

3. The hospital and doctor costs for an infant with a birth defect can be worrisome. What effect are these costs having on your finances? (51)

_____ It is having no effect as all costs are covered (1)
_____ It has a little effect as most costs are covered (2)
_____ It has a large effect as many costs are not covered (3)
_____ I don't know the effect (4)

4. Of the following services (persons), check those that you remember talking to and/or seeing about your child.

_____ hospital nurse (53)
_____ obstetrician (54)
_____ pediatrician (55)
_____ public health nurse (visiting nurse)56)
_____ urologist doctor (kidneys&bladder)(57)
_____ orthopedic doctor (legs and bones)(58)
_____ neurosurgeon (back and head)(59)
_____ clinic nurse (60)
_____ early intervention school program (61)
_____ physical therapist (62)
_____ occupational therapist (63)
_____ social worker (64)
_____ orthotist (brace man) (65)
_____ religious advisor (pastor, priest, religious counselor) (66)
_____ speech therapist (67)
_____ opthamologist (eye doctor) (68)
_____ parent from parent group (69)
_____ none(70)
_____ Other (specify)(71)_______________

5. Give the number of the above people who came to your home at least once (74-75)

__________. Don't know __________

6. How would you rate the time you spend on the following activities concerning your child with myelodysplasia. Circle the number for each activity.

	None	Little				Much
(11) Special care (crêde, exercises, etc.)	0	1	2	3	4	5
(12) Playing with	0	1	2	3	4	5
(13) Caring for (feeding, bathing, etc.)	0	1	2	3	4	5
(14) Worrying about	0	1	2	3	4	5
(15) Enjoying	0	1	2	3	4	5
(16) Taking to appointments	0	1	2	3	4	5
(17) Talking with your spouse	0	1	2	3	4	5
(18) Talking with relatives	0	1	2	3	4	5
(19) Trying to get babysitters	0	1	2	3	4	5

PART III

FOR THE FOLLOWING QUESTIONS, PLEASE CIRCLE THE NUMBER ON THE SCALE WHICH REPRESENTS HOW YOU FEEL NOW ABOUT THE QUESTIONS BEING RATED.

PLEASE TRY TO ANSWER ALL SCALES.

1. The amount of talk with your friends and/or relatives regarding your concerns and problems.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (20)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (21)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (22)
2. The amount of time you spend with your spouse.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (23)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (24)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (25)
3. The amount of discussion of your concerns and problems with your spouse.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (26)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (27)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (28)
4. The amount of time you spend with neighbors.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (29)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (30)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (31)
5. The amount of time you spend in leisure/recreational activities.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (32)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (33)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (34)
6. The amount of help from your spouse with family tasks such as care of children, house repairs, household chores, etc.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (35)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (36)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (37)
7. The amount of help from relatives (do not include spouse) with family tasks such as care of children, house repairs, household chores, etc.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (38)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (39)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (40)
8. The amount of time with health professionals (doctors, nurses, social workers, etc.) related to your child with myelodysplasia.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (43)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (44)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (45)

9. The amount of help from your friends with family tasks such as care of children, house repairs, household chores, etc.

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much (46)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much (47)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much (48)

10. If you don't have other children, check here_______ and omit questions, 11,12,13, & 14.

11. The amount of problems with your other children.

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much (49)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much (50)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much (51)

12. The amount of time you spend with your other children.

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much (52)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much (53)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much (54)

13. If none of your children are in school, check here____ and omit question 14.

14. The amount of time your other children miss school.

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much (55)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much (56)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much (57)

15. The amount of disagreements with your spouse.

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much(11)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much(12)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much(13)

16. The amount of time you are ill.

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much(14)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much(15)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much(16)

17. The amount of time you spend doing housework (cooking, cleaning, washing, yardwork, etc.).

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much(17)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much(18)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much(19)

18. The amount of time you miss work (including housework).

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much(20)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much(21)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much(22)

19. The amount of time your spouse misses work. (including housework).

   a. How much is there now?
   Little 1 2 3 4 5 6 7 much(23)

   b. How much should there be?
   Little 1 2 3 4 5 6 7 much(24)

   c. How important is this to me?
   Little 1 2 3 4 5 6 7 much(25)

20. The amount of emotional support from friends and/or relatives.
    - a. How much is there now?
      Little 1 2 3 4 5 6 7 much (26)
    - b. How much should there be?
      Little 1 2 3 4 5 6 7 much (27)
    - c. How important is this to me?
      Little 1 2 3 4 5 6 7 much (28)

21. The amount of emotional support from your spouse.
    - a. How much is there now?
      Little 1 2 3 4 5 6 7 much (29)
    - b. How much should there be?
      Little 1 2 3 4 5 6 7 much (30)
    - c. How important is this to me?
      Little 1 2 3 4 5 6 7 much (31)

22. The amount of satisfaction with your marriage.
    - a. How much is there now?
      Little 1 2 3 4 5 6 7 much (32)
    - b. How much should there be?
      Little 1 2 3 4 5 6 7 much (33)
    - c. How important is this to me?
      Little 1 2 3 4 5 6 7 much (34)

23. The amount of satisfaction with the sexual relations with your spouse.
    - a. How much is there now?
      Little 1 2 3 4 5 6 7 much (35)
    - b. How much should there be?
      Little 1 2 3 4 5 6 7 much (36)
    - c. How important is this to me?
      Little 1 2 3 4 5 6 7 much (37)

24. What is most difficult for you now? (38)

25. What is most helpful for you? (39)

THANK YOU FOR YOUR TIME IN COMPLETING THIS QUESTIONNAIRE.

IF YOU HAVE ANY QUESTIONS OR COMMENTS, PLEASE FEEL FREE TO ASK THEM OF THE INTERVIEWER OR WRITE THEM IN THE FOLLOWING SPACE.

