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# UNIVERSITY OF SAN DIEGO Hahn School of Nursing and Health Science DOCTOR OF PHILOSOPHY IN NURSING

# FUNCTIONING AMONG TAIWANESE FAMILIES WITH A CHILD HAVING DUCHENNE MUSCULAR DYSTROPHY

By

Jih-Yuan Chen, MSN, RN.

A dissertation presented to the

# FACULTY OF THE HAHN SCHOOL OF NURSING AND HEALTH SCIENCE

# UNIVERSITY OF SAN DIEGO

In partial fulfillment of the

requirements for the degree

# DOCTOR OF PHILOSOPHY IN NURSING

May / 2004

**Dissertation Committee** 

Susan L. Instone, DNSc, RN, CPNP, Chairperson

Diane C. Hatton, DNSc, RN, CS

Allen J. Orsi, Ph.D., RN

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#### ABSTRACT

# FUNCTIONING AMONG TAIWANESE FAMILIES WITH A CHILD HAVING DUCHENNE MUSCULAR DYSTROPHY

A cross-sectional study was designed to determine the factors that affect functioning among Taiwanese families with a child having Duchenne Muscular Dystrophy (DMD). This research investigated the relationships between the degree of a child's disability, family characteristics, family health, family hardiness, family support, and family functioning from a parental perspective. A total sample of 126 parents of children with DMD completed basic demographic information, the Family Assessment Device, the Family Hardiness Index, the Duke Health Profile, and the Family APGAR.

Pearson Correlation Coefficient test was performed to examine relationships between independent and dependent variables. To determine if the levels of child's mobility, family characteristics, family hardiness, family health, and family support had significant impact on the dependent variable (family functioning), the Hierarchical Multiple Regression Model was used and indicated that four variables significantly contributed to the variance in family functioning: access to care (age when diagnose with DMD), family hardiness, family health, and family support. The model as a whole explained 68 % of variance in family functioning ( $R^2$ = .679, F (4, 121) = 64.08, p= .00).

Beta coefficients indicated that the later children were diagnosed with DMD and the lower the parental scores on family hardiness, family health, and family support (less support) were related to poorer family functioning after controlling for the variable differences. This study revealed that the earlier children are detected with DMD, the greater the likelihood their families will have greater hardiness, health, and support, all of which contribute to healthy family functioning. In addition, family hardiness and family support were predictors of family health and the age when the children were diagnosed with DMD and family support were the predictors of family hardiness. The results suggest that health professionals encountering children with early signs of DMD should urged their families to promptly seek evaluation, treatment, and the social support services available to DMD children and their families in Taiwan.

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#### **CHAPTER I: INTRODUCTION**

# Overview

Little is understood about how Taiwanese families function when they have a child with Duchenne Muscular Dystrophy (DMD). Resilience, as well as hardiness, are important qualities found in families coping with other life stressors, and may also be factors in how DMD families function. The sacrifices families must make when a child has DMD - related disabilities are not temporary. Instead they become a way of life for the whole family and often require services from many agencies, which may result in increased financial costs, social isolation, as well as restriction of life-styles and career opportunities (Failla & Jones, 1991; Gottlieb, 1998; Patterson & McCubbin, 1983). The reciprocal impacts on the families are circular and continuous (Patterson, 2002).

# Background and Significance

DMD, the second most common genetic disease in humans, is an X-linked disease of the muscle caused by mutation of the Xp21 gene. This gene encodes a rod like cytoskeletal protein called dystrophin that afflicts only boys who inherit the disease from their mothers (Emery, 1993; Nicholson, 1993). The world wide incidence, based on live male births, is around 200 to  $300 \times 10^{-6}$ , but the mutation rate is approximately 70 to 100 x  $10^{-6}$  (Emery, 1993; Laing, 1993).

As children get older, DMD takes a slow and arduous course that leads to parental strains. The children's emotional responses in the form of problem behavior resulting from social isolation and poor interpersonal skills, has been found to predict maternal

stress and anxiety (Huang & Dai, 1998; Nereo, Fee, & Hinton, 2003; Nereo & Hinton, 2003).

The progression of the children's disabilities induces the family to change and influences the entire family system (Botvin, Radford, & Neumann, 1984; Siegel, Davidson, Kornfeld, & McCready, 1983). Family structure, process, and functioning change the most as a result of the demands on family relationships, activities, and goals of the family social system (Thompson, Zeman, Fanurik, & Sirotkin-Roses, 1992). Families also change roles to meet the demands for achieving positive family functioning (Epstein, Bishop, Ryan, Miller, & Keitner, 1993). Some families adapt and others become depleted of family energy and resources; this difference has received little attention in the pediatric literature (Thompson et al., 1992).

The theoretical and empirical basis of a family-oriented approach has not been widely addressed, even in Taiwan, limiting the efforts of family health and human service providers involved in family health promotion. Health professionals have the responsibility to strengthen the family/child's coping resources and make the children's environment more accommodating to their special needs, as well as to assist their families to enrich their lives through interventions that enhance meaning and satisfaction in caregiving through positive experience and encounters.

The ability to maintain a balance between change and stability has been referred to as a measure of healthy family functioning. When families are able to utilize their strength and abilities, they are able to recover from the stress and challenge and minimize a negative outcome. The resilient family that adapts to stress has a higher level of functioning. Families that function well can solve problems, share affection, and meet the needs of individual family members.

If nursing professionals could evaluate family resilience and discuss interventions to support it, they could promote family functioning. And if the family of a DMD child can describe how they have dealt with the disability and anticipated loss, this might help others in similar situations to deal with their own sense of loss, fatigue, and distress. This study attempts to discover which family characteristics, supports, strengths, resources, and functioning buffer the impact of a stressful life and improve understanding of why some families thrives and other families do not. The findings of this study may contribute to the development of interventions that will help promote resilience in family members living with a child having DMD.

# Statement of the Problem

The progressive disabling condition of DMD creates disruptions in the physical, social, emotional, and spiritual life of the affected child and his family. The chronic stress experienced by these families challenges their coping mechanisms as they adjust and continue to function. Families may experience growth and integration, balance and stability, or disorder and disintegration (Bubolz & Whiren, 1984). For family members, a DMD diagnosis heralds deformity with immobility, creating the need for important social services, such as special education programs, respite programs, and insurance coverage. DMD challenges a family to maintain normal functioning while struggling with loss.

The philosophy and policy trends of normalization and de-institutionalization encourage families to raise children with developmental disabilities at home. As more disabled children stay at home, families must become more diverse in their skills to meet

the challenges and special risks that accompany a DMD diagnosis. In addition, the ability of these families to function at this level must also be viewed in light of a Taiwanese society which is evolving into one that is increasingly multiracial, multicultural, and multilingual (Braverman, 2001; Wisensale, 1993).

Analysis of longitudinal and cross-sectional research on the factors that influence how families' function indicates mixed results. These factors are the severity of the child's disability and family characteristics, health, support, and family hardiness. Several studies suggest that families with psychological problems score lower on family functioning than control groups (Baigas, 2002; Friedmann et al., 1997; Keitner et al., 1991; Keitner, Miller, & Ryan, 1993; Miller, Epstein, Bishop, & Keitner, 1985). Other studies have not supported these findings (Epstein et al., 1993; Kim, 2002) and there has been little information about cultural influences on family functioning (Roncone et al., 1998; Shek, 2002; Stevenson-Hinde & Akister, 1995). Therefore, research is needed to better understand how Taiwanese DMD families function.

#### Purpose of the Study

The overall purpose of this study was to explore the factors associated with functioning among Taiwanese families with a child having DMD. The specific aims to achieve the purposes of this study were as follows:

- 1. Describe the child's level of disability, access to care, and family characteristics.
- 2. Describe family health, family hardiness, family support, and family functioning experienced by the parents with DMD children.
- 3. Describe the relationship among DMD child's level of disability and access to care (age when diagnosed with DMD), family health, family characteristics

(family employment and family annual income), family supports, family hardiness, and family functioning.

- 4. Determine how the child's level of disability and access to care, family health, family characteristics, family support, and family hardiness predicted family functioning in families with a DMD child
- 5. Test the model of Family Stressors, Resources, and Functioning with families with a child with DMD.

# Summary

Little is understood about how families function when they have a child with DMD. Several studies focused on the impact and coping of families with disabled children. Given the research gaps, and lack of information about the functioning of culturally diverse families with a disabled child, this study involved a group of Taiwanese families to discover whether the factors associated with family functioning found in the literature were characteristic of them.

#### CHAPTER II: LITERATURE REVIEW

This literature focuses on Duchenne muscular dystrophy (DMD), the impact of DMD on children's health, the impact of DMD on families, and cultural and religious meaning of DMD in Taiwan. With this foundation, the various conceptual models of family functioning will be examined and critiqued.

# Duchenne Muscular Dystrophy

DMD is a neuromuscular disorder that presents as a chronic progressive disease that is physically incapacitating. It affects only boys and is inherited from their mothers (Emery, 1993). As carriers, women usually show no sign of the disease but they are capable of passing the condition on to their own sons. All affected daughters are carriers and the disease is never transmitted from father to son (Laing, 1993). In two thirds of cases there is a family history of the disorder; the remainder (70 to  $100 \times 10^{-6}$ ) are spontaneous mutations (Emery, 1993; Laing, 1993). Absence of effective treatment for DMD has led to develop new approaches for carrier detection and prenatal diagnosis (Alcantara et al., 2001; Laing, 1993; Wang et al., 2001). Creatinine phosphokinase raised that is the best screen for neonatal diagnosis of DMD (Bradley, Parsons, & Clarke, 1993).

In Taiwan, the risk of a carrier having an affected child is one in four, with an incidence of 1 in 3000 to 1 in 3500 live male births; the mutation rate for DMD is about 1 in 10,000, which is very high in comparison with other genetic disorders (Laing, 1993). From 1981 to 2002, it is estimated that of 3,060,000 live male births there may have been approximately 1028 to 1199 DMD children born in Taiwan assuming each one lives to

the age of 21 (Department of Health, 2001). The amount spent on medical care is over NT\$ five million during the life of a DMD child (TMDA, 2002).

General speaking, children are diagnosed with DMD between 3 and 11 years of age (Appleton & Nicolaides, 1995). The median age at diagnosis is 2 years (Siciliano et al., 1999). Most children are diagnosed with DMD after age two, but before their fifth birthday (Roland, 2000).

## Impact of DMD on Children's Health

Duchenne defined the disease as being characterized by progressive muscular weakness, first affecting the lower limbs and then later the upper limbs. The most obvious features in the early stage are enlargement of the calf muscle (called pseudohypertrophy) which is due to an excess of adipose and connective tissue, and a wadding gait (Emery, 1993). More often mothers notice that there is a delay in their child's learning to walk; in 56% of children with DMD walking was delayed until at least 18 months and roughly a quarter did not walk until they were at least 2 years old. In 90% of cases, the onset was before 5 years old. The affected child was never able to run properly (Emery, 1993). The major symptoms at onset were muscle weakness (31.8%) and falling down easily (31.8%). The onset of illness before age 5 was 36.4% (Chen, Chen, Jong, & Yang, 2002), and losing the ability to climb stairs occurred at a mean age of 9.3 +/- 1.4 years (range, 5.8 to 13.8 years) (Vignos, Wagner, Karlinchak, & Katirji, 1996).

The affected children experience progressive muscle weakness, manifested by difficulty getting from a sitting to a standing position. The first clinical symptoms such as waddling gait, walking unsteadily with a tendency to fall easily, walking on toes,

difficulties of rising off the floor, squatting, and climbing stairs appear between 3 to 6 years of age (Hoffman, Brown, & Kunkel, 1987). In the early stage of the disease, this is evaluated by the Gower's maneuver, where the child tries to stand by using his hands to climb up his thighs, pushing down on them, and extending his hips and trunk in order to stand. An increase in interstitial connective tissue in the affected muscles, with the production of abundant fibrous and adipose issue, appears in the later stage (Emery, 1993).

Patients with DMD become unable to walk between 6 and 12 years, with a mean age of 10 years resulting in wheelchair dependency (Kilmer, Abresch, & Fowler, 1993). As the disease progresses and muscle weakness becomes more profound, the loss of hip extension and ankle dorsiflexion become the primary predictors of an inability to walk (Bakker, de Groot, Beelen, & Lankhorst, 2002). Kyphoscoliosis develops and facial and neck muscles weaken. Eventually, he becomes confined to a wheelchair because of flexion contractures of the elbows, knees, and hips. By twelve, the feet may turn inward and downward (Emery, 2002).

Vignos et al. (1996) found that operative procedures combined with bracing and physical therapy, including daily passive stretching exercises and prescribed periods of standing and walking, were successful in controlling contractures of the lower extremities for as long as seven years after treatment. Their management allowed DMD boys to walk until a mean age of 13.6 years and to stand for an additional two years after the ability to walk with braces had been lost.

In addition, 20 % of the affected boys have an IQ of less than 70 (Emery, 2002). Thus, the disease affects the DMD child's physical strength, school achievement, and

social activities with friends (Lue, Chen, Jong, & Lin, 1993). Most of the DMD children are lonely; they see other children enjoying friendships and an active social life, while their world is more restricted. The lack of physical activity and recreational opportunities can lead to the development of obesity as well as subsequent withdrawal, depression, and isolation (Adrian, 2002). Westernization of Taiwanese society has contributed to a rise in obesity as more people have access to high calorie fast food. In 1990, 25 % of boys and 18 % of girls in elementary school were obese (Lin, 1990). DMD children have a deficit of activities due to their weakened muscles; therefore, it is easier to become obese if they over eat. Their caregivers can exhaust their physical strength by taking care of their obese children.

The clinical definition of DMD includes becoming wheelchair bound by age 12, and death usually by the end of the third decade (Laing, 1993). The muscles that control respiration and cardiac function fail, leading to death. Respiratory failure invariably occurs in the second decade (Rideau et al., 1995). Death occurs by the early 20s or before they reach 25 years of age usually due to a simple cold or complicated pneumonia, and 9-50% die from cardiac failure (Emery, 1993; Parent Project Muscular Dystrophy, 2002). These children may require total care from their families for 6 to 8 years before they die. Survival to age 20 is estimated at 25% (Holroyd & Guthrie, 1986). Thus, the disease's progression has distressing consequences for the children and their families for 15-25 years (Firth, Gardner-Medwin, Hosking, & Wilkinson, 1983).

In a 40-year longitudinal study that evaluated the orthopedic treatment and physical therapy of 144 boys with DMD from 1953-1994, the major causes of death were pulmonary insufficiency (61%), pneumonia (31%), and cardiomyopathy (7%). Ninety-

four percent of the patients had a functional classification of "confined to wheelchair" or "bed" at the time of death. The mean age at the time of death was 18.1 +/- 3.2 years (range, 11.8 to 24.6 years) during the 1960's, 19.0 +/- 2.8 years (range, 13.7 to 27.5 years) during the 1970's, and 18.8 +/- 3.4 years (range, 13.1 to 26.4 years) during the 1980's. Five of these subjects were between thirty-one and thirty-three years old and needed ventilator support (Vignos et al., 1996). Vignos et al (1996) reported "with the numbers available, we could not detect a significant difference among the treatment groups or time-periods with regard to the age at the time of death" (p .1849).

Anecdotal data over eight years from my clinical experience as a pediatric nurse in Taiwan confirms the progressive, debilitating nature of DMD. Most of these children are obese, lonely and living in worlds that are restricted, especially after they graduate from primary school and do not attend high school. Practical problems such as transportation, difficulty with the physical labor of lifting, the need for increased medical attention, and the tremendous financial burden upon the family have all been observed to some degree. How these burdens affect Taiwanese families' ability to function, however, are not well understood.

# Impact of DMD on the Family

# Family Perception of Having a DMD Child

Some studies have indicated that parents are significantly concerned about problems relating to the care of DMD children including the practical problems of daily living, emotional problems, the drain on other personal relationships, and the exclusion of other family needs (Firth et al., 1983; Siegel et al., 1983). About 62% of the parents (N = 65) in one study experienced these problems: lifting, housing, difficultly using public transportation, and concern about what they should tell their sons about the disease (Firth et al., 1983). In another study, the major needs of 61 parents with a DMD child in Taiwan were getting information (71%) about coping with the disease, accessing health care (68%), the progression of disease (65%); and support groups (48%). The major concerns were how to comfort the child and help him be happy (89%), how to maintain a close couple relationship (79%), and how to overcome exhaustion (73%) (Chen, Chen, Jong, Yang, & Lue, 2003).

Some parents are able to accept and adapt to the disability by finding a meaningful pattern of life. Gagliardi (1991) found that the response of families living with a DMD child were characterized by various stages of adaptation. The "recognition stage" lead to "disillusionment" and the realization that "society confirms the impossibility of normalcy". Families then moved to the "work out stage" to adjust to the disability and maintain the "dynamic of the family: who's disabled anyway", to by adjusting to "a smaller world", and then deciding whether to "let go or hang on", with the realization that "things must change" (pp. 162-163 ). The intervention implications of these studies include restructuring psychosocial services for the entire family and providing a network of liaison services to assist the family as the disease progresses. *Family Stress and Family Coping* 

The prior studies indicated that over half of the families had psychological adjustment problems (Buchanan, LaBarbera, Roelofs, & Olson, 1979; Thompson et al., 1992) because of the stressors of their sons' decreasing independent abilities and behavior problems (loneliness and depression), marital conflict, increase in daily chores,

duties, and responsibilities of caring for the child, and lack of support from extended family and school personnel. These problems lead the parents to utilize denial, isolation, magical thinking, or overprotection and to feel a sense of guilt and hopelessness. A study of 112 mothers found that their stress was elevated because of negative behaviors of their DMD children, especially in social interactions (Nereo et al., 2003). On the other hand, 75 % of families reported a positive couple relationship and 72 % of parents with DMD children were satisfied with their marital status (Chen et al., 2002).

The chronic stress of living with a child who has a progressively deteriorating illness often motivates families to seek emotional support from groups and institutions around them. Extended families and schools are the major support systems. But some of the grandparents may blame the disability on in-laws, or the mothers may blame themselves for carrying the defective genes. Arguments focused on how to care for the child, discipline, constant fatigue from the labor involved, and interference from the extended family have been described (Buchanan et al., 1979).

In addition, the children often were placed in special education classes to avoid obvious physical competition with normal boys and to allay parents' fear that their children were being abused in public schools. Some of the parents also believed that some teachers did not understand their children and tended to spoil them, refusing to discipline them and excusing their behavior because of the illness, or expecting more from them than they could physically perform. On the other hand, the use of homebound teachers for these boys contributed to their social isolation (Buchanan et al., 1979).