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1977-80

Appendix A.5

Parent Survey Form A

3 Months

FAMILY CODE NUMBER _____________
DATE ____________________
FATHER ____ MOTHER ____
FORM A

PART I

THE NEXT SEVERAL QUESTIONS ARE RELATED TO YOUR ACTIVITIES AND CONCERNS IN THE YEAR (12 MONTHS) PRIOR TO THE BIRTH OF THE CHILD WITH MYELODYSPLASIA. TRY TO RECALL AS BEST YOU CAN.

1. What category most closely described your occupation? (Check only ONE) (13)

_____ Unskilled worker (1)
_____ Semi-skilled worker (2)
_____ Skilled worker (3)
_____ Professional (4)
_____ Owner of business (5)
_____ Worked in business (specify) (6) ____________________
_____ Home and family (7)
_____ Other (specify) (8)

2. Were you employed prior to the birth of your child with myelodysplasia? (14)

_____ Yes (1)
_____ No (2)

3. If yes: (15) _____ Full time (1)
_____ Part time (2)
_____ Other (specify) (3)
_____ Not applicable

4. What was the highest educational level you had completed? (Check only ONE) (16)

_____ 8th grade or less (1)
_____ 9-11 grade (2)
_____ High school (3)
_____ 1-3 years post high school (4)
_____ Bachelor's degree (5)
_____ Degree(s) beyond Bachelors (6)

5. Were there persons other than your spouse and children living with you in your home? (24)

_____ Yes (1)
_____ No (2)
Exact Number _____ (25-26)

6. If yes, state their relationship to you. (Check as many as necessary)

_____ Your relatives (27)
_____ Spouse's relatives (28)
_____ Non-related adult(s) (over 18 years of age) (29)
_____ Non-related child(ren) (under 18 years of age) (30)

7. For each item circle the number indicating how you rated your home environment.

	Low				High	
Room (space)	1	2	3	4	5	(33)
Heat/cooling	1	2	3	4	5	(34)
Food	1	2	3	4	5	(35)
Safety	1	2	3	4	5	(36)
Distance to stores	1	2	3	4	5	(37)
Clothing	1	2	3	4	5	(38)

8. How many of your adult relatives (children, parents, aunts, uncles, first cousins, sisters, brothers, grandparents) lived within 50 miles of your home? (39)

_____ No relatives (1)
_____ 1-9 relatives (2)
_____ 10 or more (3)
_____ Don't know how many (4)

9. Have you moved in the year (12 months) prior to the birth of the child with myelodysplasia? (Check only ONE) (40)

_____ Yes (1)
_____ No (2)

10. Have you or your spouse been in the hospital in the year (12 months) prior to the birth of the child with myelodysplasia? (41)

_____ Yes (1)
_____ No (2)

11. Have any of your children been in the hospital in the year (12 months) prior to the birth of the child with myelodysplasia? (42)

_____ Not applicable (no other children)(8)
_____ Yes (1)
_____ No (2)

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12. Have any relatives (other than your children or spouse) and/or close friends been in the hospital in the year (12 months) prior to the birth of the child with myelodysplasia? (43)

_____ Yes (1)
_____ No (2)

13. Did you experience the death of a close friend or close relative in the year prior to the birth of your child with myelodysplasia? (44)

_____ Yes (1)
_____ No (2)

14. Prior to the birth of your child, when you spent time away from your immediate family (spouse and children), was this time spent with (Check MOST common) (45)

_____ Other family/or friends (1)
_____ Work/or school associates (2)
_____ Alone (3)
_____ Did not spend time away from immediate family (4)

15. Before your child was born, how would you rate your physical health? (Check only ONE) (46)

_____ Poor (1)
_____ Fair (2)
_____ Good (3)
_____ Excellent (4)

16. Before your child was born, how would you rate your emotional health? (Check only ONE) (47)

_____ Poor (1)
_____ Fair (2)
_____ Good (3)
_____ Excellent (4)

PART II

LISTED BELOW ARE SOME QUESTIONS RELATED TO THE TIME BEFORE THE BIRTH OF YOUR CHILD. PLEASE CIRCLE THE NUMBER ON THE SCALE WHICH REPRESENTS HOW YOU FEEL ABOUT THE QUESTIONS BEING RATED.

PLEASE TRY TO ANSWER ALL SCALES.

1. The amount of talk with your friends and/or relatives regarding your concern and problems.

a. (20) How much was there?
Little 1 2 3 4 5 6 7 much

b. (21) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (22) How important was this to me?
Little 1 2 3 4 5 6 7 much

2. The amount of time you spent with your spouse.

a. (23) How much was there?
Little 1 2 3 4 5 6 7 much

b. (24) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (25) How important was this to me?
Little 1 2 3 4 5 6 7 much

3. The amount of discussion of your concerns and problems with your spouse.

a. (26) How much was there?
Little 1 2 3 4 5 6 7 much

b. (27) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (28) How important was this to me?
Little 1 2 3 4 5 6 7 much

4. The amount of time spent with neighbors.

a. (29) How much was there?
Little 1 2 3 4 5 6 7 much

b. (30) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (31) How important was this to me?
Little 1 2 3 4 5 6 7 much

5. The amount of time you spent in leisure/recreational activities.
   - a. (32) How much was there?
     Little 1 2 3 4 5 6 7 much
   - b. (33) How much should there have been?
     Little 1 2 3 4 5 6 7 much
   - c. (34) How important was this to me?
     Little 1 2 3 4 5 6 7 much

6. The amount of help from your spouse with family tasks such as care of children, house repairs, household chores, etc.
   - a. (35) How much was there?
     Little 1 2 3 4 5 6 7 much
   - b. (36) How much should there have been?
     Little 1 2 3 4 5 6 7 much
   - c. (37) How important was this to me?
     Little 1 2 3 4 5 6 7 much

7. The amount of help from relatives (do not include your spouse) with family tasks such as care of children, house repairs, household chores, etc.
   - a. (38) How much was there?
     Little 1 2 3 4 5 6 7 much
   - b. (39) How much should there have been?
     Little 1 2 3 4 5 6 7 much
   - c. (40) How important was this to me?
     Little 1 2 3 4 5 6 7 much

8. The amount of time with health professionals (doctors, nurses, social workers, etc.) related to your health problems.
   - a. (43) How much was there?
     Little 1 2 3 4 5 6 7 much
   - b. (44) How much should there have been?
     Little 1 2 3 4 5 6 7 much
   - c. (45) How important was this to me?
     Little 1 2 3 4 5 6 7 much

9. The amount of help from your friends with family tasks such as care of children, house repairs, household chores, etc.
   - a. (46) How much was there?
     Little 1 2 3 4 5 6 7 much
   - b. (47) How much should there have been?
     Little 1 2 3 4 5 6 7 much
   - c. (48) How important was this to me?
     Little 1 2 3 4 5 6 7 much

10. If you don't have other children, check here ____ and omit 11, 12, 13, and 14.

11. The amount of problems with your other children.
    - a. (49) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (50) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (51) How important was this to me?
      Little 1 2 3 4 5 6 7 much

12. The amount of time you spent with your other children.
    - a. (52) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (53) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (54) How important was this to me?
      Little 1 2 3 4 5 6 7 much

13. If none of your children are in school, check here ____ and omit question 14.

14. The amount of time your other children missed school?
    - a. (55) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (56) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (57) How important was this to me?
      Little 1 2 3 4 5 6 7 much

15. The amount of disagreements with your spouse.
    - a. (11) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (12) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (13) How important was this to me?
      Little 1 2 3 4 5 6 7 much

16. The amount of time you were ill.
    - a. (14) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (15) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (16) How important was this to me?
      Little 1 2 3 4 5 6 7 much

17. The amount of time you spent doing housework (cooking, cleaning, washing, yardwork, etc.)
    - a. (17) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (18) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (19) How important was this to me?
      Little 1 2 3 4 5 6 7 much

18. The amount of time you missed work (including housework).
    - a. (20) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (21) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (22) How important was this to me?
      Little 1 2 3 4 5 6 7 much

19. The amount of time your spouse missed work (including housework).
    - a. (23) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (24) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (25) How important was this to me?
      Little 1 2 3 4 5 6 7 much

20. The amount of emotional support from friends and/or relatives.
    - a. (26) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (27) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (28) How important was this to me?
      Little 1 2 3 4 5 6 7 much

21. The amount of emotional support from your spouse.
    - a. (29) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (30) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (31) How important was this to me?
      Little 1 2 3 4 5 6 7 much

22. The amount of satisfaction with your marriage.
    - a. (32) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (33) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (34) How important was this to me?
      Little 1 2 3 4 5 6 7 much

23. The amount of satisfaction with the sexual relations with your spouse?
    - a. (35) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (36) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (37) How important was this to me?
      Little 1 2 3 4 5 6 7 much

THE FOLLOWING QUESTIONS RELATE TO YOUR PREGNANCY.

1. What was your general feeling about the pregnancy and the baby. (38) ___

_____ Easy pregnancy and thought baby would be all right (1)
_____ Easy pregnancy but felt there was something wrong with baby (2)
_____ Difficult pregnancy but thought the baby would be all right (3)
_____ Difficult pregnancy and thought there was something wrong with baby (4)
_____ Don't know (5)

2. What was your spouse's general feeling about the pregnancy and the baby. (39)

_____ Easy pregnancy and thought baby would be all right (1)
_____ Easy pregnancy but felt there was something wrong with baby (2)
_____ Difficult pregnancy but thought baby would be all right (3)
_____ Difficult pregnancy and thought there was something wrong with baby (4)
_____ Don't know (5)

3. The doctor and hospital costs for a pregnancy can be worrisome. What effect did these costs have on your finances? (Check only ONE) (40)

   _____ It had no effect as all costs were covered by insurance or Medicaid (1)
   _____ It had no effect as we planned for the costs (2)
   _____ It had some effect, as some costs were not covered (3)
   _____ It had a large effect, as many costs were not covered (4)
   _____ Don't know (5)

4. Which pregnancy resulted in the child with myelodysplasia? (41-42)

   Exact number _____

5. Was the pregnancy which resulted in the child with myelodysplasia a planned pregnancy? (43)

   _____ Yes (1)
   _____ No (2)

6. What is the birth date of your child with myelodysplasia?

   _____/ _____/ _____
   Month (44-45) Day Year (46-47)

PART III

THE NEXT SEVERAL QUESTIONS ARE RELATED TO YOUR EXPERIENCES AT THE TIME YOUR CHILD WAS BORN.

1. When were you told your child had a birth defect? (51)

   _____ During pregnancy (specify month) (1) _____
   _____ During labor (2)
   _____ Delivery room (3)
   _____ Within first 24 hours (4)
   _____ 2-3 days (5)
   _____ After 3 days (specify time) (6) _____

2. Who told you your child had a birth defect?

   _____ Doctor who delivered infant (56)
   _____ Pediatrician (57)
   _____ Neurosurgeon (58)
   _____ Nurse (59)
   _____ Spouse (60)
   _____ Other (specify) (61) _____

3. How old was your child when transferred to Children's Hospital? (62)

   _____ Within first 24 hours (1)
   _____ 2 days (2)
   _____ 3 days (3)
   _____ 4 days (4)
   _____ Other (specify) (5) _____
   _____ Not transferred to Children's Hospital (6)

4. What is the distance you traveled from your home to Children's Hospital? (Or to other hospital if not transferred) (63)