Parental coping by preserving their own emotional well-being, reducing family conflict, and improving family supportiveness showed a significant positive correlation

with a measure of parent adjustment (Buchanan et al., 1979). Parents with higher monthly incomes who lived in cities were both more aggressive and more afraid of other's criticism. The children's ability to cope was compromised because they were isolated, could not get peer support, and could not freely express their thought and feelings and easily felt rejected (Chen et al., 2003). Another study found that parents with disabled children revealed significantly more avoidant coping, lower sense of coherence, and less emphasis on family members' interrelations and personal growth than did the control group (Margalit, Raviv, & Ankonina, 1992).

## Reaction of Siblings

Siblings may need to change their role to care for their affected brothers. Botvin et al. (1984) found that there may be a tendency for older sisters to adopt a maternal role and to overprotect their ill brother. In their study, sisters tended to be very defensive in response to the actions and attitudes of people outside the family toward their sibling. Siblings may experience some degree of emotional distress and they may need help with feelings of jealousy because the affected child seems to get more attention (Botvin et al., 1984). However, Nereo et al. (2003) reported that stress of DMD children was not significantly different from that of their siblings.

# Disclosure Issues

Fitzpatrick and Barry (1986) found that most parents were unable to discuss the condition with their sons. None of the boys had asked about the progression of their disorder. Some siblings were informed of the affected boy's condition but they were not told about the disease progression. Fitzpatrick et al. (1990) conducted a retrospective case-control study to compare the patterns of communication and use of professional

support systems in Irish and American families with DMD boys. The results indicated that difficulty in communication with their spouse (73% Irish and 24% American) and with their affected sons (94% Irish and 52% American) were reported by significantly more Irish parents than by their American counterparts. More Irish parents (56%) never talked about DMD with their sons.

## Informational Needs of Families

Smith, William, Sibert, and Harper (1990) found that only 68% of the mothers (N=201) in their study were aware that infants could be screened during the neonatal period, and more multiparous than primparous mothers were aware of such screening. Ninety-four percent of these mothers would accept a screening test for DMD, and 75 % would want to know soon after birth whether their babies had a disabling condition. Seventy percent of these mothers would consider termination of their pregnancy for medical reasons. In fact, DNA studies of cultured amniotic fluid cells at 14 weeks gestation, the absence of the X-chromosomal fragment of DXYS19X located in XY21.2-pter or Xp22.3 and analysis of several STR loci of dystrophin, followed by multiplex PCR, lead to the diagnosis of a male fetus affected by DMD; and quantitative multiplex PCR confirmed the deletion in female carriers (Jakubiczka et al., 2000). In addition, Chen et al. (2003) found that families also needed information about physiotherapy, genetic issues, and support groups to prevent them from selecting useless rituals or remedies for their DMD children.

*Summary.* These studies suggest that emotional support, parent education, and other services can improve satisfaction and communication to help families resolve their problems and function better. The data also suggest that little effort has been made to

inform parents about neonatal screening so that early diagnosis and supportive care are not delayed unnecessarily.

Cultural and Religious Meaning of DMD in Taiwan

# Common Cultural and Family Value

By 1970, Taiwan had undergone a transformation from an agricultural society to an industrial and technological country. Many Taiwanese families have become smaller nuclear families while maintaining traditional Chinese spiritual beliefs. These include life philosophies of "giving birth to new life (生生不息 Sheng-sheng buo-hsin )", "unity of heaven and man (天人合一 Tien- Jem houi-yii)", "way of heaven (天道 Tien-tao)", and "way of man (人道 Jem-tao)". This spiritual foundation gives meaning to the lives of Taiwanese families as they are "playing out one's inherent nature" ( 盡性 chin-hsing) (*Traditional Chinese Culture in Taiwan: Philosophy*, 2002, para 4). These beliefs may help parents understand the meaning of their child's illness and help them function by developing hardiness to overcome their gradual loss.

In Chinese societies the son carries the family lineage. There are three things that are unfilial and having no progeny is the greatest of these. Chinese lineage reflects and reinforces structural features of Chinese society and functions to maintain that society (Freedman, 1979). Therefore, having a disabled son who will die prematurely is a severe blow to Taiwanese families.

The values of dragon son / phoenix daughter. According to an idiom of Mandarin "wang nan tso lung, wang nu tso huang," one "hopes their son will become a dragon, and hopes their daughter will become a phoenix." "Dragon in this context means success for the male in any endeavor; phoenix for the female means she will get a good education,

have a good career, and marry a successful man" (Marsh, 1996, p. 284). Parents' attitudes about socializing their children are defined by their ideas about discipline and corporal punishment as a means of bringing children up properly for a successful life. According to Marsh (1996), two thirds of parents believed that their children needed more discipline and thought that in order to bring children up to be fine human beings, physical punishment was sometimes necessary. For families with children with DMD, how to deal with emotional problems without using physical punishment becomes an important challenge.

*The values of harmony and yin-yang.* Harmony, including the concepts of yin and yang and the five basic forces (metal, wood, water, fire, and earth), are important for understanding health and illness in Chinese culture. These concepts imply that human beings and nature are interrelated and interdependent to maintain harmony. To reestablish the harmonious state, traditional Chinese medicine uses herbs and food to correct the disturbance and imbalance in the body systems. People in Taiwan prefer to receive western medicine for treating acute illnesses, but in the recovery stage, they prefer to use traditional Chinese medicine to restore energy and balance in their bodies. The parents of children with DMD also wish to find harmonious therapies for their children.

## Buddhism

According to the Buddhist philosophy of life, life is pain and suffering because of ignorance and desires. Disability is a consequence of deeds done in previous lives and is associated with evil spirits or karma. Therefore, when a son has a disability and suffers an early death, both the child and the family have to tolerate the pain and discomforts and

undergo treatment together. For example, family members go to the temple to get a special charm as a blessing from Buddha to keep away evil spirits, to experience pain or pleasure, and to deal with the positive and/or negative reactions to the child's disability. Most Taiwanese mothers make the sacrifice required to care for their sons if they are sustained by their religion, folk beliefs, or social support. An added burden for them is the cultural expectation that they will also care for their husband's aging parents.

# Changing Demographics in Taiwan

#### Economic Impact and National Health System

Taiwan's ratio of economically active people to the retired has begun to fall, imposing an increasing by onerous burden on the younger working population (Gold, 1996). Since the global economy has declined, a young couple might have difficultly buying a house if they have no support. These financial burden are even more challenging for families with DMD children (Bothwell et al., 2002) although national health insurance covers all residents in Taiwan.

Recently, the government and private organizations have developed daycare centers for working mothers. In addition, there are respite services for DMD children, except in rural areas, but these still need to be better organized. National health insurance has reimbursed home nursing services since 1996. As a result, in-home nursing services have become a rapidly growing health industry. The number of home nursing agencies increased from 27 in 1993 to 125 in 1997 (Long-Term Care Profession Association of ROC, 1997).

# Urbanization

Highly urbanized, often freakishly ugly high-rise apartment blocks and factories are crammed together. There has been little advance in macro-level planning or coordination, posing tremendous problems of utilities, service and green spaces. As a developing society, Taiwan has seen continuous and rapid rural out-migration that tended to rely on and maintain ties with kin in the cities (Greenhalgh, 1984). While an excellent transportation and telecommunications infrastructure has been created for able-bodied persons, public access for the disabled has not been created developed, keeping DMD children and their families socially isolated.

#### Education System

Revolutionary changes in the educational system in Taiwan have made it possible for most people (over 70 %) to enter higher education after senior high school. However, most DMD children have had to drop out of school when parents cannot provide transportation, children cannot pass the entrance examination, or the children's health is poor. It is not enough to supply one-on-one teaching at home for the DMD children, although some cities in Taiwan have systems to teach at home. The quality of life of the children and their families has to be considered when DMD children are permanently absent from the school.

## Summary

In summary, Taiwanese families utilize spiritual beliefs, especially those from Buddhism, to explain their child's disability and cope with the loss of a successful son. In addition, these children and their families remain isolated because of a lack of information, transportation, and support services. Despite these concerns, some families

have maintained positive relationships in the family. In order to better understand what else these families need to function better, several models of family functioning will be explored.

## Family Functioning and the Resiliency Model

# Definition of Family

The family is a complex system of interacting individuals who share a history and a future. Families consist of structures, roles, and functions (McGoldrick & Carter, 2003). Family structure is the number of members of the family; family roles include parents, spouse, child, other kin, etc.; and family functions involve the ability to satisfy members' physical, psychological, survival, and maintenance needs (Smith, 1995).

# Definition of Family Functioning

The basic attributes of family functioning are characterized and explained by how a family system typically appraises, operates, and behaves (McCubbin & Thompson, 1991). Family functioning also includes the ability to solve problems. The ability to maintain a balance between change and stability is another aspect of healthy family functioning (Olson, 1993).

Family functioning is a reliable predictor of parental adjustment and adaptation. Normal functioning refers to the ability to achieve family goals, meet situational and developmental challenges, and adjust to economic circumstances and cultural norms (Walsh, 1993). The important attributes of healthy family functioning include commitment, responsibility, organizational stability, adaptability, communication, problem solving, belief system, and resources (Walsh, 1993). Healthy family functioning does not mean absence of problems, but rather "the healthy family can be found in the midst problems as in family resilience" (Walsh, 2003, p. 5). Therefore, the presence of distress is not necessarily a criterion of family pathology (Epstein et al., 1993).

# Definition of Family Resilience

Resilience is the ability to function well and to be competent when faced with life stress. Resiliency is "the family's ability to use their existing strengths and resources to overcome crises and to react positively to challenges" (Berry, 2004, para 3).

McCubbin, Thompson, and McCubbin (2001) defined resiliency as:

the positive behavioral patterns and functional competence individuals and the family unit demonstrate under stressful or adverse circumstances, which determine the family's ability to recover by maintaining its integrity as a unit, the well being of family members and the family unit. (p. 5)

It is encouraging to note that some families' ability to adapt to stress leads to higher than normal levels of functioning (Patterson, 2002). McCubbin and McCubbin (1988) defined family resilience as "characteristics, dimensions, and properties of families which help families to be resistant to disruption in the face of crisis situations" (p. 247). The institute for Health and Disability (1997) states that

A "resilient family" can balance the demands of the child with a chronic condition with other family needs, maintain clear family boundaries, develop communication competence, attribute positive meanings to the situation, maintain family flexibility, maintain a commitment to the family, engage in active coping efforts, maintain social integration, and develop collaborative relationships with professionals. (p. 6)

# Resiliency Model of Family Stress, Adjustment, and Adaptation

The Resiliency Model of Family Stress, Adjustment, and Adaptation (figure 1), which is derived from a substantial body of research (McCubbin et al., 2001; McCubbin & McCubbin, 1993) on family functioning over time, emerges from studies of warinduced family crises, the study of families faced with chronic stressors and illness (Kosciulek, McCubbin, & McCubbin, 1993) and the study of native Hawaiian, Filipino, Asian, American, and African-American families faced with both normative and nonnormative stressors and crises (McCubbin et al., 2001). Therefore, the Resiliency Model may be helpful to understand the ability to function among families who have a child with DMD.



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The resiliency model is characterized as having two discernible phases: adjustment and adaptation (McCubbin, McCubbin, Thompson, & Thompson, 1998). The traits of family adjustment and adaptation are healthy, normal, invulnerable, and resilient in well-functioning families. The Resiliency Model (McCubbin & McCubbin, 1993) characterizes the family system as "a resources exchange network in which problem solving and coping are the actions for this exchange" (p. 55). Successful family adaptation is achieved when "family schema and patterns of functioning are congruent, family members' personality and growth are supported, the family 's relationship with the community is mutually supportive, and the family develops a shared sense of coherence" (p. 59). Family adaptation involves the process of restructuring and making changes in rules, boundaries, and patterns of functioning. Families who experience an excessive demand from stressors deplete their resources; but when they adapt, they can restore functional stability and promote family satisfaction (McCubbin et al., 2001).

Families of children with DMD who are resilient may able to adjust to changing circumstances and have a positive attitude toward the challenges of family life. Bonadjustment (successful adjustments) occur when the needs of individual family members are met, and functioning of the family system and its transaction with the community is not threatened. However, having a child with DMD places enormous burdens upon how families function. The disability experience affects the functioning of the whole family, creating intense stress and draining its total resources. The chronic stress may result in disruption and break down of the family's functioning when the demands on the physical and psychological energy and other resources are too great. This outcome is termed maladjustment.
Family capabilities may be inadequate to deal with the chronic hardships of DMD leading to maladjustment and crisis (McCubbin et al., 2001). A family in crisis may try to develop new patterns of functioning, marking the beginning of the second phase of the Resiliency Model, the adaptation phase (McCubbin et al., 2001). During the adaptation phase, associated stressors as severe as the disability of the DMD child may produce a pile-up of demands with the family becoming increasing vulnerable. At this point, the family engages in dynamic relational processes to introduce changes in existing patterns of functioning to help resolve stressors. The family's level of appraisal influences the family system, and affects patterns of functioning, problem solving, and coping (McCubbin et al., 2001). A family that adapts to stress in these ways leads to a higher level of functioning.

Family adaptation is the optimal outcome if a new level of balance, harmony, coherence, and a satisfactory level of functioning is achieved following the progression of a disability (McCubbin et al., 2001). The adaptation phase of the Resiliency Model differs from the adjustment phase in that the family must develop new patterns of functioning in order to successfully adapt to their situation; if not, the Resiliency Model suggests that there will be a deterioration of the family's integrity, autonomy, or ability to manage their current crisis (McCubbin et al., 2001). There are several components of this model that can determine whether families can adapt. Each of these components is discussed below.

#### Family Stressors

Stressors may threaten the stability of the family unit or place significant demands on the family's resources and capabilities (McCubbin et al., 2001). McCubbin et al.

(2001) used the Family Inventory of Life Events and Changes to measure family stressors and strains.

# Family Resources

Family resources are a family's capabilities and strengths to resist a crisis and achieve harmony and balance. Hardiness is another term that represents these capabilities and strengths. Hardiness is a manifestation of competence despite exposure to significant stressors, and is another measure of healthy family functioning. Hardiness is a term that was first identified as personal resilience (eg. health status), characterized by commitment, challenge, and control (Kobasa, 1979). Hardiness results from "resilience processes that contribute to family dynamics and the family's abilities to cope effectively and adapt in crises" (Cohen, Slonim, Finzi, & Leichtentritt, 2002, p. 183). The components of family resilience include interpersonal relationships, open emotional sharing, system flexibility to shift roles and provide support, connectedness, and family values. McCubbin et al. (2001) refer "family hardiness" to

The internal strengths and durability of the family unit and is characterized by a sense of control over the outcomes of life events and hardships, a view of change as beneficial and growth producing, and an active rather than passive orientation in adjusting to and managing stressful situations. (p. 274)

Hardy families "shared a commitment to each other", "coped with change", "cultivated a protective environment in which family members actively" promoted "esteem among each other and themselves", "developed healthy lifestyles", and "encouraged coping skills of individual members" (Thames & Thomason, 2000, p. 1)

The Family Hardiness Index, the Family Inventory of Resources for Management, and the Family Time and Routines Index have often been used to measure the characteristics of hardiness as a stress resistor and adaptation resource that reflects the internal strengths and durability of the family unit (McCubbin et al., 2001).

# Family Problem Solving and Coping

Family problem solving and coping indicate actions that reflect a family's ability to deal with stressors and hardship to maintain or restore family harmony and balance. Researchers often use the Coping Health Inventory for Parents (CHIP), the Family Crisis Oriented Personal Evaluation Scales (F-COPES), and the Family Problem Solving Communication scale (FPSC) to measure the extent of family coping ability (McCubbin, McCubbin, & Thompson, 1996).

### Family Appraisal

Family appraisal is the family's perception of the seriousness of a stressor and its effects. It addresses family beliefs and expectations regarding the stressor and is defined as a familial sense of coherence. Appraisal has been measured using the Family Sense of Coherence (Antonovsky & Sourani, 1988).

# Family Schema and Meaning

Family schema is "a structure of fundamental convictions, values, beliefs, and expectations" (McCubbin, Thompson, Thompson, Elver, & McCubbin, 1998, p. 42). A family schema includes cultural-ethical beliefs and values. Family schema is how families attach meanings to their situation. The meaning is often determined by spiritual values and beliefs. Family schema assists the development of meaning through the processes of affirmation and spiritualization. Family schema have been measured using the Family (Ethnicity) Schema Index (McCubbin et al., 2001; McCubbin, Thompson et al., 1998).

# Family Adaptation

Family adaptation is the outcome of the family's efforts to create new ways of functioning in response to family stressors and is characterized as the minimal discrepancy between demands and capabilities (McCubbin et al., 2001). Family adaptation results "in a new or satisfactory level of balance, harmony, and functioning to a crisis situation" (McCubbin et al., 2001, p. 74). Family adaptation is often measured using the Family Assessment Device.

#### Research on Family Resilience

A review of studies on family resilience identifies four factors that influence family adaptation and three groups of factors that affect family functioning in families with a disabled child. Factors influencing adaptation include (a) stress related to emotional climate and pessimism concerning the child's future, (b) sense of coherence and use of resources, (c) social support, and (d) family strengths. Family hardiness, family support, and family communication; family problem solving communication, family schema, and family meaning; and family time together, life style, and accumulation of stressors and strains influence family functioning.

# Factors Influencing Family Adaptation

Stress related to emotional climate and pessimism concerning the child's future. Dyson (1997) found that there was no difference between fathers and mothers of children with disabilities in levels of parental stress, social support, or family functioning. Parental stress was related to family problems resulting from the child's special needs, the

family's emotional climate, and parents' pessimism concerning the child's future. The high rate of parent-reported child behavior problems among children with disabilities could reflect parental distress, especially about impairment in the social skills of their children (Smith & Oliver, 2001). Nereo et al. (2003) found that disabled children's emotional behavior problems were associated with mothers' stress. Psychosocial stressors in the lives of the mothers of children with handicapping conditions may result in initial shock, crisis, emotional changes, and pressures on family and social roles, requiring adjustment of parental role expectations (Burden, 1991).

Sabbeth (1984) found that fathers were at special risk for developing feelings of helplessness in relation to a child with a disability because of a number of conditions. Seligman and Daring (1989) also indicated that fathers and mothers also differ in their initial response to the diagnosis of a child who is disabled. In addition, Damrosch and Perry (1989) noted that fathers and mothers differed with regard to adjustment patterns and coping behaviors, and Ptacek, Smith, and Zanas (1992) found that men and women cope differently with stress.

Sense of coherence and use of resources. Bristor (1991) noted that quality of life for physically disabled children may depend to a large degree on the parents' ability to care for the child completely. In addition, socio-economic or material resources, locus of control, self-esteem, relationships with the family and social network, and service response can improve adaptation (Knussen & Sloper, 1992).

Gottlieb's (1998) research emphasized stress and coping resources in singlemothers of school-age children with a variety of developmental disabilities. In a study of 152 single mothers with developmentally disabled children, he found that their sense of

coherence (life as comprehensible, manageable, and meaningful) was associated with family adaptation. Single mothers who had a strong sense of coherence and greater use of resources had more adaptive outcomes. The sense of coherence was related to the mothers' perceptions of their child with disabilities.

Lustig (1997) studied 116 parents of adult children with mental retardation and found that most were resilient and exhibited positive functioning. There were positive correlations between scores on family adaptation and social support, family sense of coherence, and family adaptability. Lustig's work led to an empirical family typology and knowledge of a family's sense of coherence.

Margalit and Yona (1991) compared the ability of family systems to cope (including perception of family climate and sense of coherence) in an Israeli kibbutz (49 families with nondisabled children and 43 families with disabled children) and in an Israeli city (48 families of disabled children, 51 families of non-disabled children). The findings indicated that parents' sense of coherence assisted them to develop effective parental skills for seeking solutions to their child's specific needs. The implication was that improving parents' perception of coherence would promote family strength.

*Social support.* Judge (1998) examined the relationship between parental perceptions of coping strategies and family strength in 69 parents of young Caucasian children with disabilities in one geographic region. The results showed that use of social support was highly associated with family strength. This study provides evidence that families' informal and formal sources of support can strengthen family adaptation.

Bennett and Deluca (1996) used in-depth interviews of 12 parents with a disabled child to investigate the use of networks. Results showed that parents got (a) emotional

support and caregiving from families and friends; (b) emotional outlets and sources of information from parent groups; and (c) ideas for action and support from professionals. The implication is that a social network has an effect on family adaptation.

McCubbin, McCubbin, and Thompson (1993) conducted a study of the impact of pressures, strengths, and capacities on family life with 200 families in Hawaii including Caucasian (N=78), Asian (N=49), Hawaiian (N=37), and mixed-race families (N=36). The results showed that social support appeared to have greater explanatory power than other indexes for family adaptation. The major strength of the study is that it used a random digit dialing process and the sample size was large enough to compare the impact of disability on families in four different races. A second strength is that the study identified the reliability and validity of each psychometric measure of family adaptation. The third strength is that the findings of the study supported two critical explanatory factors, family schema and appraisal in the Resiliency Model.

*Family strengths.* McIntyre (2000) examined the role of competency-enhancing help in the adaptation process for 77 mothers of children with special needs. They found that higher levels of competency-enhancing help were related to greater maternal adaptation as measured by maternal sense of well-being and satisfaction with family functioning. In addition, competency-enhancing help was positively related to family resources and the use of positive coping strategies.

Silberberg (2001) used multiple methods to study 605 families and found that self-identified strong families agreed with positive statements (e.g., strongly connected to each other, easily to share values and ideas, love one another, often laugh with each other, enjoy helping each other) (p. 53). She found eight qualities of family strength among 177

volunteers including communication, togetherness, commitment, sharing activities, affection, support, acceptance, and resilience. She also extracted two strong themes from 33 families: support from extended family and friends and positive co-parenting arrangements. The major implication for nursing is a strengths-based approach that focuses on available resources and skills within the family and community, and the empowerment of the family and community in building resilience.

# Factors Influencing Family Functioning

*Family hardiness, family support, and family communication.* Olsen, et al. (1999) studied 54 couples (108 parents) of young children with disabilities and found that income, family support, and incendiary communication (defined as communication that is inflammatory in nature and tends to exacerbate stressful situations) predicted parent's hardiness. McCubbin, McCubbin and Thompson (1996) found that family hardiness was positively related to family support for the mothers and fathers, but negatively related to incendiary communication. Based on their findings, the researchers suggest that families develop basic capabilities and strengths, which foster the development and growth of family members and protect them from major disruption during family changes or transitions.

Family problem solving communication, family schema, and family meaning. McCubbin, Thompson et al. (1998) gathered self-report data from 101 parents of Native Hawaiian preschool children. Results showed that poor family problem solving, communication, and lack of family hardiness to be significant predictors of family dysfunction. Family schema was indirectly related to family dysfunction, primarily through coherence, hardiness, and family problem-solving communication. The authors

constructed a series of path models to account for indirect relationships of family schema and sense of coherence on levels of family functioning. A major strength of this study was the use of the Resiliency Model as the basis of the study and use of a randomly selected sample, thus making it a more reliable representation of the intended population of Native Hawaiians. But the small sample size limits generalizability of the findings.

Cannors' and Donnella's (1998) anthropological study explored parents' perceptions and coping abilities in eight Navajo families with autistic children and 24 families without autism. Results showed that parents were concerned about their children's social competency and residential placement. The implications are that professionals should encourage the family to become involved in early childhood special education, advocate for a family-centered approach, look at their own expectations, provide a loving and caring relationship, to protect the child.

Garwick, Kohrman, Titus, Wolman, and Blum (1999) designed a grounded theory study of 63 family caregivers of school children with chronic physical health impairments and used the Impact-on-Family Scale to discover how Hispanic, African-American, and European American families explain the cause of childhood chronic conditions and the indicators of resilience reflected in these explanations. The categories of explanation for the cause of childhood chronic conditions were: biomedical and environmental explanations, traditional and fatalistic beliefs, cause unknown, and personal attributions.

The major strength of the study is that the impact of traditional ethnocultural beliefs on families' explanations is most evident in descriptions of folk beliefs about illness and religious/spiritual interpretations of the chronic conditions. The influence of culture is also apparent in expressions of fatalistic and superstitious beliefs that reflect the

family's worldview, thus contributing the development of a substantive theory. A second strength is related to the development and refinement of psychometric measures focused on the impact of attitudes and ethnic differences. The third major strength is the contribution to our understanding of family explanations of their knowledge and attitudes toward minority patients and on the families' perceptions of cross-cultural health care behaviors. An important contribution of this research is a greater awareness and sensitivity of cultural differences in the meaning the family attributes to the cause of the child's condition.

Cohen et al. (2002) used a qualitative grounded theory method to study fifteen Israeli women whose families underwent crisis events. The authors found that family abilities, flexibility to shift roles, and the willingness of family members to give up their personal needs for someone else and to accept other people's feelings promoted family resilience. Other contributing factors included a sense of humor, trust, and providing a sense of security. The implications from this study focused on improving communication. However, the sample size was small and did not include males so the ability to generalize findings is limited.

*Family time together, life style, pile-up of stressors and strains*. McCubbin (1998) used regression analysis with data gathered from 184 African American enlisted military personnel and their spouses to determine factors most influential in helping them adjust to overseas assignments. Military life style (coherence) and confidence in spouse's self-reliance, spouse employment, and spouse's assessment of family time together emerged as important factors associated with family functioning. Critical variables of the accumulation of stressors and strains, and particularly family strengths, support, and

coherence were of central importance in explaining African-American enlisted family functioning in the face of reassignment.

Finally, Hawley (2000) based his study on the family resilience and narrative therapy model. Barriers to effective family communication and relationships were finances, differences over religion, father's depression, and interference from mother's ex-husband. A focus on family strengths and successes, developmental path, overcoming obstacles to achieve well - being, and obtaining outside resources led to improved family functioning.

These studies represent a wide variety of disciplines including epidemiology, sociology, psychology, and psychiatry. Most studies identified a broad range of background conditions, personal characteristics, social relations and community resources that may be helpful to understanding family functioning among DMD families. Many of these studies support various aspects of the Resiliency Model. In addition, the studies often explored the concept of resilience from multiple family dimensional processes including belief systems, organizational patterns, and communication processes. Family functioning research has contributed to a recognition of the need for interventions, such as personal or social support networks, self-help groups based on conventional wisdom, strength-based approaches to family support to facilitate family functioning. However, the Resiliency Model cannot be used to measure the family adaptation of Taiwanese families because there are no reliability and validity measures in Mandarin to assess the several aspects of the model (e.g. family appraisal, schema, and meaning). Furthermore, the model does not consider the inherited, progressive, life-threatening

nature of a condition like DMD as a family stressor. Therefore, a modified version of the model was used to examine the family functioning of DMD families in this study.

Conceptual Model of Family Stressors, Resources, and Functioning

The Conceptual Model of Family Stressors, Resources, and Functioning is proposed in Figure 2 and includes the measured concepts (variables), and empirical indicators (instruments). Figure 2, represents the relationship among the variables of family stressors, resources, and functioning. Independent variables were the child's disability and access to care. The dependent variable was family functioning. The mediating variables were family health, family characteristics, family support, and family hardiness. Family health, family characteristics, family support, and family hardiness of a child's disability and antecedents of family function.

A brief discussion of each variable and its relationship to the other variables will be presented next.



Figure 2 Conceptual Models for Family Stressors, Resources, and Functioning

# Family Stressors

In the Model, family stressors (the child's disability and access to care) place demands on family resources. As the DMD child's condition worsens the families may become more vulnerable. Families in crisis try to resolve stressors by using family resources, reviewing the meaning of life, and engaging in dynamic relational processes to guide changes in existing patterns of family functioning to resolve their stressors (McCubbin & McCubbin, 1993). The following literature review will focus on terminology, measurement and their relationship to family functioning.

*Child's disability*. Disability means an incapacity or disqualification. A child with a disability is deprived of physical or mental abilities. It is a long-term impairment adversely affecting specific normal daily activities (Kenneth, 2001). A disability makes the DMD children depend on families to give them assistance and that stressors contributes to an accumulation of demands on the families. Holroyd and Guthrie (1979) reported that physical incapacitation of chronically ill children with neuromuscular or psychiatric disease was a predictor of burden. Snowdon, Cameron, and Dunham (1994) found that severity of child's condition and behavior problems are the significant stressors for the families with developmental disabilities. The severity of disability was defined on the child's independence, measured by the Barthel Index, to evaluate daily activity conditions.

The Barthel Index (BI) is probably the most widely used generic disability measure. It was developed in 1955 as a simple index of independence useful in scoring disability. Independence means that the person needs no assistance at any part of the task (Mahoney & Barthel, 1965). The BI was as good as any other single simple index for

clinical purposes but might be limited in the context of research (Wade & Collin, 1988). Van der Putten, Hobart, Freeman, and Thompson (1999) suggested that the Functional Independence Measure had no advantages over the BI in evaluating changes in disability due to therapeutic interventions. This has important clinical implications, as the BI is quicker and simpler to rate.

Access to care. Once children are diagnosed with DMD, their parents are in continual contact with health professionals regardless of the children's age. Generally, the parents will gather or research formal knowledge about the disease from professional and begin to access supportive care system when children are diagnosed. Care assists the process of emotional adjustment to the child's disability, enabling parents to access service and benefits, and improve parents' management of the child's behavior (Pain, 1999). Only after diagnosis do parents learn what information is helpful (Pain, 1999). Some parents felt that information was hard to get because they didn't know the right question to ask (Beresford, 1994). Some researchers have concluded that professionals can provide support to parents by providing resources and expertise and helping create and maintain an open, honest, and collaborative relationship with parents (Bennett & DeLuca, 1996; McCallion & Toseland, 1993). This study identified the age when a child was diagnosed with DMD, noting that earlier detection implied families had early access to professional care.

# Family Resources

Family resources are the capacities of families to respond to the crisis of their child's illness so they can regenerate personal energy (McCubbin et al., 2001). Snowdon et al. (1994) suggest that hardiness, health, esteem, and communication are coping

resources. The components of family resources in the proposed Model include family characteristics (employment and family annual income), family health, family support, and family hardiness. These are viewed as mediating variables that provide family energy to address each member's physical, mental, social, and spiritual needs in order to solve problems and maintain healthy family functioning.

*Family health.* To bring an ill, handicapped, or disabled child into the world is one of the most heartbreaking events parents ever face. Chronic emotional stress was reported by parents to be the most significant problem in coping with a child who has DMD due to the unrelenting, constant demands of medical, physical and emotional care required by the disease. There is no doubt that the physical, psychological, social, and emotional health and well-being of family members are essential protective and recovery factors in promoting resilience in families (McCubbin, McCubbin, Thompson, Han, & Allen, 1997) and are often used as the outcome measure of resiliency (McCubbin et al., 1997). Therefore, the degree of family health may explain the variability in resiliency in the family system. Parkerson, Broadhead, and Tse (1991) developed the Duke Health Profile (Duke) to measure self-reported health, quality of life, and functional health status. It has been used primarily for research on health-related outcomes in the clinical setting. In this study family health parental health, measured by the Duke Health Profile, to evaluate physical, social, mental, perceived health, anxiety, depression, disability, selfesteem, and pain.

*Family characteristics*. Family characteristics are a family's capabilities and strengths to resist a crisis and promote family resilience to maintain patterns of functioning to achieve harmony and balance. Based on Smilkstein's (1978) acronym

SCREEM-the major family characteristics are social, culture, religious, economic, educational and medical. When families have social support, cultural satisfaction, economic stability, high education, and access to medical care, they are better able to function.

As DMD progresses, demands upon the parents also increase. The financial burden of a chronically ill child falls most heavily on middle-class Taiwanese families because parents are often searching for alternative therapy that is not covered by insurance. Palfrey et al. (1989) found that educational level and socioeconomic factors had significant effects on parental stress. Reid and Renwick (2001) found that familial stress is not significantly related to any of the socio-demographic measures. But Canning, Harris, and Kelleher (1996) reported that family stress was related to family income. Svavarsdottir (1997) found that the number of children and family income were positively correlated with family hardiness, indicating mothers of children with asthma who had more children and higher income reported higher hardiness (r = .27). There was no relationship between family hardiness and parents' age, and length of marriage. So, parents' employment and annual income were the most important variables of family characteristics in the model.

*Family support*. Family support is family members' satisfaction with their family's responsiveness and caring for their needs. These include adaptation, partnership, growth, affection, and resolve. Family support services can encourage the use of cognitive coping strategies to facilitate healthy functioning in families with disabled children (Summers, Behr, & Turnbull, 1989). Yu (2002) used the Family APGAR (FAPGAR) to evaluate family support and found that there was a positive correlation

with self-efficacy for the families of tuberculosis patients. Some researchers translated the FAPGAR into Mandarin as"家庭關懷度指數 chia ting guan huai du zhi shu" that retranslated into English means "family caring index" (Smilkstein, 1978). A low score was shown to be predictive of psychosocial problems in children, patients, and families (Chen, 1988; Chen, Chen, Hsu, & Lin, 1980; Tsai, Chang, & Tseng, 1993; Tyan, Chie, & Chang, 1988). This study used the meaning of "family caring index" to measure perceived family support in the five domains of adaptation, partnership, growth, affection, and resolve (Gardner et al., 2001).

*Family hardiness*. Family hardiness is the internal strength of a family system and durability of family unit characterized by a sense of control to over life events and hardships by the family working together to solve problems. Leske (2003) defined "hardiness as the family's internal strengths and durability" that help a family adapt over time by "an ability to work together to find solutions to difficulties" (p. 33). Lambert and Lambert (1999) defined "hardiness as a constellation of attitudes, beliefs, and behavioral tendencies that consist of three components: commitment, control, and challenge" (p. 11).

For this study, family hardiness was conceptualized as the energy resource used to help facilitate adjustment and adaptation over time by serving to release the negative effects of stressors and demands. In addition, being able to view change as beneficial and growth-producing and an active rather than passive orientation in adjusting to and managing stressful situations is also important to family hardiness. The attributes of family hardiness include commitment, challenge, and control (McCubbin et al., 2001). Family hardiness is a mediating factor to decrease the effects of stressors and demands on the family and maintain normal family functioning.

McCubbin, et al. (2001) developed "the Family Hardiness Index (FHI) to measure the characteristics of hardiness as a stress resistance resource and adaptation resource in families, which would function as a buffer or mediating factor of stressors and demands and as a facilitation of family adjustment and adaptation" (p. 274). Henkle (1994) found that family hardiness is an important resistant resource for the burden and stress of family caregiving. Kamya (1997) found that family hardiness would explain the variance in caregiver well-being and suggested that future research should be on caregivers of the functionally impaired. Donnelly (1994) reported that parents of children with asthma viewed their families as hardy, and found that there was a significant relationship between family hardiness and family stress.

Olsen, et al. (1999) used hardiness to describe people who remained healthy even while experiencing high amounts of life stress. They defined hardiness as the sum of 3 components: control, commitment, and challenge, just as Lambert and Lambert (1999) did. A few studies have explored the construct of family hardiness (Failla & Jones, 1991; McCubbin, Thompson, & McCubbin, 1996) and influencing factors that included family stress, family support, emotional distress, family coping strategies, family appraisal, and demographic factors (Campbell & Demi, 2000; Mellon & Northouse, 2001; Olsen et al., 1999).

Mellon and Northouse (2001) explored the quality of life of families of long-term survivors of a cancer; they found that there was significant positive relationship between family hardiness and family quality of life (r = .37); significant negative relationships between family hardiness and family stressors (r = -.26); and family hardiness and

patients' fear of recurrence (r = -.24). Family hardiness made a unique contribution to the variance in family meaning of the cancer illness.

# Family Functioning

Family functioning is the outcome of the families' ability to use family resources and other sources of support. In this study, problem solving, communication, role, affective responsiveness, affective involvement, behavior control, and healthy family functioning were conceptualized as attributes of the resilient DMD family (Epstein, Bishop, Ryan, Miller, & Keitner, 2003). In addition, family health, family support and family hardiness were viewed as indicators of adaptation of the DMD family. Successful family functioning has been found to reduce demands on the family system and brings resources to manage the situation (McCubbin et al., 2001).

Several different attributes of family functioning have been described. Epstein, Bishop, and Levin (1978) defined them as problem solving, communication, role, affective responsiveness, affective involvement, and behavior control. Olson, Sprenkle, and Russell (1979) identified the attributes of family functioning as family cohesion, adaptability, and communication; Beavers and Hampson (1993) described them as competence and style; Suttiamnuaykul (2001) noted that basic attributes appropriate to culture, society, economic, and political policy were important for families to function well.