   _____ Less than 50 miles (1)
   _____ 51 to 100 miles (2)
   _____ Over 100 miles (3)

5. What were you told regarding having contact with your child while she/he was in the hospital? (64)

   _____ See and care for child as much as you want (1)
   _____ See and care for the child a few times (2)
   _____ See and care for the child once (3)
   _____ Do not see and care for the child at all (4)

6. What were you told to expect regarding your child? (65)

   _____ Defect can be surgically closed (1)
   _____ Defect can be surgically closed but surgery not recommended (2)
   _____ Defect cannot be surgically closed but child will be re-evaluated (3)
   _____ Defect cannot be surgically closed under any circumstances (4)
   _____ Defect cannot be surgically closed, no check-ups needed (5)

7. What were you told were the possibilities for care for your child? (Check all possibilities you were told)

_____ Place the child in a nursing home (66)
_____ Place the child in a State Hospital or institution (67)
_____ Take the child home (68)
_____ Child would not leave the hospital (69)
_____ Place the child in foster care (70)
_____ Place the child for adoption (71)
_____ Other (specify) (72) ______________
____

8. What was most helpful to you the first few weeks after the child was born? (73)

____
____
____

9. What was least helpful to you the first few weeks after the child was born? (74)

____
____
____

10. How would you describe your child's present condition in terms of how you were told it would be? (Check only ONE) (75)

_____ Child has died as told would happen(1)
_____ Child has died but not expected (2)
_____ Child alive and better than told (3)
_____ Child alive and same as told (4)
_____ Child alive and worse than told (5)
_____ Child alive with different problems than told (6)

11. Were you informed of the opportunity to talk with parents of children with the same problems? (11)

_____ Yes (1)
_____ No (2)
_____ Don't remember (3)

12. If yes, how old was your child when you learned about the opportunity to talk with other parents? (12)

_____ Within first 14 days after birth (1)
_____ 15 days to 1 month (2)
_____ 5 weeks to 3 months (3)
_____ Over 3 months (4)
_____ Don't remember (5)

13. If you were informed about other parents, who told you? (Check as many as necessary)

_____ Not informed (13)
_____ Another parent (14)
_____ Social Worker (15)
_____ Doctor (16)
_____ Nurse (17)
_____ Friend (18)
_____ Spouse (19)
_____ Other family member (20)
_____ News media: TV, Radio, Newspaper (21)

14. When did you first talk with another parent of a child with myelodysplasia? (22)

_____ Within the first week (1)
_____ Two weeks to one month (2)
_____ Within the first six months (3)
_____ After first six months (4)
_____ Have not talked with another parent (5)

15. Were you given written information about myelodysplasia (Spina Bifida)? (23)

_____ Yes (1)
_____ No (2)
_____ Don't know (3)
_____ Other (specify) (4) ______________
____

16. Were you informed of the possibility of financial assistance for the special medical expenses with your child? (24)

_____ Yes (1)
_____ No (2)
_____ Don't know (3)

PART IV

LISTED BELOW ARE SOME QUESTIONS RELATED TO YOUR INVOLVEMENT WITH YOUR CHILD AND HEALTH PROFESSIONALS AT THE TIME YOUR CHILD WAS BORN AND THE FIRST FEW DAYS FOLLOWING THE BIRTH. PLEASE CIRCLE THE NUMBER ON THE SCALE WHICH REPRESENTS HOW YOU FEEL ABOUT THE QUESTIONS BEING RATED.

PLEASE TRY TO ANSWER ALL SCALES.

1. The need you had to care for your child at the time of birth.

a. (27) How much was there?
Little 1 2 3 4 5 6 7 much

b. (28) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (29) How important was this to me?
Little 1 2 3 4 5 6 7 much

2. The amount of time you saw your child.

a. (30) How much was there?
Little 1 2 3 4 5 6 7 much

b. (31) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (32) How important was this to me?
Little 1 2 3 4 5 6 7 much

3. The amount of time you touched or held your child.

a. (33) How much was there?
Little 1 2 3 4 5 6 7 much

b. (34) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (35) How important was this to me?
Little 1 2 3 4 5 6 7 much

4. Encouragement from the doctors to see your child.

a. (36) How much was there?
Little 1 2 3 4 5 6 7 much

b. (37) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (38) How important was this to me?
Little 1 2 3 4 5 6 7 much

5. Encouragement from the nurses to see your child.

a. (39) How much was there?
Little 1 2 3 4 5 6 7 much

b. (40) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (41) How important was this to me?
Little 1 2 3 4 5 6 7 much

6. Encouragement from spouse to see your child.

a. (42) How much was there?
Little 1 2 3 4 5 6 7 much

b. (43) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (44) How important was this to me?
Little 1 2 3 4 5 6 7 much

7. Encouragement from family members/friends (other than spouse) to see your child.

a. (45) How much was there?
Little 1 2 3 4 5 6 7 much

b. (46) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (47) How important was this to me?
Little 1 2 3 4 5 6 7 much

8. The opportunity for you as a parent to decide if the child was to have surgery.

a. (48) How much was there?
Little 1 2 3 4 5 6 7 much

b. (49) How much should there have been?
Little 1 2 3 4 5 6 7 much

c. (50) How important was this to me?
Little 1 2 3 4 5 6 7 much

9. The opportunity to decide about placement for your child (i.e., home, hospital, nursing home, institution).
    - a. (51) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (52) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (53) How important was this to me?
      Little 1 2 3 4 5 6 7 much

THE NEXT FEW QUESTIONS ARE RELATED TO THE TIME A FEW WEEKS AFTER THE BIRTH OF YOUR CHILD.