Family functioning has been studied among families with children with various serious conditions, resulting in conflicting findings. For example, some studies report a negative relationship between family functioning and the children's conditions: major depressive disorders, depression (Fornari, Wlodarczyk-Bisaga, Matthews, Sandberg, &

Katz, 1999; Stein et al., 2000; Tamplin & Goodyer, 2001), suicide adolescent depression (King, Segal, Naylor, & Evans, 1993), psychiatric disorders (Friedmann et al., 1997), eating disorders of bulima nervosa (Fornari et al., 1999), anorexia nervosa (Gowers & North, 1999), mental retardation, down syndrome, physical disability (Luescher, Dede, Gitten, Fennell, & Maria, 1999), epilepsy (Pal, Chaudhury, Das, & Sengupta, 2002), traumatic brain injury (Rivara et al., 1996), or oppositional defiant disorder (Tamplin, Goodyer, & Herbert, 1998). Other studies found that there were no relationships between the children's condition and family functioning: fractures (Loder, Warschausky, Schwartz, Hensinger, & Greenfield, 1995), developmental disability (Dyson, 1997), or anorexia nervosa (Dare & Key, 1999) - in family functioning. There were no significant differences between parents from families with healthy and unhealthy children (Keitner et al., 1995). Furthermore, the severity of involvement of cerebral palsy children did not seem to influence parents' perception of family functioning (Magill-Evans, Darrah, Pain, Adkins, & Kratochvil, 2001).

Early studies of family functioning focused on the family's economic functioning. The pioneer LePlay (Silver, 1982), who conducted the first study of family functioning in the 1850s, submitted that family functioning was related to health and well-being of the family. His research was based on analysis of family budgets. Other pioneer studies of family functioning supported LePlay's important idea of economic functioning (Schwab, Gray-Ice, & Prentice, 2000). After 1859, some family research focused on hereditary influences on mental health and illness that were related to procreative and social functioning of the family. These studies pointed to the importance of the family's basic reproductive function.

At the end of the 20<sup>th</sup> century, abortion, family values, and day care became the key issues and replaced reproductive functioning. Researchers were interested in hereditary patterns of mental illness and mental retardation in the family. Families of patients with depression were likely to experience more dysfunction than families without psychiatric disorders (Friedmann et al., 1997; Keitner et al., 1991; Keitner et al., 1993). Fifty to seventy percent of families of depressed patients perceived their own family functioning as unhealthy (Keitner et al., 1995).

Lately, repeated studies have found the negative effects of depression or chronic mental illness on family well-being. Keitner (1990) found significant associations between depressed patients and the quality of family functioning, especially impaired role functioning. Friedmann et al., (1997) found that "having a family member in an acute phase of psychiatric illness was a risk factor for poor family functioning" (p.357). And 80% of the families with anxiety disorders and 74.8% of the families with major depression had unhealthy functioning in communication, with 50-80% of the various patients' families with impaired general function.

The correlation between behavioral–emotional symptoms and family dysfunction has been found in other studies (Heru & Ryan, 2002; Keitner, Ryan, Miller, & Norman, 1992; Lindeman et al., 2002). Scahill et al. (1999) found that children with attentiondeficit hyperactivity disorder (ADHD) were more likely to live in low-income families with higher levels of family dysfunction. Poor family functioning at 5 years after a child was sexually abused was associated with low self-esteem and behavior problems (Tebbutt, Swanston, Oates, & O'Toole, 1997).

Acreman (2002) found that gender of child, family income, single parent status, parental level of education, family functioning, parental depression and school readiness as predictors of academic resilience. Baigas (2002) indicated that lower rates of visualmotor, academic, adaptive, and social development after four months were found in children whose families scored more dysfunctional on the FAD. And there were significant differences between learning disability (LD) and non-LD families on structure and interaction on five of the seven FAD scales: roles, behavior control, communication, affective-responsiveness, and general functioning. However, there was a positive relationship between healthy family functioning and socioeconomic level. Vandsburger (2001) suggested that the effects of family hardiness and social support on family functioning in families experiencing economic pressure did not fit these data.

Kim (2002) found that intra-family and extra-family resources were significant predictors of family functioning. Whether the child had a disability and the age of the child (adolescent versus young adult) were not significant predictors. Researchers have found that dyadic relationships within the family, especially parent-child relationship, are related to the functioning of the family (Hayden et al., 1998).

#### Conclusion

The disability of the DMD child induces the family to change and experience many challenges over their life-time, putting their family functioning at-risk. A complete understanding of how well DMD families' function, however, is unknown. With little information about Taiwanese family functioning in general, and conflicting data about the functioning of families with seriously ill children from in larger populations, further research is needed.

The DMD population in Taiwan has been studied to improve understanding of family stress, parents' coping, social support, and quality of life (Chen et al., 2002; Chen et al., 2003; Huang & Dai, 1998; Kao, 1998). However, knowledge about family support, family hardiness, and family functioning in the DMD family is needed before health care professionals can provide family-centered interventions that promote family health, adaptation, and better family functioning.

# CHAPTER III: METHODOLOGY

This descriptive correlational study used a cross-sectional, predictive design to explore the family functioning of 126 parents aged 28 to 61 years in Taiwan who have a child with DMD. The design looked at an event at one specific point in time (Rubin & Babbie, 1997). The study used the Conceptual Model of Stressors, Resources, and Functioning (Figure 2), a revised version of the Resiliency Model, because several instruments used for the latter have not been tested or translated into Mandarin. For example, only one subscale of the Family Hardiness Index, has been translated.

Table 1 showed the concepts, variables, and instruments in the present study. Child's disability and access to care were measured by self-report. The degree of the child's disability and reported age when diagnosed with DMD were the family stressors. Family resources included family characteristics, family health status, family support, and family hardiness. These were measured with a demographic sheet including parents' employment and family annual income, the scales of the individual Duke Health Profile (Duke), the Family APGAR (FAPGAR), and the Family Hardiness Index (FHI). Family functioning was measured with a scale of the individual Family Assessment Device (FAD). All instruments were translated into the Chinese language and used in Taiwan. This study utilized the data collected from the parents. Participants individually completed each measurement. They were excluded if the DMD children, siblings, or grandparents helped parents answer the questionnaires.

# Table 1

Concepts	Variables	Instruments		
Family Stressors	Child Disability Daily activity dependent Access to Care Age when diagnosed with DMD	Barthel Index (BI) Demographic Sheet		
Family Resources	Family Health Family Hardiness Family Support Family Characteristics Family annual income Parents' employment	Duke health profile (DUKE) Family Hardiness Index (FHI) Family APGAR (FAPGAR) Demographic Sheet		
Family Functioning	Family Functioning	Family Assessment Device (FAD)		

Concepts, Variables, and Instruments in the Current Study

### Identification of Study Population

The study used a convenience sample to recruit parents of children with DMD into the study. Although convenience samples have advantages for multiple reasons including recruiting time, accessibility, and low expense, there was a chance that respondents might not have returned the questionnaires if they hadn't received a followup phone call, a stamped return envelope, and assurance of confidential communication. In addition, the sample may not have been representative of the population and those who were more uncomfortable might have refused to participate in the study. The question of generalizability was addressed (Polit & Hunger, 1999), in part, by having a representative sample of parents from each of the families with DMD children in the Taiwan Muscular Dystrophy Association (TMDA). Therefore, results can be generalized only to the DMD group of the TMDA (86.4% of the participants were members of this organization). The target population for this study were parents of children with DMD who participated in TMDA's groups or used medical resources from Kaohsiung Medical University Hospital (KMU hospital) for diagnostic evaluation, support, and medical care. Pediatric neurologists diagnosed the children's DMD through muscle biopsy and serological tests (creatine phosphokinase-CPK and lactate dehydrogenase-LDH) and provided follow-up care. The TMDA, created in 1995, developed support groups for DMD families and expanded to other families with family members with different types of muscle dystrophy. There were three branches of the organization, located in the south, north, and central areas of Taiwan.

#### Subject Sample

A convenience sample of 126 parents participated in this study. They came from a total pool of 125 DMD families (245 parents) in the TMDA, as well as outpatients from Kaohsiung Medical University (KMU) hospital. The response rate to questionnaires was 62% (based on mailings to 203 parents who had agreed to receive the questionnaires). Forty-six couples (58%) completed the questionnaires; eight fathers (10% of families), and 26 mothers (32% of families) also completed the questionnaires. The subjects who declined to participate had multiple reasons, including death, divorce, separation, illness; others had no forwarding address or gave no reason.

# Demographic Description of the Subjects

The demographic characteristics of the 126 parents are found in Table 2. The majority of parents were female (57%). On average, mothers and fathers were in their early 40s with the parents' mean age of 43 (SD= 6.1) and a range of 28 to 61. The majority of parents in the study were Taiwanese (76%), high school graduates (35%),

# Table 2

# Demographic Characteristics of Parents for Current Study

Characteristics	5	No	%	
Gender	Male	54	42.9	
	Female	72	57.1	
Parent age	<=35 years	13	10.3	Mean age $= 43$ years
U	36-40 years	28	22.2	SD = 6.1 years
	41-45 years	48	38.1	Range = $28-61$ years
	>=46 years	27	21.4	
Ethnicity	Taiwanese	96	76.2	
-	Chinese	13	10.3	
	Haika	15	11.9	
	Aboriginal	2	1.6	• •
Education	Elementary	15	11. <del>9</del>	
	Primary school	37	29.4	
	High school	44	34.9	
	College school	16	12.7	
	University or higher	14	11.1	
Occupation	Laborer of farmer	33	26.2	
•	Technique	14	11.1	
	Government officer	13	10.3	
	Professional	12	9.5	
	Business	15	11.9	
	None or homemaker	39	31.0	
Marital status	Married	114	90.5	
	Separated	1	.8	
	Widowed	5	4.0	
	Divorced	.3	2.4	
	Remarried	3	2.4	
Religion	Buddhism	63	50.0	
	Taoist	36	28.6	
	Christian	5	4.0	
	Catholic	1	.8	
	None	21	16.7	

doing work as laborers or farmers (26%), married (91%), and Buddhist (50 %).

Demographic Description of the Children with DMD and Their Families

The age of the children ranged from 3-25 years (mean = 14.3, SD = 4.6). Twenty-three percent of the children were eighteen years old or more, and 41% were teenagers. Forty-six percent of the children could not raise their hand to their mouth, and 78% needed wheelchair assistance. Seventy-three percent of the children still attended school or received education at home (Table 3).

Sixty percent of families were living in an urban area, 76% were nuclear families, and 66% had an adolescent child. The majority of families had only one child (44%) and 42% had two children (Table 3).

# Sample Size and Data Analysis

There were two types of statistical techniques, Pearson correlation and multiple regression, used to analyze the data. A power of .93 was reached with the sample of 126 subjects with an effect size of .3 and set alpha at .05 on the Pearson correlation. A power of .86 was reached with the sample of 126 subjects with an effect size of .15 and alpha set at .05 on the regression (Cohen, 1988).

# Procedure

# Access to Study Population

The investigator contacted the leaders of KMU Hospital and the TMDA, the pediatric and adult neurologists, and the social worker of the TMDA to present the study and obtain permission to contact eligible participants. Permission was obtained (see letters of support, Appendix A-G).

# Table 3

Characteristic		Frequency %				
Child age	Range = 3-25 years Mean =14.3 years SD = 4.6 years					
Child's upper	Can raise hand to mouth	43	52.8			
extremity function	Can not raise nand to mout	n 3/	40.2			
Child's lower	With wheelchair assistance	62	77.5			
extremity function	Without wheelchair assistant	nce 18	22.5			
Child education in	Attended	58	72.5			
School or at home	Not attended	22	27.5			
Location	Rural	32	40.0			
	Urban or Municipal	48	60.0			
Family status	Nuclear	61	76.3			
	Extended	19	%   52.8   46.2   77.5   22.5   72.5   27.5   40.0   60.0   76.3   23.8   1.3   22.5   66.2   10.0   8.8   43.8   42.4   5.0			
Developmental	Preschool	1	1.3			
stage of children in	School	18	22.5			
family	Adolescent	53	66.2			
-	Adult	8	10.0			
Sibling number	0	7	8.8			
	1	35	43.8			
	2	34	42.4			
	3	4	5.0			

Demographic Characteristics of the Children with DMD and Families (N=80)

# Data Collection

Eligible subjects (parents of children with DMD) were mailed a letter by the TMDA or were invited by the neurologists to participate in the study. Then the investigator made a phone call to ask the subjects to participate in the study; if the subjects agreed to fill out the questionnaires the investigator sent each family a cover letter with two sets of questionnaires and informed consents, which outlined the purpose and procedures of the study and assurance of confidentiality. The letter further informed them that the investigator would respect their right to refuse to participate if they were in distress. It was once again emphasized to the participants that all their responses would be held in strictest confidence and locked in a file cabinet, and that only a number would be used for subject identification. A telephone number was included in case there were further questions or consultations.

Parent participants were instructed to individually answer the questionnaires separately and avoid discussion of the questions with others. Each subject took approximately 40 minutes to complete the questionnaires. Each subject received a phone call from the investigator within the first week after mailing the questionnaires and again two weeks later to remind him or her to complete and return them. The researcher enclosed a payment envelope, consent forms, and another set of forms if the subjects lost them. The researcher also offered assistance by phone to help them complete the questions, and later contacted them if the questionnaires were not completed. The instrument data was entered by the investigator and rechecked to prevent artificial errors and loss of the sample. The researcher used SPSS (version 11) to survey the data and conduct the analysis.

#### Instruments

### The Demographic Sheet

The demographic sheet included information about the child's level of disability (using criteria from the Barthel Index-BI) and the age when the child was diagnosed with

DMD. Family resources included parent's age and education, ethnicity, employment status, marital status, family location, religion, family income, and family size.

# Barthel Index

The BI is a 10-item instrument measuring disability in terms of a person's level of functional independence in personal activities of daily living (Mahoney & Barthel, 1965). The BI contained items about feeding, moving from wheelchair to bed and return, grooming, transferring to and from a toilet, bathing, walking on a level surface, going up and down stairs, dressing, and continence of bowels and bladder. It was rated by observation. The BI has been used to measure disability in both adults and children.

Item scores (based on different levels of independence: independent = 0; need help or major help = 5; independent, minor help, or continent = 10; maximum independent = 15) were summed up to generate a total score. There were two items on a two-point scale, six items on a three-point scale, and two items on a four-point scale (Appendix H). Scores on each rating form were added for an overall score, with higher scores indicating greater independence. The scores ranged from 0 (totally dependent) to 100 (fully independent). This study used Shah, Vanclay, and Cooper's (1989) suggestion that scores of 0-20 indicated total dependency, 21-60 indicated severe dependency, 61-90 indicated moderate dependency, 91-99 indicated slight dependency, and 100 indicated complete independence.

The BI is an ordinal rating scale. Each item was rated in terms of whether the child could perform the task independently, with some assistance, or was dependent on help based on observation. The scores for each of the items were summed to create a

total score. The higher the score, the more independent the person was (Mahoney & Barthel, 1965).

# Validity

Wade and Hewer (1987) reported validity correlations ranged from .73 to .77 with an index of motor ability for 976 stroke patients. Wylie and White (1964) and Wylie (1967) found that the BI correlated well with clinical judgment and predicted mortality or ability to be discharged to a less restrictive environment.

### Reliability

The BI had evidence of reliability and validity (Collin, Wade, Davis, & Horne, 1988). Sherwood, Morris, Mor, et al. (1977) reported high alpha reliability ranging from .953 to .965 for three samples of hospital patients suggesting that the test was internally consistent as a measure of self-care activities. Shah et al. (1989) reported alpha internal consistency coefficients of .87 to .92 for the original scoring system and .90 to .93 for a revised scoring system. It was .88 in the current study.

# Family Characteristics

The demographic questionnaire included parental education, ethnicity, family religion, family annually income, parental employment, satisfaction with medical care, sibling health, family structure, family size, family development stage, parental age, and family location. The demographic variables of employment and annual income were used to measure family characteristics.

# The Duke Health Profile

The DUKE is a 17-item measure of adult health - related quality of life and functional health status. The 17 items are divided into 6 scales, measuring positive functional health and 5 scales measuring negative functional health. The six scales of functional health include: physical, mental, social, general, perceived health, and self-esteem; higher scores indicate better health-related quality of life (greater functional health). The five scales of negative functional health include: anxiety, depression, anxiety-depression, pain, and disability; higher scores indicated greater dysfunctional health (Parkerson, 2002).

The physical, mental, social, and perceived health scales and the disability scale are independent of each other in that none of their items are shared, whereas the other scales are not independent because they shared single or multiple items extracted from the independent scales. The DUKE consists of 10 summary scores: physical health (5 items), mental health (5 items), social health (5 items), perceived health (1 item), self-esteem (5 items), anxiety (6 items), depression (5 items), pain (1 item), perceived health (1 item), and disability (1 item). A general health score was obtained by combing averages of the first subscales. Some items contributed to several summary scores; for example, number 4, "I gave up too easily," contributed to the mental health, self-esteem, and depression scales. Responses were made on a three-point scale (see Appendix I for the rating scale). A total score was calculated based upon a summary of the 17 items score was the mean of the raw scores transformed from a scale of 0-2 to a scale of 0-100 (raw scores of 0, 1, and 2 become final scores of 0, 50, 100) (see Appendix I for the DUKE scores of procedures). The higher score indicated better health (Parkerson, 2002). *Validity* 

The DUKE obtained adequate validity by using the Family Strengths and Family Inventory of Life Events (Parkerson et al., 1991). The 7-item anxiety-depression

subscale of the DUKE (DUKE-AD) had been used as an effective screener for DSM-III-R major anxiety and depression. Validity had been strongly supported for the instrument. Tsai et al. (1993) also showed that the subscale scores of the DUKE were significantly correlated with demographic and clinical variables. The predicted relationships among the DUKE score and clinical variables supported the construct validity of the DUKE. The DUKE had been found to have significant correlations with the Psychological Symptom Scale, Tseng's Depression Scale, Chinese Health Questionnaire, and Family APGAR to support convergent and discriminate validity.

### Reliability

Reliability estimated for the DUKE is the following: most of multi-item scales had Cronbach's alpha reliability coefficients in the .60 and .70, while the single item scales had test-retest coefficients in the .40 and .50 (Parkerson, 2002). Tsai, Chang, and Tseng (1993) compared the Chinese version of the DUKE with 557 adult outpatients' and 323 adults seeking general health examinations; they found that one-week interval test-retest reliability for the DUKE was .51 to .85 and internal consistent Cronbach alpha was .49 to .70. The study found that internal consistent Cronbach alpha for the DUKE was .60, mental health was .52, and social health was .69.