1. The amount of encouragement to care for your child a few weeks after birth.
    - a. (56) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (57) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (58) How important was this to me?
      Little 1 2 3 4 5 6 7 much
2. The need you had to care for your child a few weeks after birth.
    - a. (59) How much was there?
      Little 1 2 3 4 5 6 7 much
    - b. (60) How much should there have been?
      Little 1 2 3 4 5 6 7 much
    - c. (61) How important was this to me?
      Little 1 2 3 4 5 6 7 much
3. During the first few weeks after your child was born, how would you rate your physical health? (Check only ONE) (62)
    ____ Poor (1)
    ____ Fair (2)
    ____ Good (3)
    ____ Excellent (4)
4. During the first few weeks after your child was born, how would you rate your emotional health? (Check only ONE) (63)
    ____ Poor (1)
    ____ Fair (2)
    ____ Good (3)
    ____ Excellent (4)

THANK YOU FOR YOUR TIME IN COMPLETING THIS QUESTIONNAIRE.

IF YOU HAVE ANY QUESTIONS OR COMMENTS, PLEASE FEEL FREE TO ASK THEM OF THE INTERVIEWER OR WRITE THEM IN THE FOLLOWING SPACE.

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W.S.U. College of Nursing
Children's Hospital of Michigan
NIH 00632-01
1977-80

Appendix A.6

Parent Survey # 3, 4, 5

6, 12, 18 Months

FAMILY CODE NUMBER____________
MOTHER____FATHER____
DATE______________
3 4 5

PARENT SURVEY

THE PURPOSE OF THIS QUESTIONNAIRE IS TO IDENTIFY THE EFFECTS THE BIRTH OF AN INFANT WITH SPINA BIFIDA (MYELODYSPLASIA) HAS ON A FAMILY AND TO IDENTIFY WHAT FAMILIES EXPECT OF PROFESSIONALS IN THE CARE OF THEIR CHILD. YOUR COMPLETION OF THE QUESTIONNAIRE MAY GIVE US SOME SUGGESTIONS ABOUT PROVIDING THE TYPE OF ASSISTANCE THAT MAY BE HELPFUL TO YOU AS A PARENT OF A CHILD WITH MYELODYSPLASIA.

PART I

THIS SERIES OF QUESTIONS ASKS SOME BACKGROUND QUESTIONS ABOUT YOU. PLEASE CHECK THE APPROPRIATE RESPONSE OR FILL IN THE EXACT NUMBERS AS INDICATED.

1. **What category most closely describes your occupation?** (Check only ONE)(13)

____Unskilled worker (1)
____Semi-skilled worker (2)
____Skilled worker (3)
____Professional (4)
____Owner of business (5)
____Work in business (specify) (6)
____
____Home and family (7)
____Other (specify) (8)
____

2. **Are you employed now?** (14)

____Yes (1)
____No (2)

3. **If yes:** ____Full time (1) (15)
____Part time (2)
____Other (specify) (3)
____

4. **What is the highest educational level you have completed?** (Check only ONE) (16)

____8th grade or less (1)
____9-11 grade (2)
____high school(3)
____1-3 years post high school (4)
____Bachelor's degree (5)
____Degree(s) beyond Bachelor's (6)
____other(specify)(7)________________

5. **What is your present marital status?** (Check only ONE)(17)

____Married to the father/mother of child with myelodysplasia (1)
____Married but not to father/mother of child with myelodysplasia (2)
____Single (3)
____Single and living with father/mother of child with myelodysplasia (4)
____Divorced (5)
____Separated (6)
____Other (specify)(7)
____

6. **What is the total number of pregnancies you or your spouse have had?** (18-19)

Exact number__________

7. What is the total number of living children you have now? (20-21)

Exact number__________

8. How many children do you have living at home at the present time? (22-23)

Exact number__________

9. **Are there persons other than your spouse and children living with you in your home?** (24)

____Yes (1)
____No (2)
Exact Number ____ (25-26)

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NIH 00632-01

10. If yes, state their relationship to you. (Check as many as necessary)

____ Your relatives (27)
____ Spouse's relatives (28)
____ Non-related adult(s) (over 18 years of age) (29)
____ Non-related child(ren) (under 18 years of age) (30)

11. For each item, circle the number indicating how you rate your home environment now.

	Low				High	
Room (space)	1	2	3	4	5	(33)
Heat/cooling	1	2	3	4	5	(34)
Food	1	2	3	4	5	(35)
Safety	1	2	3	4	5	(36)
Distance to stores	1	2	3	4	5	(37)
Clothing	1	2	3	4	5	(38)

THE NEXT QUESTIONS WILL BE RELATED TO YOUR ACTIVITIES AND CONCERNS SINCE YOU COMPLETED THE LAST QUESTIONNAIRE (3 or 6 MONTHS). TRY TO RECALL AS BEST YOU CAN.

12. How many of your adult relatives (children, parents, aunts, uncles, first cousins, sisters, brothers, grandparents) live within 50 miles of your home? (Check only ONE) (39)

____ No relatives (1)
____ 1-9 relatives (2)
____ 10 or more (3)
____ Don't know how many (4)

13. Have you moved in the last 3-6 months (Check only ONE). (40)

____ Yes (1)
____ No (2)

14. Have you or your spouse been in the hospital in the last 3-6 months (Check only ONE). (41)

____ Yes (1)
____ No (2)

15. Have any of your other children been in the hospital in the last 3-6 months (Check only ONE). (15)

____ Yes (1)
____ No (2)
____ Not applicable (no other children)(8)

16. Have any relatives (other than your children or spouse) and/or close friends been in the hospital in the last 3-6 months (Check only ONE). (43)

____ Yes (1)
____ No (2)

17. Have you experienced the death of a close friend or close relative in the last 3-6 months (Check only ONE). (44)

____ Yes (1)
____ No (2)

18. When you spend time away from your immediate family (spouse and children), is this time spent with (Check most common). (45)

____ Other family/or friends (1)
____ Work/or school associates (2)
____ Alone (3)
____ Do not spend time away from immediate family (4)

19. How would you rate your physical health now (Check only ONE).

____ Poor (1)
____ Fair (2)
____ Good (3)
____ Excellent (4)

20. How would you rate your emotional health now? (Check only ONE). (47)

____ Poor (1)
____ Fair (2)
____ Good (3)
____ Excellent (4)

PART II

THE FOLLOWING QUESTIONS ARE RELATED TO YOUR CHILD WITH MYELODYSPLASIA FOR THE TIME SINCE YOU COMPLETED THE LAST QUESTIONNAIRE (3 OR 6 MONTHS).