In addition, the DUKE had been used mostly for primary care patients, but also for normal medical students and insurance policyholders, and for patients with chronic lung disease, insulin-dependent diabetes, end-stage renal disease requiring hemodialysis, and cardiac and musculoskeletal disorders (Medical Outcome Trust, 2001). The DUKE had been translated into a Chinese version (Medical Outcome Trust, 2001).

# Family APGAR

Smilkstein (1978) designed the FAPGAR to evaluate adult satisfaction with social support from the family. The components of FAPGAR include adaptation, partnership, growth, affection, and resolve. Adaptation is "utilization of intra and extra familial resources for problem solving when family equilibrium is stressed during a crisis" (p.1232). Partnership is "the sharing of decision making and nurturing responsibilities by family members" (p. 1232). Growth is "the physical and emotional maturation and self-fulfillment that is achieved by family members through mutual support and guidance" (p. 1232). Affection is "the caring or loving relationship that exists among family members" (p. 1232). Resolve is "the commitment to devote time to other members of the family for physical and emotional nurturing. It also usually involves a decision to share wealth and space" (p. 1232).

The FAPGAR is a 5-item measure of perceived family support (Smilkstein, 1978). Each item allowed three responses (2 = almost always, 1 = some of the time, 0 = hardly ever) (Appendix J). The total scores range from 0 to 10 (low to high satisfaction with family support). Lower scores indicate more parental distress (Gardner et al., 2001). All items were summed for a total score.

# Validity

*Construct validity.* A correlation of .64 was found between FAPGAR and a therapist's rating of family functioning of mental health outpatients. Good et al. (1979) noted a correlation of .80 with the Pless-Satterwhite Family Function Index (r = .80). Foulke, Reeb, Graham, and Zyzanski (1988) used 140 families to explore the relationship between the FAPGAR and the Family Adaptation and Cohesion Evaluation Scales

(FACES). They found that FAPGAR is highly correlated with the Cohesion Scale of FACES (r = .70) and moderately correlated with the Adaptability Scale (r = .59) to support construct validity.

*Criterion validity.* Moos and Moos (1981) reported a correlation of .54 (p = .01) with the FACES III cohesion sub-scale, and a correlation of -.40 (p. = .01) with Family Environment Scale to support the criterion validity. The Family Disruption from Illness Scale (FDIS) correlated significantly in the expected direction with all measures of family functioning: Family APGAR, r = -.23 (Gragert & Ide, 2003). Gwyther, Bentz, Drossman, and Berolzheimer (1993) found that the FAPGAR failed to detect family dysfunction found by psychological interview, but there was a strong relationship with the Minnesota Multiphasic Personality Inventory (MMPI) for 198 patients: 58 irritable bowel syndrome patients (IBS), 67 IBS nonpatients, and 73 normal subjects.

Gardner et al. (2001) suggested that low scores on the FAPGAR might measure parental distress, reflecting parental depression. Chen, Chen, Hsu, and Lin (1980) reported that well-adjusted Taiwanese students (N=1164) had higher scores in each subscale of the FAPGAR than the maladjusted students (N=1377). They also found that adopted children had significantly lower FAPGAR scores than biological children, and separated students had significantly lower FAPGAR scores than those living with parents. Chen (1988) reported that there was a relationship between the stimuli that children (N=100) perceived as stressful in the hospital and their scores on the adaptation and partnership subscale of FAPGAR. Lee et al. (1992) found that low FAPGAR scores could independently predict depressive symptoms among 397 patients with active
pulmonary TB and the FAPGAR was significantly related to those who received TB treatment.

*Discriminant validity.* FAPGAR scores of married graduate students were significantly higher than scores of community mental health clinic patients (Smilkstein, Ashworth, & Montano, 1982). Good, Smilksteine, Good, Shaffer and Arons (1979) found a significant difference between the FAPGAR scores of the psychiatric outpatients and healthy adults groups. In addition, Hilliard, Gjerde, and Parker (1986) found significant differences in the mean FAPGAR score (respondents rating five-Likert scale) between nonsymptomatic patients (mean = 38) and patients with suggestive symptoms (abdominal pain of uncertain etiology, urticaria, peptic ulcer, irritable bowel syndrome) or clear symptoms (anxiety, depression, suicide attempt, marital dysfunction) (mean = 32). The results supported discriminate validity. In terms of psychometric validity, they also found a false-negative rate for the FAPGAR (19%) (insensitivity to psychological problems).

### Reliability

The instrument has obtained satisfactory reliability scoring, ranging from .80 to .89 (Gillis, Neuhaus, & Hauck, 1990; Kirkevold, Gortner, Berg, & Saltvold, 1996; Smilkstein et al., 1982). The Cronbach  $\alpha$  was .80, a high internal consistency for a sample of 291 women and 238 men whose average age was 19.7 years (Smilkstein et al., 1982). Inter-item correlations ranged from .24 to .67, and the inter-spouse correlation for the FAPGAR was .67 (Good et al., 1979; Smilkstein et al., 1982). Moos and Moos (1981) reported an alpha coefficient of .84. Kirkevold, Gortner, Berg, and Saltvold (1996), and Good et al. (1979) noted a split-half reliability coefficient of .93. Two-week interval test-

retest reliability was .83 among 100 Taiwanese students (Chen et al., 1980). The Cronbach  $\alpha$  of this study was .89.

The Family APGAR has been used to screen for lack of family social support (Murphy et al., 1998). Several researchers used the FAPGAR to evaluate family relationship of HIV-I infected patients (Lee, 1999; Lee, Chuang, & Shen, 1994; Lee & Lin, 1989), and cardiac inpatients (Lee, 1991). It has also been used to look at family relationships in terms of health status, neurosis, severe mental symptoms, and coping strategies. Further, it has been used in the study by Chen, et al. (1980) in Taiwan when the instrument was translated into Chinese.

### Family Hardiness Index

The FHI was "developed to adapt the concept of individual hardiness to the family unit" and consists of three components: commitment, challenge, and control (McCubbin et al. 2001, p. 273). According to McCubbin et al. (2001), commitment represents "family sense of internal strengths, dependability, and ability to work together to manage the difficulties" ( p. 277). Challenge means "family efforts to be innovative, active, and to experience new things and to learn" (family believes that hardship is normal for life to change) (p. 277). Finally, control is defined as the "family sense of being in control of family life rather than being shaped by outside events and the victim of circumstances" (is the tendency to believe and act in a way that influence the course of life's events) (p. 277).

The Index was a 20-item instrument with a four-point scale that was constructed to measure three components: commitment-8 items, challenge-6 items, and control-6 items (McCubbin, McCubbin, & Thompson, 1986). Scoring of the FHI is done by the summation of the chosen response, which represents the degree to which the personal agree with the statement at the present time (0=False, 1=Mostly false, 2= Mostly true, 3=True). Nine of the items must be revised in order to ensure they are positively directed (3=False, 2=Mostly false, 1= Mostly true, 0=True) (Appendix K). Items were summed for a total score in the present study. High score indicates greater levels of family hardiness.

#### Validity

The concurrent validity was measured by examining the relationship with various indices, validity coefficient ranging from .15 to .23 for coherence, flexibility, and stability (McCubbin et al., 2001). The FHI correlated with Family Time and Routines, .23 (McCubbin et al., 2001) and with FACES II, .22 (Olson, Potner, & Bell, 1982). Construct validity was verified by factor loading that was reported to be in the range of .52 to .85.

Svavarsdottir (1997), using a sample of families of young children with asthma, found that a sense of coherence and general well-being were positively correlated with family hardiness, indicating a higher sense of coherence (r = .75 for the mothers' score, r = .73 for the highest score of the parents, r = .81 for the mean of the parents' score, r = .60 for the fathers' score). Higher reported physical and emotional well-being correlated with higher family hardiness (r = .70 for the mothers' score, r = .60 for the fathers' score) and also suggested that family hardiness was also positively correlated with family adaptation (r = .57 for the parents' highest score, r = .72 for the mean of the parents).

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Campbell and Demi (2000) investigated the relationship among emotional distress, grief, and family hardiness in 20 adult children of missing-in-action fathers. They found the FHI subscale, commitment and control, was negatively correlated with all three Bereavement Experience Questionnaire-Short Form (BEQ-24) subscales, and the BEQ-24 Existential Loss was negatively correlated with two of the FHI subscales, challenge and control.

Family hardiness has been noted as a key variable in influencing family adaptation and family well-being (McCubbin & McCubbin, 1991; Newby, 1996; Svavarsdottir, 1997). Svavarsdottir's (1997) study suggested that family hardiness could predict family adaptation and the well being of mothers and fathers caring for children with asthma. Leske (2003) did not find significant differences in family strengths of hardiness and family well being and adaptation for patients who had trauma after surgery. Leske et al. (1998) suggested that the only significant variable of hardiness (family strength) to influence family adaptation was problem-solving communication. Ladewig et al. (1992) indicated that family hardiness and coping played a more important role in relation to long-term outcomes than for initial response to a crisis event, supporting predictive validity.

### Reliability

The overall internal reliability for the original study for the FHI is .82 with subscale reliabilities of .73 to .82 (Sawin & Harrigan, 1994). Subsequently, studies reported a reliability of .73 for caregivers' burden among family members caring for patients receiving chemotherapy (Carey, Oberst, McCubbin, & Hughes, 1991), .80 reliability for the families of children with developmental disabilities for the total FHI

(Failla & Jones, 1991), and subscale reliabilities from .49 to .77 (Failla & Jones, 1991). For the three subscales, the internal reliabilities were .81, .80, and .65 (McCubbin et al., 2001). Kuo (2000) measured the Chinese Family Hardiness Index with preterm labor families (using Cronbach's alpha) with fathers reported at .81, and .77 for mothers. A test-retest study at one month of families dealing with a technology-dependent chronic illness was .94 (Carey et al., 1991). McCubbin et al. (2001) reported that test-retest reliability was .86. The Cronbach  $\alpha$  of the study was .81. The subscale of commitment was .69, challenge was .62, and control was .56.

There was no normative data on the FHI, but the FHI has been used in various populations of the chronically ill, such as persons with cancer (Mellon & Northouse, 2001; Northouse et al., 2002), disability (Failla & Jones, 1991; Olsen et al., 1999), asthma (Svavarsdottir, 1997), arthritis (Lambert, Lambert, Klipple, & Mewshaw, 1990), and hemodialysis (White, Richter, Koeceritz, & Lee, 2002). A few studies focused on immigrants (Kamya, 1997), and victims of political violence (Campbell & Demi, 2000; Khamis, 1998) and traumatic events (Ladewig & Jessee, 1992; Leske, 2000; Leske & Jiricka, 1998). Family hardiness has been studied in families of children with a cardiac condition and families who have a child with diabetes (H. I. McCubbin et al., 1996).

### McMaster Family Assessment Device (FAD - 60 items)

The McMaster Model of Family Functioning (MMFF) is based on systems, role, and communication theories, and evolved from work with non-clinical families (Sawin & Harrigan, 1995). The model identified six dimensions: problem solving, communication, roles, affective responsiveness, affective involvement, and behavior control. Six of the scales on the FAD reflected the dimensions of family functioning outlined in the MMFF

(Epstein, Bishop, & Levin, 1978). Additionally, Epstein, Baldwin, and Bishop (1983) selected the items most highly intercorrelated, which resulted in the creation of a general functioning dimension, which assessed overall health of the family.

Epstein et al. (2003) identified "family problem solving as a family ability to resolve problems to a level that maintains effective family functioning" (p. 587), "communication as the exchange of verbal information within a family" (p. 589), "family role as the repetitive patterns of behavior by which family members fulfill family functions" (p. 590), "affective responsiveness as the ability to respond to a given stimulus with an appropriate quality and quantity of feelings" (p. 594), "affective involvement as the family shows interest in and values the particular activities and invest themselves in one another" (p. 595), and "behavior control as the pattern a family adopts for handling behavior in three areas-physically dangerous situations, involving meeting and expressing drives and psychobiological needs, and interpersonal socializing behavior" (p. 596).

The present study used the FAD which consisted of 60 items (with seven items added to three of the scales to increase reliability of the original 53-item version) (Bernstein, Garbin, & McClellan, 1983). The scales and dimensions of the FAD included: 6 items for problem solving, 9 items for communication, 11 items for roles, 6 items for affective responsiveness, 7 items for affective involvement, 9 items for behavior control, and 12 items for general functioning. Epstein, Baldwin, & Bishop (1983) developed the FAD-3 in the United States. Responses are made on a four-point scale "strongly agree, agree, disagree, to strongly disagree." One total score ranging from 1 to 4, a lower score corresponds to greater health (Appendix L).

# Validity

The construct validity of the FAD was appropriate (Browne, Arpin, Coey, Fitch, & Gafni, 1990). The FAD scores have been related to family function focused on parenting (McFarlane, Bellissimo, & Norman, 1995), psychological well-being (Byles, Bryne, Bolye, & Offord, 1988; Martin, Rozanes, Pearce, & Allison, 1995; Wenniger, Hageman, & Arrindell, 1993); and to the parent-child relationship scale (Wamboldt, Wamboldt, Gavin, & Mctaggart, 2001). Shek (2002) showed that the FAD scores were significantly correlated with measures of trait anxiety, existential well-being, life satisfaction, and sense of mastery. Kabacoff, Miller, Bishop, Epstein, and Keitner (1990) used oblique multiple group confirmatory factors analysis to show that over 90 % of the FAD items were loaded on factors hypothesized by the McMaster Model. These findings support the construct validity of the FAD. The predicted relationship between the scales of the FAD, FACES (96 items) and the Family Unit Inventory (FUI) provided adequate evidence of the concurrent validity for the FAD (Kabacoff, Miller, Bishop, Epstein, & Keitner, 1990; Van der Putten et al., 1999). The relationship between the FAD and FACES II, a revised version of FACES (30 items), did not correspond to theoretical predictions, but a more linear relationship was obtained.

The FAD was able to discriminate psychiatric patients and healthy employees or university students to support its discriminant validity (Epstein, Baldwin, & Bishop, 1983; Miller et al., 1985; Shek, 2002). Miller et al. (1985) used mean cutoff scores for each subscale, which ranged from 2.1 to 2.4, to discriminate between healthy and unhealthy families. It was able to discriminate between healthy families, and psychiatric families when compared to families rated by clinicians. Lampher (1999) found that students who

were at high risk for suicide ideation scored significantly higher on the FAD, which suggested family dysfunction. Those data supported discriminative validity of the FAD. The FAD has been found to have low correlations with social desirability (r = .06-.19), moderate correlations with global measures of marital functioning such as the Dyadic Adjustment Scale (r = .47), and the Locke-Wallace Marital Satisfaction Scale (r = .59), and theoretically consistent correlations with other measures of family functioning (Miller et al., 1985). Keitner, et al. (1992) and Miller, et al. (1992) reported that the FAD was predictive of recovery from major depression.

### Reliability

The FAD has been found to have high levels of internal consistency ranging from .72 to .92 across a variety of different types of families (Epstein et al., 1983), and acceptable levels of test-retest reliability ranging from .66 to .76 (Miller et al., 1985). Roncone and colleagues (1998) reported that test-retest reliability for the Italian FAD ranged from .69 to .91. Shek (2002) reported that test-retest for the Chinese secondary school students ranged from .52 to .81; and the alpha reliability was acceptable, ranging from .61 to .91 except for affective responsiveness (.44) and behavior control (.56). Chen (2002) reported that the acceptable alpha reliability of the Chinese FAD version ranged from .52 to .82, except for behavior control (.52). Wang and Phinney (1998) reported alpha reliability of .29 to .74 in the evaluation of immigrant Chinese and Anglo-American mothers. The Cronbach  $\alpha$  of this study was .67 for problem solving, .81 for general function, .60 for communication, .62 for roles, .64 for affective involvement, .67 for affective responsiveness, and .36 for behavior control (Table 4).

# Table 4

# Internal Consistency Reliability of Measuring the Subscales of the Family

	Number of Item	Alpha
Problem solving score	6	.67
Communication score	9	.60
General	12	.81
Role	11	.62
Affective responsivenes	s 6	.67
Affective involvement	7	.64
Behavior control	9	.36

Functioning	Described in	the	Current Study (N=126)

The FAD has been used to assess family functioning in different countries such as Australia (Sawyer, Sarris, Baghurst, Cross, & Kalucy, 1988), Hungary (Keitner et al., 1991), Italy (Roncone et al., 1998), the Netherlands (Wenniger et al., 1993), the United Kingdom (Stevenson-Hinde & Akister, 1995), Hong Kong (Shek, 2002; Shek, Lai, & Lai, 1998), and Taiwan (Huang, 1994); and different populations with psychiatric disorders (Friedmann et al., 1997), anorexia nervosa (Gowers & North, 1999), depression (Keitner, 1990; Stein et al., 2000), cardiac rehabilitation (O'Farrell, Murray, & Hotz, 2000), psychopathology (Lieb et al., 2000), traumatic brain injury (Max et al., 1998; Rivara et al., 1996), and adolescence (McFarlane et al., 1995).

The particular strength of the FAD is the number of languages in which the instrument is available, making it possible to study and compare families from a variety

of cultures. Sawin et al. (1994) has recommended the FAD as a convenient, easy, and rapidly administered instrument that is useful in clinical and research settings to evaluate family functioning. It has been translated into twelve languages. Tutty (1995) reported that the FAD holds excellent psychometric properties.

### Summary

Using a descriptive correlation study with a cross-sectional and predictive design, this quantitative research study explored factors associated with family functioning in families with a DMD child. One hundred and twenty-six parents with DMD children participated in the study. The participants answered four separated instruments that measured family health, family hardiness, family support, and family functioning and then a family demographic sheet that included the children's degree of disability. Instruments achieved appropriate alpha internal consistency coefficients. Sample size and power analysis were used for Pearson correlation and multiple regression, with a moderate effect and alpha = .05 selected.

# CHAPTER IV RESULTS

This chapter will report factors associated with functioning among families who have DMD children. These factors included family health, family support, family hardiness, and age when diagnosed with DMD. The demographic characteristics of the subjects, DMD children, families; and the subscales of the instruments reliability were presented in the preceding chapter. The presentation of the results is organized by each aim of the study.

# Data Analysis

Data were analyzed using SPSS (version 11.0) and AMOS (version 4.0). Internal consistency reliability using Cronbach's alpha coefficients ranged from 0.81 to .92 (Table 5), indicating high internal consistency reliability for the instruments used in the present study. Hierarchical multiple regression and path analysis were used to test the model.