1. What is the situation of your child with myelodysplasia? (49)

_____ cared for in our home (1)
_____ cared for in nursing home (or state home) (2)
_____ cared for in own home and nursing home (3)
_____ cared for in other relative's home (4)
_____ cared for in foster home (5)
_____ cared by adoptive home (6)
_____ deceased (7)
_____ other (specify) (8) _____________

2. To take your child to appointments and/or to visit your child do you (Check the one most common). (50)

_____ drive the family car (1)
_____ your spouse drives the family car (2)
_____ ride with friends/or relatives (3)
_____ ride city bus (4)
_____ ride taxi (5)
_____ ride special medical transport van (6)
_____ do not visit or take to appointments (7)
_____ other (please specify) (8) _________

3. The hospital and doctor costs for an infant with a birth defect can be worrisome. What effect are these costs having on your finances? (51)

_____ It is having no effect as all costs are covered
_____ It has a little effect but most costs are covered
_____ It is having a large effect as many costs are not covered
_____ I don't know the effect

4. Of the following services (persons), check those that you remember talking to and/or seeing about your child since you completed the last questionnaire (3 or 6 months).

_____ hospital nurse (53)
_____ obstetrician (54)
_____ pediatrician (55)
_____ public health nurse (visiting nurse)56)
_____ urologist doctor (kidneys&bladder)(57)
_____ orthopedic doctor (legs and bones)(58)
_____ neurosurgeon (back and head)(59)
_____ clinic nurse (60)
_____ early intervention school program (61)
_____ physical therapist (62)
_____ occupational therapist (63)
_____ social worker (64)
_____ orthotist (brace man)(65)
_____ religious advisor (pastor, priest, religious counselor)(66)
_____ speech therapist (67)
_____ opthalmologist (eye doctor)(68)
_____ parent from parent group (69)
_____ none (70)
_____ other (specify)(71) _____________

5. Give the number of the above people who came to your home at least once (74-75)
_____________. Don't know ___________

6. How would you rate the time you spend on the following activities concerning your <u>child with myelodysplasia</u>? Circle the <u>number</u> for <u>each</u> activity.

	None	Little				Much
(11) Special care (crede, exercises, etc.)	0	1	2	3	4	5
(12) Playing with	0	1	2	3	4	5
(13) Caring for (feeding, bathing, etc.)	0	1	2	3	4	5
(14) Worrying about	0	1	2	3	4	5
(15) Enjoying	0	1	2	3	4	5
(16) Taking to appointments	0	1	2	3	4	5
(17) Talking with your spouse	0	1	2	3	4	5
(18) Talking with relatives	0	1	2	3	4	5
(19) Trying to get babysitters	0	1	2	3	4	5

PART III

FOR THE FOLLOWING QUESTIONS, PLEASE CIRCLE THE NUMBER ON THE SCALE WHICH REPRESENTS HOW YOU FEEL NOW ABOUT THE QUESTIONS BEING RATED.

PLEASE TRY TO ANSWER ALL SCALES.

1. The amount of talk with your friends and/or relatives regarding your concerns and problems.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (20)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (21)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (22)
2. The amount of time you spend with your spouse.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (23)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (24)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (25)
3. The amount of discussion of your concerns and problems with your spouse.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (26)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (27)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (28)
4. The amount of time you spend with neighbors.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (29)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (30)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (31)
5. The amount of time you spend in leisure/recreational activities.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (32)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (33)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (34)
6. The amount of help from your spouse with family tasks such as care of children, house repairs, household chores, etc.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (35)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (36)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (37)
7. The amount of help from relatives (do not include spouse) with family tasks such as care of children, house repairs, household chores, etc.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (38)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (39)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (40)
8. The amount of time with health professionals (doctors, nurses, social workers, etc.) related to your child with myelodysplasia.
   - a. How much is there now?
     Little 1 2 3 4 5 6 7 much (43)
   - b. How much should there be?
     Little 1 2 3 4 5 6 7 much (44)
   - c. How important is this to me?
     Little 1 2 3 4 5 6 7 much (45)

9. The amount of help from your friends with family tasks such as care of children, house repairs, household chores, etc.

a. How much is there now?
Little 1 2 3 4 5 6 7 much (46)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (47)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (48)

10. If you don't have other children, check here________and omit questions, 11,12,13, & 14.

11. The amount of problems with your other children.

a. How much is there now?
Little 1 2 3 4 5 6 7 much (49)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (50)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (51)

12. The amount of time you spend with your other children.

a. How much is there now?
Little 1 2 3 4 5 6 7 much (52)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (53)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (54)

13. If none of your children are in school, check here____and omit question 14.

14. The amount of time your other children miss school.

a. How much is there now?
Little 1 2 3 4 5 6 7 much (55)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (56)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (57)

15. The amount of disagreements with your spouse.

a. How much is there now?
Little 1 2 3 4 5 6 7 much (11)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (12)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (13)

16. The amount of time you are ill.

a. How much is there now?
Little 1 2 3 4 5 6 7 much (14)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (15)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (16)

17. The amount of time you spend doing housework (cooking, cleaning, washing, yardwork, etc.).

a. How much is there now?
Little 1 2 3 4 5 6 7 much (17)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (18)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (19)

18. The amount of time you miss work (including housework).

a. How much is there now?
Little 1 2 3 4 5 6 7 much (20)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (21)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (22)