Table 5

Measures of Central Tendency for Child Disability, Family Resources (Family

Health, Family Hardiness, and Family Support), and Family Functioning								
Empirical indicator (Instruments)	Items	Alpha	Range 7	Theoretical range	Mean	SD		
Child disability (BI)	10	.88	10-100	0 - 100	38.65	25.40		
Family health (DUKE)	17	.81	29.41 - 100	0 - 100	67.48	15.79		
Family hardiness (FHI)	20	.81	20-58	0 - 60	41.24	7.70		
Family support (FAPGAR)	) 5	.89	0-10	0 - 10	6.63	2.86		
Family Functioning (FAD)	60	.92	1.43 - 2.63	1 - 4	2.10	.29		

The specific aims for the study were as follow:

Aim 1: Describe the child's level of disability, access to care, and family characteristics

*Children's level of disability and access to care.* The age range of the children when diagnosed with DMD was 1-15 years (mean = 6.2, SD = 2.8), which indicated when the children began to have access to professional care (Table 6).

The total Barthel Index score of the children at the time of the study ranged from 10 to 100 (mean: 38.65, SD = 25.4) (Table 5). Thirty-eight (47.5%) of the DMD children had a rated score of 21-60, indicating severe dependency and twenty-eight (35%) of the DMD children had a rated score of 0-20, indicating total complete dependency (Table 6).

*Family characteristics.* Forty-four percent of the families reported annual income of less than \$10,000 (NT\$ 360,000); 10% were over \$30,000 (NT\$ 1,080,000). Low-income families were the majority in this study. Fifty-six percent of parents were employed and 44% were unemployed, retired, or homemakers (Table 6). *Aim 2: Describe for family health, family hardiness, family support, and family functioning* 

*Normative data of family health.* The total family health score rated by the individual parents ranged from 29-100 (mean: 67.5, SD = 15.8) with higher scores reflecting higher functional health (Table 5). Fifty-two percent of the parents reported a family health score greater than 67.7, indicating better health. Twenty-one percent of the parents reported a family health score lower than 55.9, indicating dysfunctional health. The higher the score the better the health. Therefore, most of the parent's (52%) reported that they were overall healthy.

# Table 6

Characteristic		Frequency	%	
Age when	Range = $1-15$ years			
diagnosed with	Mean = $6.2$ years			
DMD	SD = 2.8 years			
	Barthel Index score	a		
Disability Level	0- 20	28	35.0	
·	21-60	38	47.5	
	61 -90	11	13.7	
	>=91	3	3.8	
Annually income <sup>b</sup>	<=10,000	35	43.8	
•	10,001 - 15,000	13	16.2	
	15,001 - 20,000	14	17.5	
	20,001 - 25,000	6	7.5	
	25,001 - 30,000	4	5.0	
	> 30,000	8	10.0	
Employment	Employed	71	56.3	
• •	Unemployed	5	4.0	
	Retired or homemaker	50	39.7	

DMD Children's Disability and Access to Care, and Family Characteristics(N=80)

<sup>a</sup> Total raw score <= 60 indicated severe dependency

Total raw score>60 indicated mild dependency

<sup>b</sup> 1 US\$ = 36 NT\$

The mean scores and standard deviation of the subscales of family health are presented in Figure 3. The mean of physical, mental, and social health scores of the parents were 66.1 (SD = 19.5, range 20-100), 64.1 (SD = 19.7, range: 20-100), and 65.2 (SD = 21.2, range 10-100) respectively; six percent of the parents scored lower than 40 for the three subscales indicating poor physical, mental, and social health. Four percent of the parents scored lower than 40 (mean = 71.5, SD = 21.5, range 20-100) for the self-esteem subscale indicating impaired self-esteem. Twelve percent of the parents scored lower than 50.0 for the perceived health (mean = 78.2, SD = 34.9, range 0-100)

indicating impaired perceived health. Eight percent of the parents scored higher than 50.0 for the pain (mean = 43.3, SD = 26.3, range 0-100) indicating current pain. Eight percent of the parents scored higher than 50.0 for the disability (mean = 7.9, SD = 19.4,



range 0-100) indicating disability. Nine percent of the parents scored higher than 58.3 (mean = 33.9, SD = 19.5, range 0-88.33) indicating anxiety. Seven percent of the parents scored higher than 60 (mean = 37.6, SD = 20.3, range 0-90) indicating depression. Nine percent of the parents scored higher than 57.1 (mean = 35.4, SD = 18.8, range 0-85.72) indicating anxiety-depression.

*Normative Data of Family Hardiness.* The ranged obtained on the total family hardiness scale (FHI) was: 20-58 (mean = 41.2, SD = 7.7) with higher scores reflecting greater hardiness (Table 5). The majority of families reported high hardiness scores (49%), indicating a hardier family, and ten percent of the parents scored less than 32 indicating a weaker family.

Three subscales of family hardiness are presented in Figure 4. The mean of the commitment score was 18.22 (SD = 3.62, range 6-24), seven percent of the parents scored lower than 13 indicating low commitment. Eight percent of the parents scored less than 8 on the challenge subscale (mean = 11.57, SD = 3.1, range 0-18), indicating a low degree of challenge. Five of the parents scored less than 7 on control (mean = 11.38, SD = 3.0, range 4-17), indicating a low sense of control.



*Normative Data of Family Support.* The range obtained on the total family support (FAPGAR) was 0-10 (mean = 6.63, SD = 2.9) with higher scores reflecting more support (Table 5). Over 50% of the parents scored higher than 6, indicating greater family support; 35% of the parents scored less than 6 indicating lower family support.

The mean and standard deviation of the five subscales of family support are shown in Figure 5. Sixteen percent of the subjects scored less than 1 on adaptability, indicating that they lacked the utilization of resources to solve problems; nineteen percent scored less than 1 on partnership, indicating the lack of sharing of decision making and nurturing responsibilities. Thirteen percent scored less than 1 on growth, indicating their lack of emotional maturation and self-fulfillment; ten percent scored less than 1 on affective, indicating their lack of a caring and loving relationship, and eight percent scored less than 1 on resolve indicating the lack of commitment to devote time to other family members.



*Normative Data of Family Functioning*. The total family functioning scores on the FAD ranged from 1.43 to 2.63 (mean = 2.10, SD = .29) (Table 5). Nine percent of the parents scored higher than 2.46, indicating family dysfunction, and 48 % of the parents scored lower than 2.14, indicating positive functioning.

Seven subscales of the FAD are presented in Figure 6. The mean of problem solving was 1.95 (SD = .39, range 1.0-3.67). Ten percent of the parents scored higher than 2.33, indicating worse problem solving, and 45 % of the parents scored less than 2.0, indicating positive problem solving. Four percent of the parents scored higher than 2.56 on communication (mean = 2.11, SD = .35, range 1.0-3.11), indicating worse communication, and 37% scored less than 2.11, indicating positive communication. Six percent of the parents scored higher than 2.64 on role (mean = 2.22, SD = .33, range 1.36-3.18), indicating worse role functioning, and 46% scored less than 2.18, indicating positive role functioning. Ten percent of the parents scored higher than 2.50 on affective



responsiveness (mean = 2.11, SD = .45, range 1.0-3.67), indicating worse affective responsiveness, and 35% scored less than 2.17, indicating positive affective responsiveness. Four percent of the parents scored higher than 2.71 on affective involvement (mean = 2.13, SD = .40, range 1.14-3.14), indicating less affective

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involvement, and 44 % scored less than 2.14, indicating more affective involvement. Six percent of the parents scored higher than 2.67 on behavior control (mean = 2.23, SD = .31, range 1.44-3.0), indicating worse behavior control, and 42% scored less than 2.11, indicating better behavior control. Six percent of the parents scored higher than 2.58 on general functioning (mean = 2.01, SD = .43, range 1.08-3.17), indicating worse general functioning, and 46% scored less than 2.0, indicating better general functioning. *Aim 3: Describe the relationship among child's disability and access to care (age when diagnosed with DMD), family resources (family characteristics, family health, family support, family hardiness), and family functioning* 

Pearson correlation coefficients were used with interval data and with nonnumeric data with dummy variables or dummy coding to explore what factors were associated with family functioning. A correlation matrix among child's disability and access to care, family resources, and family functioning appears in Table 7.

Correlation between child disability and access to care and family functioning. From the correlational analysis, the family functioning score had a significantly small positive correlation with age when diagnosed with DMD (r = .20, p = .02), but was not significantly correlated with child's dependency level (r = .06, p = .52) (Table 7). Detecting the disease early increased family functioning.

Correlation between family characteristics and family functioning. Table 7 shows that the family functioning score has no significant correlation with family annual income (r = .17, p = .06) and parents' employment (r = .06, p = .48) (Table 7). The results indicate that family annual income and parents' employment were not correlated with family functioning.

# Table 7

Intercorrelation Among Child Disability and Access to Care, Family Resources,

and Family Functioning									
Variable	1	2	3						
1 Age when diagnosed of DMD	1.00								
2 Child disability (dependency)	22*	1.00							

2 Child disability (dependency)	22*	1.00			
3 Annual income	.26**	13	1.00		
4 Employment	09	.07	27**	1.00	
5 Family hardiness <sup>a</sup>	21*	.11	16	.05	1.00
6 Family health <sup>a</sup>	.01	05	06	.19*	.51** 1.00
7 Family support <sup>a</sup>	01	08	10	.13	.55** .53** 1.00
8 Family functioning <sup>b</sup>	.20*	.06	.17	06	74**60**66**

\*\* Correlation is significant at the 0.01 level (2-tailed).

\* Correlation is significant at the 0.05 level (2-tailed).

<sup>a</sup> Higher score is higher, better, or greater

<sup>b</sup> Lower score is better

## Correlation of family hardiness, family health, family support, and family

functioning. Table 7 also shows that family functioning had significantly high negative correlation with family hardiness (r = -.74, p = .00), and moderate negative correlation with family health (r = -.60, p = .00) and family support (r = -.66, p = .00). The higher the score of family functioning is, the lower the score on the family hardiness, family health, and family support. The family functioning score is a reverse score. Therefore, healthy family functioning was associated with higher family hardiness, greater family support, and better family health.

4 5 6 7

The variance shared between family functioning and family hardiness was 55% (95% CI for r = .643-.804). The variance shared between family functioning and family health was 36% (95% CI for r = .475-.701). The variance shared between family functioning and family support was 44% (95% CI for r = .597-.783). In other words, the independent variables-family hardiness, family health, and family support- were significant variables accounting for 55%, 36%, and 44% of variance in family functioning, respectively.

In addition, family hardiness had a significantly moderate positive correlation with family health (r = .51, p = .00) and family support (r = .55, p = .00); family health had a significantly moderate positive correlation with family support (r = .53, p = .00). Family hardiness accounted for 26% of the variance of family health (95% CI for r = .383-.639). Family support accounted for 30% of the variance of family hardiness (95% CI for r = .432-.677). Family support accounted for 28% of the variance of family health (95% CI for r = .384-.643).

The high or moderate negative correlation coefficients among the independent variables of family health (r = -.60), family support (r = -.66), and family hardiness (r = -.74) with family functioning suggest the absence of multicollinearity. Indications of multicollinearity are high correlations between independent variables (> .80); if correlations are above .95, there are serious problems (Glantz & Slinker, 1990). These higher negative associations indicated that parents with higher family hardiness, family health, and family support scores reported lower family function scores (meaning healthier family functioning).

*Correlation between child disability, access to care, and family resources.* Table 7 shows that there were no significant correlations between child disability and family annual income, parents' employment, family hardiness, family health, family support, or family functioning. Age when diagnosed with DMD was significantly correlated with family hardiness (r = -.21, p = .02) and family annual income (r = .26, p = .003) but not significantly correlated with family health (r = .01, p = .88) or family support (r = -.01, p = .94) (Table 7). The variance shared between family hardiness and access to care (age when diagnosed with DMD) was 4% (95% CI for r = .037-.361). Access to care accounted for 7% of the variance of family annual income (95% of CI for r = .094-.418).

Correlation between family characteristics and family resources. The correlation of parents' employment with family annual income (r = -.27, p = .002) and family health (r = .19, p = .04) was significantly low (Table 7). However, these three variables were correlated with each other. The results reflect that parents working fulltime report a healthier family when the annual income is over \$15,000.

Aim 4: Determine how the child's level of disability and access to care, family health, family characteristics, family support, and family hardiness predict family functioning

The relationships among the child's disability (dependence) and access to care (age when diagnosed with DMD), family resources (family characteristics, family health, family support, and family hardiness), and family functioning were determined in two ways. First, the Pearson correlation coefficient (dummy coding was applied to transform category data to 0 and 1) was used with significance determined at the .05 level, and second a hierarchical multiple regression procedure was used with significance determined at the .05 level.

In order to determine each predictor variable with the best parameter possible, numeric data (parents' employment and family annual income) were converted to dummy coding (employment coding 1 = employed, 0 = unemployed); family annual income was coded as 1 = <\$15,000, 0 > \$15,000); furthermore, the raw score of the Barthel Index was coded as 1 > 60 to represent mild dependence and  $0 \le 60$  to indicate severe dependence. There were several statistical assumptions to investigate prior to doing the multiple regressions. The assumptions to examine were normality, homoscedasticity (equal variance), linearity, and independence of individual variables and the residuals (Hair, Anderson, Tatham, & Black, 1998).

For each value of the independent variable, the distribution of the dependent variable, family functioning, was normal distribution. The variance of the distribution of family functioning was constant for all values of the independent variables-family hardiness, family health, and family support. The relationship between family functioning and each independent variable was linear, and all observations were independent.

To examine scatterplots and normal probability plots of the residuals of the dependent variable, family functioning, and independent variables, family hardiness, family health, or family support, the assumption of normal distribution with constant variance held; the residuals of family functioning plotted against any independent variable, family hardiness, family health, or family support fell in a band centered around zero with a constant width (null plot) produced a straight line, so the data presented was

consistent with the regression model (Glantz & Slinker, 1990; Hair et al., 1998). The residuals were homoscedasticity and normally distributed about the plane regression line.

In partial regression plots for each independent variable, the equation showed that the relationship of the dependent variable, family functioning, to the independent variable, family hardiness, family health, and family support, were linear. The absence of curvilinear relationships had a significant effect in the regression equation, both in slope and scatter of the points, which were demonstrated in partial plots of the dependent and independent variables in the present study.

All predictive variables were entered in regression analysis and detected multicollinearity. The results fit the assumption of collinearity by the condition index (CI) of each variable that was lower than 30. There were no more than two predictors with coefficient variances over .5; all correlation coefficients less were than .75 and the variance inflation factor (VIF) value of each variable was never over 2 (Table 7 & Table 8). No high multicollinearity evidence can avoid redundant information in the independent variables taken as a whole and a decrease in two variables happens to contain the same information (Glantz & Slinker, 1990).

Based on the theoretical model and the unique contribution of individual predictors on the criterion variable, all variables were significant determinants of family functioning: the total variance of explanation ( $\mathbb{R}^2$ ),  $\mathbb{R}^2$  change, and part correlations in the criterion variable among the cluster of variables having significantly (F value) entered in the hierarchical multiple regressions.

*Hierarchical regression analysis on family functioning*. Hierarchical multiple regression analysis (Table 10) was performed for the first block with all three family

resource clusters entered into the equation model. These were family hardiness, family health, and family support (multiple R = .743 for the predictive variable of family hardiness on family functioning, .802 for adding the second variable of family health, and .817 for adding the third predictive variable of family support, all with/at a p = .00).

The level of accurate prediction is the coefficient of determinants  $R^2$ , compared to the simple regression model value of  $.743^2$  or .5520, which uses only family hardiness; when family health is added to the regression analysis,  $R^2$  increase to  $.802^2$  or .6432. The means inclusion of family health in the regression analysis increases the prediction by 9.08 %. When family support is added to the regression analysis,  $R^2$  increases to  $.817^2$ or .6675, which increases the prediction by 2.42 %.

The first block, which contained family hardiness, family support, and family health, was significant and accounted for 66.8% of the variance in family functioning. This indicated that a higher family hardiness score, higher family support score, and more healthier parents were related to lower family functioning score (meaning better family functioning). In the second block, the child's level of disability and access to care variables resulted in the entry age when diagnosed with DMD. The child's disability did not enter the equation. The age when diagnosed with DMD (multiple R = .824, p = .00) showed significant bivariate correlation with family functioning. The age when diagnosed with DMD was entered in the equation, the added R<sup>2</sup> changed to .011, and the R<sup>2</sup> was .679. The second predictor accounted for 1.1 % of variance in family functioning after controlling for the first three predictors. The age when diagnosed with DMD was related to family functioning, indicating that an earlier diagnosis of DMD led to earlier

# Table 8

Hierarchical Multiple Regression for Family Stress with Family Resources on Dependent Variable Family Functioning

(N=126)

Mod	el	R	R <sup>2</sup>	Adjust R	2 F	В	SE	Beta	Т	Part cor (sr)	VIF
1	(Constant) Family hardiness Family health Family support	.817	.668	.660	81.82*** (df=3,122)	3.27 020 004 030	.088 .002 .001 .007	482 198 290	37.217*** -7.316*** -3.306** -4.366***	382 160 228	1.593 1.536 1.625
2	(Constant) Family hardiness Family health Family support Diagnosed age	.824	.679	.669	64.08*** (df=4,121)	3.17 020 004 030 .011	.100 .002 .001 .007 .005	445 212 303 .110	31.807*** -6.599*** -3.304** -4.592*** 2.066*	340 170 236 .106	1.713 1.554 1.638 1.076

\*P<.05, \*\*p<.01, \*\*\*p<.001

access to professional care that was related to lower family functioning scores (meaning better family functioning).

In the third block of variables, none of the two family characteristics entered the model. As a result of the shared variance, the variable (multiple R= .824, p= .00) significantly correlated family functioning in the final regression model, including three family resources clusters and one disabled child and access to care cluster that explained 67.9 % of the variance in family functioning. About 67.9 % of the variance in the criterion variable family functioning was explained by first family hardiness, family health, and family support (66.8%), and second by age when diagnosed with DMD (1.1%). All results came out to be significant. The change R<sup>2</sup> was also significant for the second step (F = 64.08, df = 4, 121, p = .00). According to the data of part cor (squared semi-partial)-sr, the total variance explained by the four independent variables is a unique variance explained (part cor<sup>2</sup>-sr<sup>2</sup>) by family hardiness .1156, family health .0289, family support .0557, and age when diagnosed with DMD .0112.

The overall equation for predicting family functioning was:

Family functioning = 3.17- .02 (family hardiness)- .004 (family health)- .03 (family support)+ .01 (age when diagnosed of DMD). The final regression statistics for the full model was significant (R = .824, F <sub>(4, 121)</sub> = 64.08, p = .00).