19. The amount of time your spouse misses work. (including housework).

a. How much is there now?
Little 1 2 3 4 5 6 7 much (23)

b. How much should there be?
Little 1 2 3 4 5 6 7 much (24)

c. How important is this to me?
Little 1 2 3 4 5 6 7 much (25)

20. The amount of emotional support from friends and/or relatives.
    a. How much is there now?
    Little 1 2 3 4 5 6 7 much (26)
    b. How much should there be?
    Little 1 2 3 4 5 6 7 much (27)
    c. How important is this to me?
    Little 1 2 3 4 5 6 7 much (28)

21. The amount of emotional support from your spouse.
    a. How much is there now?
    Little 1 2 3 4 5 6 7 much (29)
    b. How much should there be?
    Little 1 2 3 4 5 6 7 much (30)
    c. How important is this to me?
    Little 1 2 3 4 5 6 7 much (31)

22. The amount of satisfaction with your marriage.
    a. How much is there now?
    Little 1 2 3 4 5 6 7 much (32)
    b. How much should there be?
    Little 1 2 3 4 5 6 7 much (33)
    c. How important is this to me?
    Little 1 2 3 4 5 6 7 much (34)

23. The amount of satisfaction with the sexual relations with your spouse.
    a. How much is there now?
    Little 1 2 3 4 5 6 7 much (35)
    b. How much should there be?
    Little 1 2 3 4 5 6 7 much (36)
    c. How important is this to me?
    Little 1 2 3 4 5 6 7 much (37)

24. What is most difficult for you now? (38)

25. What is most helpful for you? (39)

THANK YOU FOR YOUR TIME IN COMPLETING THIS QUESTIONNAIRE.

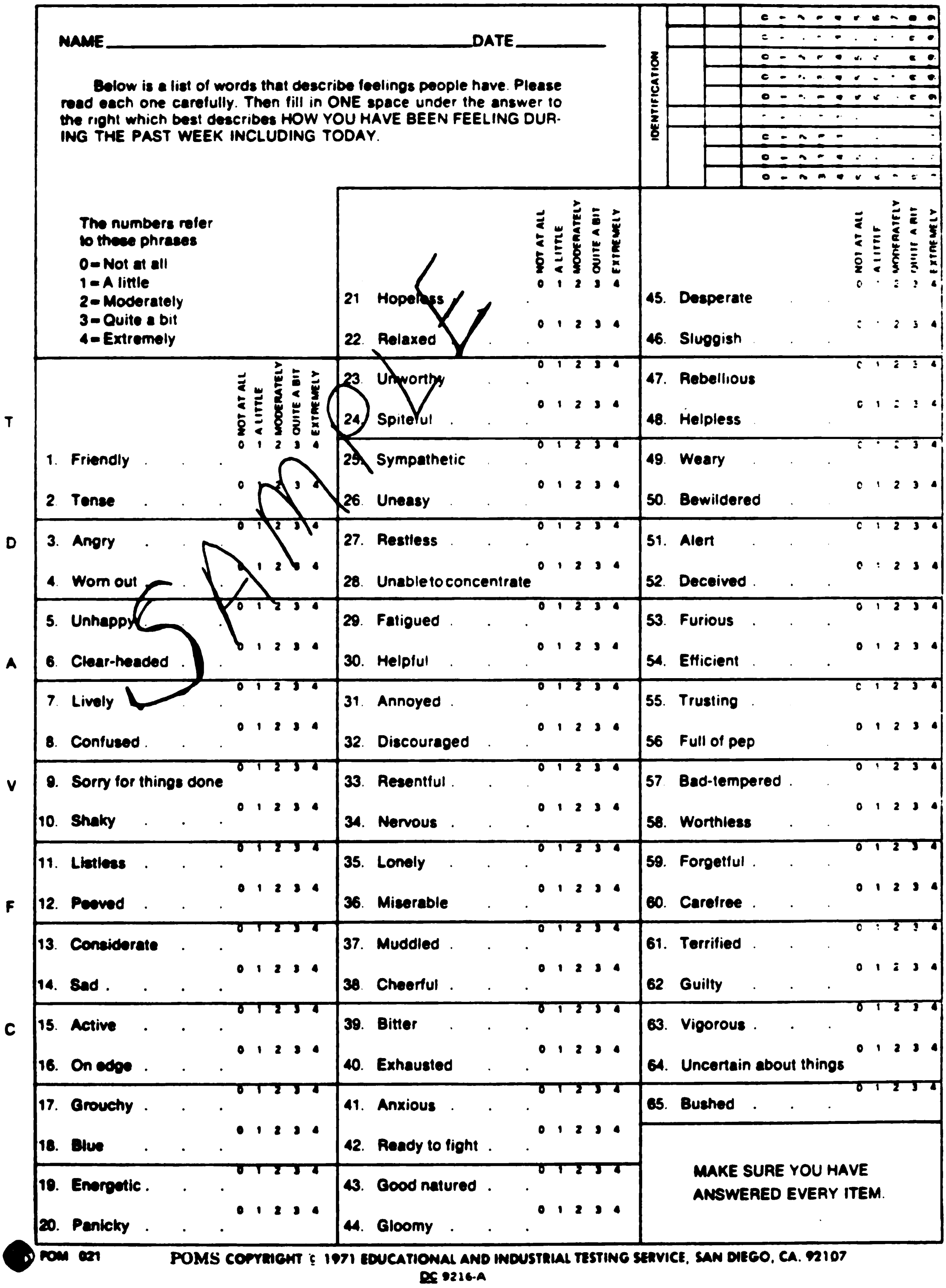
IF YOU HAVE ANY QUESTIONS OR COMMENTS, PLEASE FEEL FREE TO ASK THEM OF THE INTERVIEWER OR WRITE THEM ON THE REVERSE SIDE.