Aim 5: Test the model of family stressors, resources, and functioning

In the initial path analysis, standardized beta coefficients indicated that family hardiness, family support, family health, and age when diagnosed with DMD were the independent predictors in the regression equations on family functioning. In addition, family health and family support were predictors in the regression on family hardiness, and family health was a predictor in the regression on family support. However, the age when diagnosed with DMD was not a predictor in the regression on family health and family support. The path analysis was not a good fit for the model ( $x^2 = 0$ , df = 0, RMSEA = .44).

The model that was tested was drawn with the path analysis in figure 7. The standardized regression coefficient (beta) was used to examine the total effect of family resources (family hardiness, family health, and family support), and family stressors (age when diagnosed with DMD), which had both direct and indirect effects on family functioning. The standardized regression coefficient (path coefficient) may be used to decompose the correlation in the model into direct and indirect effects, corresponding to direct and indirect paths reflected in the arrows in the model. This is based on the rules of a linear system. The total causal effect of variable i on j is the sum of the values of all the paths from i to j (Norris, 2001).

Considering "family functioning" as the dependent variable in the model and the independent variables -age when diagnosed with DMD, family hardiness, family health, and family support, the indirect effect was calculated by multiplying the path coefficients for each path from each independent variable oneself to family functioning. The path coefficients suggest that family hardiness (beta = -.45, p = .00), family support (beta = -.30, p = .00), and family health (beta = -.21, p = .001) were directly predicted to have a positive effect on family functioning. The direct effect of age when diagnosed with DMD (beta = .11, p = .04) was also directly predicted to have a positive effect on family functioning. There was an indirect effect of age when diagnosed with DMD on family functioning. There was an indirect effect of age when diagnosed with DMD on family functioning by two paths (a) family hardiness, indicated by -.21 x (-.45) or .0945,



and (b) family hardiness through family health, indicated by - .21 x .32 x (-.21) or .0141. The indirect effect of family hardiness on family functioning via family health was .32 x (-.21) or -.0672. The indirect effect of family support on family functioning was - .3579 via three paths (a) family heath, indicated by .35 x (-.21) or - .0735, (b) family hardiness, indicated by .55 x (-.45) or - .2475, and (c) family hardiness through family health, indicated by .55 x .32 x (-.21) or - .0369. There was no indirect effect of family health on family functioning.

In sum, the standardized total effect on family functioning by age when first diagnosed with DMD was .22, family hardiness was - .52, family health was - .21, and family support was - .66 (See Figure 7 or Table 9). Forty-three percent (.0945/.22) of the effect of early disease diagnosis on family functioning was under the influence of family hardiness and 6.4% (.0141/.22) was under the influence of family hardiness through family health; 50% (.11/.22) of the total effect was mediated. By this computation of the total effect of family hardiness on family functioning, 13.5% (- .0672 /- .52) of the total

# Table 9

Standardized Direct and Indirect Effects for Family Stressors, Family

	Independent variable									
Dependent variable	Age when	Family	Family	Family						
	diagnosed with	hardiness	health	support						
	DMD									
Family functioning										
Total	.22	52	21	66						
Direct	.11	45	21	30						
Indirect	.11	07		36						
Family health										
Total	07	.32		.53						
Direct		.32		35						
Indirect	07			.18						
Family hardiness										
Total	21			.55						
Direct	21			.55						

Resources on Family Functioning, Family Hardiness and Family Health

effect was mediated by family health. Eleven percent (-.0735/- .66) of the effect of family support on family functioning was under the influence of family health, 37.9% (- .2475/.66) was under the influence of family hardiness, and 5.6% (- .0369/-.66) was under the influence of family hardiness through family health. Fifty-four percent (- .3579/- .66) of the total effect of family support on family functioning, -.3579, was mediated by family health, family hardiness, and combining family hardiness with family health.

In addition, the direct effect of age when diagnosed with DMD (beta = -.21, p = .02) was predicted to have a negative effect on family hardiness. The path coefficients indicating the standardized direct effects, as well as the standardized total effect of age when diagnosed with DMD on family hardiness was -.21. The standardized direct effect,

as well as the standardized total effect of family support on family hardiness, was .55. The standardized direct effect, as well as the standardized total effect of family hardiness on family health was .32. Path analysis decomposed total effect of family support on family health (.53) into direct effect of family support on family health (.35) and the effect of family support that was indirect through family hardiness (.18). The standardized indirect effect, as well as the standardized total effect of age when diagnosed with DMD through family hardiness on family health, was - .07 (see Table 9).

# Conclusion

The researcher used path analysis to test a model of the effects of family resources and child disability and access to care on family functioning in families with DMD children. A higher score on the perceived health measure indicated a higher level of positive health and a lower level of negative health. A higher score of family support indicated higher support to the DMD family. A higher score of family hardiness indicated greater levels of family hardiness. A higher score of family functioning indicated lower levels of family functioning. Results of hierarchical regression analysis of possible predictors of family functioning in a sample of 126 parents with DMD children showed higher scores of family hardiness, family health, and family support; earlier diagnosis of DMD was associated with healthier family functioning (lower score of family functioning) and higher family hardiness. The parents who reported higher scores of family hardiness and family support were associated with better health. Results of the path analysis revealed that family hardiness, family health, family support, and age when diagnosed with DMD accounted for 68% of the variance of family functioning; family hardiness and family support accounted for 35% of the variance of family health; and family support and age when diagnosed with DMD accounted for 34.5% of the variance of family hardiness.

### CHAPTER V: DISCUSSION

Based on the results from this study of parents of children with DMD, the major components of the Conceptual Model of Family Stressors, Resources, and Functioning were supported. In this chapter, the findings of the study will be discussed, first by examining the major concepts and variables in the context of findings from other studies, followed by the implications of the major findings from this study.

Family Stressors, Resources, and Functioning Status Family Stressors

The age of the child's diagnosis with DMD varied from 1 to 15 years. Because one-third of the children were diagnosed late, their access to professional care was limited. Eighty-three percent of families needed to completely assist with their daily activities. Families who received accurate information about the disease too late had difficultly coping with the chronic disability. Overload and uncertainty caused families much stress.

# Family Resources

*Family health.* These parents had less physical, mental, and social health; lower self-esteem and perceived their health poorer than middle age female and male policyholders of a health insurance company (Parkerson, 2002). Except for physical health, the parents reported lower scores on all positive health subscales compared with 50-65 year old policyholders (Parkerson, 2002). They also reported more anxiety, depression, pain, and disability (Parkerson, 2002). Nereo et al. (2003) found that

mothers of children with DMD have higher stress than a normative group because of problem behaviors of children with DMD, especially in social interactions (Nereo et al., 2003). These findings suggest that the parents in this study may have had more tension and poorer health status. The parents not only had physical overload because of caring for the children who needed complete assistance, but they also experience anxiety and depression because of their children's behavior problems and the progressive, and lifethreatening nature of DMD.

*Family hardiness.* The mean of family hardiness for 126 parents with DMD children was less than the mean of mothers and fathers with young children of asthma; mothers and fathers of children with cardiac conditions; and mothers and fathers of children with diabetes (McCubbin et al., 2001). These differences between children with DMD and other difficult conditions in different cultures should be explored, as well as the differences in the mean scores in different diseases or phenomenon within the same country. Less than 50% of the families in the study were hardy; families with lower hardiness scores had difficulty finding resources, were anxious or depressed, and had health problems.

# Family Support

Parents in this study had lower scores of family support than did families with and without other chronic conditions reported in the literature (Gwyther et al., 1993). Thirty-five percent of the parents in the study scored lower than 6 on the FAPGAR, a higher rate than Smucker, Wildman, Lynch, and Revolinsky's (1995) study, which found that only 15 % of families with well children score this low. Furthermore, Gardner's et al (2001)

found that 31% of families with a child having psychosocial problems scored below 6 on the FAPGAR.

### Family Functioning

For every dimension except behavior control, these parents had better functioning than clinician-rated healthy and unhealthy families; but communication score and affective involvement were worse than the healthy group (Miller et al., 1985). Hawley's (2000) study emphasized that inability to communicate was the major obstacle to family functioning. And in the early stage, several scholars have utilized communication in their research framework (Epstein et al., 1978; Olson et al., 1979). Psychiatric families had a significantly higher score on all the FAD subscale scores except behavior control (Miller et al., 1985). Chen and Liu (1989) reported that there were differences associated with problem solving, communication, role, affective involvement, behavior control, and general function between normal families and psychiatric patients' families.

Relationships Among Family Stressors, Resources, and Functioning Family Stressors, Family Resources, and Family Functioning

Overall, of all the major variables investigated in this study, earlier detection of DMD, and higher scores on family hardiness, family health, and family support were associated with better family functioning. This suggests that early diagnosis of DMD in children may provide early access to professional care and the resources parents need to adapt and function well. It may be, however, that highly functioning families are more likely to seek on encourage evaluation of their child's grass motor delay sooner than families with poorer functioning, a possibility this study did not address, given its cross-sectional design.

The child's disability level was not significantly correlated with family functioning, family hardiness, family support, or family health. While this finding was surprising, others have found that higher family functioning was not related to the child's disability status (Kim, 2002; Olsen et al., 1999); whereas others reported an inverse relation between family functioning and severity of adolescent's and young adult's injury or disability status (Magill-Evans et al., 2001). Failla et al. (1991) found that mothers who had a developmentally disabled child showed a significantly positive relationship between family hardiness and family functioning; and between family hardiness and coping behavior, which are related to strengthening the relationships within the family. The present study did not find that the degree of the child's disability was positively related to family hardiness, family support, and family health. This may have been due to an increasing awareness and knowledge of the children's condition, and unlimited access to healthcare. The significant relationship between child health, family health, and family functioning has been found in other studies (Shek, 2002; Wells & Whittington, 1993) and reinforces the notion that the promotion of child health is central to all interventions in families with DMD children.

In addition, a strong positive correlation among family hardiness, family health, family support, and healthy family functioning suggests evidence of construct validity or criterion validity of family functioning. Bristol (1987) and Bennet and DeLuca (1996) reported that support from significant others and social support has a positive affect on family functioning. Wu and Huang (1997) reported that there was a significant relationship between family functioning and expressed emotion for the caregivers of family members with psychiatric conditions. Heru and Ryan (2003) found that family

functioning and depression were closely associated with the caregivers of patients with chronic or recurrent mood disorders. The findings from this study suggest that lower family functioning was associated with less family hardiness and worse psychological health, which in turn was associated with less family support and a later age of diagnosis with DMD.

# Family Characteristics, Family Hardiness, Family Support, and Family Health

The socioeconomic factors of family annual income and parents' employment were not found significantly related to family functioning. These may not have been the best parameters to assess because of centering the same classification of the sample. These findings are similar to previous studies in that none of the family characteristics have been found to be associated with family functioning (Lieb et al., 2000; Merikangas, Avenevoli, Dierker, & Grillon, 1999). However, parental education, number of children, severity of the child's condition, and the child's diagnosis have been found to correlate significantly with family hardiness, social support, stress, and coping (Huang, 1996).

The significant correlation between family support and family hardiness found in this study supports Olsen et al. (1999) and McCubbin et al. (1996) who found that family support was positively correlated with family hardiness. However, there was no evidence that income was related to family hardiness and family support. The DMD children from families with low annual income and low level of employment rate in this study were eligible to receive government support for special education, rehabilitation, and other specialized care. This may have contributed to their resiliency.
# Determining the Predictive Variables Effect on Dependent Variable Predictive Variables Effect on Family Functioning

Family stressors. The child's disability level did not enter the equation model of Family Stressors, Resources, and Functioning, which indicated that the child's disability level was not a significant predictor of family functioning. The child disability was not associated with family hardiness, family health, and family support. The level of dependency of the child did not significantly enter the equation of hierarchical multiple regressions on the family functioning. One explanation for this finding is that by the time of the study parents had already adjusted to their child's diagnosis, making it possible for them to develop healthy family communication and a positive parenting style.

Access to care was a significant positive predictor of family functioning, suggesting that early detection of the disease may have allowed the family access to needed services and support. Fifty percent of the total effect of access to care was an indirect effect on family functioning. This result suggests that both family hardiness and family hardiness combined with family health, is an important mediator for access to care on family functioning.

*Family characteristics*. Sociodemographic variables were found to play an unimportant role in explaining the dependent variable, family functioning, in the study. Of the family demographic characteristic domains, parents' employment and family annual income in this study were not consistent with other studies which found that income was negatively related to family functioning or well-being (Friedmann et al., 1997). The other characteristics were not considered in this study because the sample size was not large enough to use more than eight variables so the results could not be

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inferred and generalized (Hair et al., 1998; Thorndike, 1978). The low level of income reported from respondents may have skewed the results of the study than if the sample was more representative of the general population. However, all the children in this study had insurance coverage for their DMD-related health care.

*Family health.* The findings in this study were consistent with other studies which found that family health or psychological morbidity was directly or indirectly related to family functioning (Magill-Evans et al., 2001), but was not support most studies' finding that severity of child disease was related to family functioning (Fornari et al., 1999; Gowers & North, 1999; King et al., 1993; Luescher et al., 1999; Pal et al., 2002; Stein et al., 2000; Tamplin & Goodyer, 2001; Tamplin et al., 1998). In this study family health only directly affected family functioning; there was no indirect effect on family functioning. Family health contributed to a small percent of unit variance explaining family functioning. But family health was a mediating factor for family hardiness, family support, and access to care in their effects on family functioning (see figure 7).

This study did not explore the effect of subscales of family health on family functioning. Stein's (2000) findings that fathers of children with major depression scored significantly lower on the FAD scales of behavioral control and general functioning, compared to the fathers of other high-risk children; and mothers of high-risk children had significantly lower scores on the roles and affective involvement dimensions of the FAD compared with mothers of low-risk children. Heru and Ryan (2003) found that family functioning and depression were closely associated with the caregivers of patients with chronic or recurrent mood disorders. *Family hardiness.* Huang (1996) found that family hardiness was a stronger predictor of family stress, coping, and family functioning than social support in families of children with developmental disability. Failla et al. (1991) found that family hardiness, family stressors, and functional support could predict the mothers' satisfaction with family functioning. They suggested that family hardiness could diminish the effects of stress, increase the use of support, and facilitate adaptation.

Results of this study suggest that the desirable outcome -family functioning- is associated with family hardiness, and that family hardiness mediated family functioning through age when diagnosed with DMD are important findings. One explanation of these findings is that parents' experience with difficult challenges at an earlier time in their family's life may have resulted in parents who were stronger and more resilient because of access to professional care or because they were already hardy prior to their child's diagnosis. Family hardiness was the major mediating factor for age when diagnosed with DMD and the effect of family support on family functioning. Family health was a mediating factor for family hardiness on family functioning (see Figure 7).

Hardy people have a tendency to see life events as less stressful than others, an ability to cope more effectively with stressful events, and a more conscientious approach to health care. The results suggest that psychosocial interventions that focus on promoting family functioning must not only address the challenges and internalized stigma that affected energy and self-esteem, but should also promote the development of health-seeking behaviors. Otherwise, unhealthy family functioning may result in families with lower level of commitment to themselves, less sense of control in their lives, and less of a tendency to view change as a positive life challenge.

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*Family support.* Family support may be a crucial factor for the family of children with a disability (Bailey, Skinner, Rodrguez, Gut, & Correa, 1999; Haveman, 1997; Sayger & Bowersox, 1996). Support from significant others and social support both formally and informally positively affect family functioning (Bennett & DeLuca, 1996; Bristol, 1987). Olsen et al. (1999) proposed that family support was positively related to family hardiness for both fathers and mothers with disabled children. A large body of evidence suggests that social support is an important factor in mediating stress and enhancing coping in families of children with disabilities, with considerable support networks outside of the family (Dyson, 1997; Hadadian, 1994; Trivette & Dunst, 1992).

While significant, family support contributed to a small percent unit variance explaining of family functioning (see Table 8) after controlling the other independent variables. But the standardized total effect of family support on family functioning was high because family health and family hardiness were mediators to influence family supports indirect effect on family functioning. Dunst, Trivette, and Deal (1988) reported that informal support results in more optimal family functioning, while Haveman et al. (1997) emphasized the influence of formal support for families of children with disabilities. Other researchers have also found that extra-family resources are important for healthy family functioning.

*Summary*. The most important findings of the Family Stressors, Resources, and Functioning Model were that four variables directly or indirectly affected family functioning: age when diagnosed with DMD, family hardiness, family support, and family health had direct effects on family functioning and the first three variables had indirect effects on family functioning. However, all four variables accounted for 68% of the variance in family functioning. Failla and Jones (1991) reported that family hardiness, functional support, family stressors, and parental age among families of children with developmental disability accounted for 42% of the variance in family functioning.

In reviewing the study data about direct and indirect effects of predictors on family functioning, the path coefficient between family hardiness and family functioning was in a higher range and in the direction predicted. The association between these two variables was strong when the total combined score and the family functioning score were used. This result is similar to a study that found family hardiness was positively associated with family functioning in parents of children with asthma (Donnelly, 1994). Others have argued that family hardiness and family health are related to family functioning in term of the child's health and severity of symptom (Carpiniello, Piras, Pariante, Carta, & Rudas, 1995; Luescher et al., 1999; Walker, Van Slyke, & Newbrough, 1992). The positive relationship between hardiness and healthy family functioning suggests that parents of children with DMD who had greater internal strengths to endure stressors also had greater health. In addition, from a health promotion perspective, the findings support the development of family hardiness through family support services that could be incorporated into health promotion programs in the long-term.

## Predictive Variables Effect on Family Health

Family support and family hardiness were predictors, which were direct and indirect effect on family health. One report suggests that families with low family support had significantly higher psychosocial dysfunction than families with adequate family support (Murphy et al., 1998). Family hardiness was found to influence perceived psychological distress; and supportive social resources might directly affect functioning among parents with disabled children (Sloper, 1999), parents with a developmental disability (Failla & Jones, 1991), and family of children with disability (Snowdon et al., 1994). Bigbee (1992) found that hardiness may have a direct effect as well as a buffering effect in the stress-illness relationship. Several authors proposed that hardiness functions as a buffer or mediating factor that may enhance coping or reduce harmful effects of stress (Failla & Jones, 1991; H. I. McCubbin et al., 1996). In addition, family hardiness also has been found to be a significant factor in health promotion (Donnelly, 1994). These findings suggested that health promotion should focus on the creation of interventions to strengthen family hardiness and support.