Myelodysplasia Study
W.S.U. College of Nursing
Children's Hospital of Michigan
NIH 00632-01
1977-80

Appendix A.7

Profile of Mood States (POMS)

Neonatal, 3, 6, 12, 18 Months



NAME ________________________ DATE __________

IDENTIFICATION

Below is a list of words that describe feelings people have. Please read each one carefully. Then fill in ONE space under the answer to the right which best describes HOW YOU HAVE BEEN FEELING DURING THE PAST WEEK INCLUDING TODAY.

The numbers refer to these phrases
0 = Not at all
1 = A little
2 = Moderately
3 = Quite a bit
4 = Extremely

NOT AT ALL 0 — A LITTLE 1 — MODERATELY 2 — QUITE A BIT 3 — EXTREMELY 4

T 1. Friendly 0 1 2 3 4
2. Tense 0 1 2 3 4
D 3. Angry 0 1 2 3 4
4. Worn out 0 1 2 3 4
5. Unhappy 0 1 2 3 4
A 6. Clear-headed 0 1 2 3 4
7. Lively 0 1 2 3 4
8. Confused 0 1 2 3 4
V 9. Sorry for things done 0 1 2 3 4
10. Shaky 0 1 2 3 4
11. Listless 0 1 2 3 4
F 12. Peeved 0 1 2 3 4
13. Considerate 0 1 2 3 4
14. Sad 0 1 2 3 4
C 15. Active 0 1 2 3 4
16. On edge 0 1 2 3 4
17. Grouchy 0 1 2 3 4
18. Blue 0 1 2 3 4
19. Energetic 0 1 2 3 4
20. Panicky 0 1 2 3 4

21. Hopeless 0 1 2 3 4
22. Relaxed 0 1 2 3 4
23. Unworthy 0 1 2 3 4
24. Spiteful 0 1 2 3 4
25. Sympathetic 0 1 2 3 4
26. Uneasy 0 1 2 3 4
27. Restless 0 1 2 3 4
28. Unable to concentrate 0 1 2 3 4
29. Fatigued 0 1 2 3 4
30. Helpful 0 1 2 3 4
31. Annoyed 0 1 2 3 4
32. Discouraged 0 1 2 3 4
33. Resentful 0 1 2 3 4
34. Nervous 0 1 2 3 4
35. Lonely 0 1 2 3 4
36. Miserable 0 1 2 3 4
37. Muddled 0 1 2 3 4
38. Cheerful 0 1 2 3 4
39. Bitter 0 1 2 3 4
40. Exhausted 0 1 2 3 4
41. Anxious 0 1 2 3 4
42. Ready to fight 0 1 2 3 4
43. Good natured 0 1 2 3 4
44. Gloomy 0 1 2 3 4

45. Desperate 0 1 2 3 4
46. Sluggish 0 1 2 3 4
47. Rebellious 0 1 2 3 4
48. Helpless 0 1 2 3 4
49. Weary 0 1 2 3 4
50. Bewildered 0 1 2 3 4
51. Alert 0 1 2 3 4
52. Deceived 0 1 2 3 4
53. Furious 0 1 2 3 4
54. Efficient 0 1 2 3 4
55. Trusting 0 1 2 3 4
56. Full of pep 0 1 2 3 4
57. Bad-tempered 0 1 2 3 4
58. Worthless 0 1 2 3 4
59. Forgetful 0 1 2 3 4
60. Carefree 0 1 2 3 4
61. Terrified 0 1 2 3 4
62. Guilty 0 1 2 3 4
63. Vigorous 0 1 2 3 4
64. Uncertain about things 0 1 2 3 4
65. Bushed 0 1 2 3 4

MAKE SURE YOU HAVE ANSWERED EVERY ITEM.

SAMPLE

POM 021

DC 9216-A

APPENDIX B

Description of Study to Precede Consent

To Be Read to Participant

It is known that the birth of a child with a birth defect affects a family in many different ways. The purpose of this study is to learn from families who have had a child with myelodysplasia (spina bifida) and to learn what has happened as a result of this birth. We expect this information will help to improve the care given to the families and children with myelodysplasia.

This study will gather information as to your activities prior to and since the time your child was born, activities at the time the child was born and your current activities and feelings from birth and for 18 months. It will take from 30 to 40 minutes of your time to complete the questionnaire. Information regarding the physical and developmental status of your child with myelodysplasia will also be recorded. The time intervals to complete the questionnaire are at the baby's birth, 3 months, 6 months, 1 year, 18 months.

Your participation in this study is completely voluntary.

The information you provide will be identified by a code number to maintain the confidentiality of your opinions. If you agree to participate, you are free to withdraw your consent and discontinue your participation at any time.

Questions you have about the study will be answered. Do you have any questions at this time?

Informed Consent

The study has been explained to me. I understand that if I agree to participate, I will:

1. Complete a brief form identifying my feelings, taking 10 minutes today, and at 3 months, 6 months, 12 months, and 18 months after my child was born;
2. Complete a questionnaire about my activities and feelings prior to and since the birth of my child with myelodysplasia (spina bifida), taking 20-30 minutes at 3 months, 6 months, 12 months, and 18 months after my child was born; and
3. The hospital records for my child with myelodysplasia will be reviewed for the results of the physical and developmental examinations.

I further understand that:

all information is confidential and my identity will not be revealed,

I am free to withdraw my consent and to discontinue my participation in the project at any time, and

any questions I have about the project will be answered.

On the basis of the above statements, I agree to participate in this project.

participant's signature

date

APPENDIX C

Appendix C

Study Sample

Subjects	Time	1 Neonatal	2 3 months	3 6 months	4 12 months	5 18 months
Parents						
Mothers		27	24	22	25	16
Fathers		19	16	13	12	8
Infants		33	36	30	29	17 *

*Number of subjects at 18 months is lower as study is in progress and results indicate number reaching 18 months of age.

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