#### Predictor Variables Effect on Family Hardiness

Access to care and family support influenced family hardiness (see figure 7). The results of hierarchical multiple regressions showed that earlier diagnosis with DMD was associated with healthier family functioning and better family hardiness. All the total effects of age when diagnosed with DMD and family support on family hardiness were derived from their individual influence of direct effect on family hardiness. *Summary* 

The study was based on the conceptualization of several factors thought to influence the variables of family stressors (child disability and access to care), family resources (family hardiness, family support, family health, and family characteristics) on the outcome variable family functioning. Results of testing supported the predicted relationships between the age when diagnosed with DMD and family functioning; they also supported the predicted relationships between family hardiness and family functioning, family health and family functioning, and family support and family functioning. The findings support the recommendation that family stress, family resources, and family functioning be operationalized as complex univariates.

Limitations of the study included the non-probability and cross-sectional design. The sample was drawn mostly from TMDA. This sample reflected the middle-aged parents of DMD families at that time. At the time of the study, a great majority of the sample had benefited resources and information from the TMDA. The findings of this study might have been different if the respondent had been drawn from non TMDA families with DMD children, where the level of disability, family annual income, and family hardiness might have differed from the sample. Although duration of illness and length of time in treatment did not correlate, or predict family hardiness, family support, family health, and family functioning, conclusions about time effects are limited by the cross-sectional design of the study.

The majority of the respondents had such similar characteristics that made it difficult to do multivariate analysis for the demographic independent variables, only by dummy coding to dichotomy categories. Thus, the participants may have over reported family support; and underreported family hardiness and family functioning. Limited attention has been given to families with DMD children, although they are a particularly vulnerable population. Other families not participating in TMDA may have different responses.

Sample size was not enough to explore the relationship and do the hierarchical regression among the subscales of each measurement. All these factors limit the generalizability of the findings to other samples of families whose children are reported as disabled. This study needs to be replicated with a longitudinal design that samples

both men and women who have a handicapped child. The study did not provide a picture of family functioning from the perspective from all family members. Therefore, it is not known how family functioning or family health would correlate with family hardiness as reported by other family members.

## Contribution to Nursing Knowledge

Knowledge of the Resilience Model and the Model of Family Stressors, Resources, and Functioning can be useful in nursing practice. A family centered and strength-based approach is needed with a focus on resources and skills to provide loving and caring relationships, effective communication, and empowerment of the family and community in building resilience. Social networks have an effect on family adaptation. Professionals can explore family support, family communication, and family hardiness in families with children with disabilities to help families develop insights and behaviors associated with hardiness. The Model can be used to evaluate outcomes of interventions that are designed to minimize threats to family integrity and to facilitate healthy adaptation in caring for a child with special needs. These findings further suggest that interventions are needed to improve communication skills, affective expression, and behavior control training.

## **Clinical Practice Implications**

The findings from this investigation provide some important targets for nursing practice with families who have disabled children. From a clinical perspective, this model addresses many of the variables that nurses confront when evaluating family nursing interventions. Clinicians who use the Family Resiliency Model or the Family Stressors, Resources, and Functioning Model could modify their interventions to build on the family's strengths and improve functioning. For example, it would be reasonable to

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assess parents' health status and focus interventions on clinical services that are convenient and affordable. It would be important to provide training on communication skills, affective expression, and parenting skills to alleviate anxiety and depression, and improve self-esteem and mental-social health. This training should take place over several weeks to allow time for the development of trust and therapeutic relationship.

The culmination of family data yields a family nursing diagnosis that may be focused on the individual parent and how he or she might help meet illness demands, or it may address the family's need for information, resources, problem-solving, education, or role negotiation. The importance of the earlier detection of the disease for faster to access to professional care needs to be emphasized, including prenatal and neonatal screening programs.

#### Implication for Nursing Education

Nursing educators should foster knowing about DMD, genetic counseling, screening, health promotion, as well as provide students with the opportunity to assess and care for DMD children and their families. With this foundation, students will begin to appreciate the need for family, school, and community interventions that can support DMD children and their families. Findings from this study can enhance curricula related to family strength-based approaches to children with disabilities or chronic illnesses and their families.

# Contributions of Family Process Research

A systemic view of hardiness and family functioning is important to understand how individuals, couples, and families cope and adapt through crisis and adversity. The family resilience framework provides theoretical understanding about how family

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resiliency and family functioning are related. This understanding can usefully inform our efforts to strengthen families in distress. The search for family resiliency and family functioning should identify key processes that can strengthen each family's ability to overcome the challenges they face in their particular life situation. Family resilienceoriented interventions in clinical practice should build on the principles and techniques common among strength-based approaches and should be systematically evaluated. Using psychometrically distinct measures of problem solving, communication, and affective responsiveness would strengthen future studies.

This study used instruments that had been previously used with other populations. The results suggest that the responses of parents of children with DMD fall within the normal range when compared with other populations. An important outcome of this study was the information on the cross-culturally validity of the instruments used. Yet the study also challenges researchers to pursue the development of instruments that are more culturally sensitive and cultural competent. Longitudinal studies have suggested that in some families of children with disabilities, symptoms of distress are chronic and persistent. Further research should begin to focus on specific interventions to improve family functioning, especially in families with later access to care.

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Appendix A Letter of Permission I: KMU Hospital



高雄醫學大學附設中和紀念醫院 807高雄市三民區十全一路100號 電話:07-3121101 傳真:07-3213931 KAOHSIUNG MEDICAL UNIVERSITY CHUNG-HO MEMORIAL HOSPITAL 100 Shih-Chuan 1<sup>st</sup> Road, Kaohsiung 807, Taiwan TEL: 886-7-3121101 FAX: 886-7-3213931

The Hahn School of Nursing and Science University of San Diego 5998 Alcala Park San Diego, Ca 92110 U.S.A.

March 27, 2003

To Whom It May Concern:

We agree that Ms. Jih-Yuan Chen to do the data collection at Kaohsiung Medical University Chung-Ho Memorial Hospital (KMUH) for her dissertation research project entitled: "Functioning among Families with a Child Having Duchenne Muscular Dystrophy" during June 2003 to September 2003.

If you have any questions, please do not hesitate to contact with me.

Yours sincerely.

Shen-Long Howng, M.D., D. Med. Sc. Superintendent Kaohsiung Medical University Chung-Ho Memorial Hospital

#### Appendix B Letter of Permission II: TMDA

Jih-Yuan Chen The Hahn School of Nursing and Science University of San Diego 5998 Alcala Park San Diego, Ca 92110 e-mail: <u>jihc@sandiego.edu</u> Tel: 1-858-278-4201

I have read the information enclosed and have had it approved by the agency IRB for the dissertation research project entitled: Functioning among families with a child having Duchenne Muscular Dystrophy."

I agree that data collection can occur at Taiwan Muscular Dystrophy Association during June 2003-September 2003

2003 3.30

V

Date

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Signature Yung-Hua Liu President, Taiwan Muscular Dystrophy Association 58, 3F-3, Chiou-Zuo 1st Road, San Ming District Kaohsiung, Taiwan, ROC 80708 E-mail address: <u>tmda168@ms22.hinet.net</u> Tel: 07-3801000

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#### Appendix C Letter of Permission III: Social Worker

19:61 cUL BURE HARL

Jih-Yuan Chen The Hahn School of Nursing and Health Science University of San Diego 5998 Alcala Park San Diego, Ca 92110 U. S. A. Į

June 3, 2003

I agree to help that Ms Jih-Yuan Chen to do the research "Functioning among Families with a Child Having Duchenne Muscular Dystrophy". I will assist to counsel and support the families of children with Duchenne Muscular Dystrophy if the families have emotional risks during their processes of participation in the study.

Hui-Fang Wong Social Worker, Taiwan Muscular Dystrophy 58. 3F-3, Chiou-Zuo 1" Rd, San Ming District E-mail address: imda 168@ms22.hinct.net Tel:07-380-1000

June 3. 2003 Date

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## **Rhode Island Hospital**

A Lifespan Partner Letter of Permission IV: Family Assessment Device

593 Eddy Street Providence, RI 02903

## RECEIPT

TO: Jih Yuan Chen 5998 Alcala Pk San Diego, CA 92110

<b>DATE:</b> February 18, 2003 <b>REFERENCE #</b> : CK# 1078
--

QUANTITY	ITEM	COST
1	McMaster Family Assessment Device	\$40.00
1	McMaster Family Assessment Device – Mandarin Version	\$5.00
	TOTAL AMOUNT PAID:	\$45.00



## BROWN UNIVERSITY Providence, Rhode Island 02912

Enclosed please find the FAD packet that you ordered. You have permission to duplicate the copyrighted Family Assessment Device, the manual scoring sheet and instructions, and the Family Information Form. We may contact you in the future to receive your feedback on the instrument.

Thank you for your interest and good luck in your future project.

Sincerely,

Christine E. Ryan, Ph.D. Director, Brown University Family Research Program Potter 3 Rhode Island Hospital 593 Eddy Street Providence, RI 02903

#### Appendix E Letter of Permission V: Duke Health Profile



DUKÈ UNIVERSITY MEDICAL CENTER Department of Community and Family Medicine Office of the Chairman

March 28, 2003

Jih-Yuan Chen 10963 Vivaracho Way San Diego, Ca 92124

Dear Jih-Yuan Chen,

This is to document that you have our permission to use the Duke Health Profile (DUKE) in your dissertation.

Best wishes,

George R. Parkerson, Jr., MD, MPH Professor of Community and Family Medicine

#### Appendix F Letter of Permission VI: Family Health Index



## KAMEHAMEHA SCHOOLS Office of the Chancellor and Chief Executive Officer

April 30, 2003

Jih-Yuan Chen 10963 Vivaracho Way San Diego, California 92124

Dear Mr. Chen:

On behalf of the developers and copyright holders of the Measure(s) that you have requested:

1) FHI: Family Hardiness Index

I would like to confirm our granting of permission to utilize this instrument for this particular investigation/study/project.

There will be no charge relating to this permission by virtue of your having required the book/CD entitled <u>Family Measures: Stress, Coping, and Resiliency</u> and have registered it accordingly.

This permission is also granted with the understanding that any revisions of these measures (e.g. language translation, etc.) will be sent to this office in its complete form to be distributed to others who may be interested in your revisions/translations.

In all cases the revisions, adaptations and the original measures, the copyright holders will remain the same as the original and also remain a property of the Kamehameha Schools and the Ke Ali'i Pauahi Foundation.

Finally, it is required that will use appropriate citation for the measure in the publication, dissertation, thesis or book. The citation that is expected in all cases will be "Published in Hamilton I. McCubbin, Ann Thompson, Marilyn McCubbin (2001) <u>Family Measures: Stress,</u> <u>Coping, and Resiliency</u>; Kamehameha Schools and Ke Ali'i Pauahi Foundation, Honolulu, Hawai'i."

Respectfully,

(Hamilton I. McCubbin, Ph.D. Chancellor/CEO 國立臺北護理學院 中華民國臺灣省臺北市 北投區 112 明德路365號 NATIONAL TAIPEI COLLEGE OF NURSING NO. 365, MING TE ROAD, PEI TOU 112, TAIPEI TAIWAN, REPUBLIC OF CHINA

March 10, 2003

Jih-Yuan Chen 10963 Vivaracho Way San Diego, CA 92124 U.S.A.

Dear Miss Chen:

You have my full permission to administer my translated Chinese version of Family Hardiness Index for use in your research. Accurate credit must be given to its source where used or described in publications. I wish you success in your research and look forward to hearing the results of your investigation. Contact me any time if you have questions regarding the application of the Family Hardiness Index.

Sincerely,

Su-Chen Kuo Associate Professor Graduate Institute of Nurse-Midwifery National Taipei College of Nursing Taipei, Taiwan, R.O.C.

#### Appendix G

#### Letter of Permission--Family APGAR Scale

Date: Mon, 9 Jun 2003 17:00:53 -0400

From: Chuck Williams < Chuck.Williams@dowdenhealth.com>

To: jihc@sandiego.edu

Subject: Family APGAR

This message was written in a character set other than your own. If it is not displayed correctly, click here to open it in a new window.

Dear Ms Chen,

We are pleased to grant you permission to use the Family APGAR in your dissertation and ask only that you cite the reference thus: "Smilkstein G. The Family APGAR: A proposal for a family function test and its use by physicians. J Fam Pract 1978; 6:1231-1239."

Regards,

**Chuck Williams** 

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Charles Williams Executive Editor Journal of Family Practice 201-782-5708

#### Appendix H

#### **Demographic Sheet**

I. Family Demographic and Medical Characteristics

Each parent should complete the instrument separately. Please mark an "v" in the box " $\Box$ " that best answers each question about your family:

Your age: years Your education level:  $\Box$  6 years  $\Box$  9 years  $\Box$  12 years  $\Box$  15 years  $\Box$  >=15 years Your gender:  $\Box$  male  $\Box$  female Your ethnicity:  $\Box$  Taiwanese □ Chinese □ aboriginal 🗆 Haika Your relationship with DMD child:  $\Box$  mother  $\Box$  father Your marital status: 
married 
separated 
widowed 
divorced If married, quality of marital relationship:  $\Box$  excellent  $\Box$  good  $\Box$  fair  $\Box$  poor Your employment status:  $\square$  employed  $\square$  retired  $\square$  homemaker Classification of your occupation: 
□ labor 
□ technique 
□ government officer  $\Box$  professional  $\Box$  business  $\Box$  farmer Your family annual income:□ <10,000 □<15,000 □<20,000 □<25,000 □ =<30,000 □>30,000 Satisfaction with child's medical care:  $\Box$  yes  $\Box$  no Living location: 
□ rural 🗆 urban 🗆 municipal Religion:  $\Box$  Buddhism  $\Box$  Taoist  $\Box$  Christian  $\Box$  Catholic  $\Box$  none  $\Box$  others, Family structure:  $\Box$  nuclear family  $\Box$  extended family Family development stage:  $\Box$  family with preschoolers  $\Box$  family with school age children  $\Box$  family with adolescents Number of other children:  $\Box 1 \quad \Box 2 \quad \Box 3 \quad \Box > 3$ Health condition of other children:  $\Box$  excellent  $\Box$  good  $\Box$  fair  $\Box$  poor explain: Family history of psychiatric disorder:  $\Box$  yes  $\Box$  no Please answer the following questions about your child with Duchenne Muscular Dystrophy:

Child's age: \_\_\_\_\_ years Child's education status: 
attend in school 
temporally not in school permanently not in school (causes: \_\_\_\_\_) Age of child when diagnosed with DMD \_\_\_\_\_ years Health condition of the child: 
excellent 
good 
fair 
poor Severity of symptom of the child: 
severe moderate mild

No:

# II. Child's Current Condition: Upper and Lower Extremities Functioning Assessment

Instructions:

Please read each statement carefully, and decide how well it describes your child's upper extremities functioning and lower extremities functioning. You should answer according to how you see your child's current function. Please mark an "v" in the appropriate box "□" about your child's upper and lower extremities functioning.

Upper Extremities Functioning Assessment

- ☐ 1.Starting with arms at the sides, the child can raise and extend the arms in a full circle until they reach above the head
- 2.Can raise arms above head only by flexing the elbow or using accessory muscles
- ☐ 3.Can't raise hands above head, but can raise an 8-oz glass of water to the mouth
- ☐ 4.Can raise hands to the mouth, but can't raise an 8-oz glass of water to the mouth
- 5.Can't raise hands to the mouth, but can use hand to hold a pen or pick up pennies from the table
- 6.Can't raise hands to the mouth, and has no useful function of hands

Lower Extremities Functioning Assessment:

- 1.Walks and climbs stairs without assistance
- 2. Walks and climbs stairs with aid of railing (<12 sec/4 standard steps)
- ☐ 3.Walks independently and climbs stairs slowly with aid of a railing (>12 sec/4 standard steps)
- 4. Walks independently and rises from chair unassisted but cannot climbs stairs
- 5.Walks independently but cannot rise from a chair or climb stairs
- 6.Walks independently in bilateral knee-ankle-foot orthoses
- 7. Walks with orthoses and assistance of one person
- 8. Stands in orthoses but is unable to walk even with assistance
- 9. Is in a wheelchair
- $\square$  10. Is confined to a bed

III. Child's daily activity assessment; The Barthel Index Directions: For each activity, please mark on "v" in the appropriate box "<sup>f</sup>" about your

child's daily activities.

Activity	Score
Feeding 0=unable	
5=needs help cutting spreading butter etc. or requires modified	l diet
10=independent	
Bathing	
0=dependent	
5=independent (or in shower)	<u> </u>
Grooming	
0=needs to help with personal care	
5=independent face/hair/teeth/shaving (implements provided)	
Dressing	
0=dependent	
5=needs help but can do about half unaided	
10=independent (including buttons, zins, laces, etc.)	<del></del>
Bowels	
()=incontinent (or needs to be given enemas)	
5=occasional accident	
10=continent	
Bladder	
0=incontinent, or catheterized and unable to manage alone	
5=occasional accident	
10=continent	
Toilet Use	<u> </u>
0=dependent	
5=needs some help, but can do something alone	
10=independent (on and off, dressing, wiping)	
Transfers (Bed to chair and back)	
0=unable, no sitting balance	
5=major help (one or two people, physical), can sit	
10=minor help (verbal or physical)	
15=independent	
Mobility (on level surfaces)	
0=immobile or $< 50$ yards	
5=wheelchair independent, including corners, > 50 yards	
10=walks with help of one person (verbal or physical) > 50 yards	
15=independent (but may use any aid; for example, stick) > 50 ya	ards
Stairs	
0=unable	
5=needs help (verbal, physical, carrying aid)	
10=independent	
Total (0-100)	):

### Appendix I Duke Health Profile (The DUKE)

Instructions: Here are some questions about your health and feelings. Please read each question carefully and check (v) your best answer. You should answer the questions in your own way. There are no right or wrong answers. (Please ignore the small scoring numbers next each blank.)

· · · · · · · · · · · · · · · · · · ·	Yes, describes	Somewhat describe	No, doesn't describe me
	me	me	at all
	exactly		
1. I like who I am	2	1	0
2. I am not an easy person to get along with	n 0	1	2
3. I am basically a healthy person	2	1	0
4. I give up too easily	0	1	2
5. I have difficulty concentrating	0	1	2
6. I am happy with my family relationships	2	1	0
7. I am comfortable being around people	2	1	0
Today would you have any physical trouble	or difficulty	/?	
	None	Some	A Lot
8. Walking up a flight of stairs	2	1	0
9. Running the length of a football field	2	1	0
During the past week: How much trouble ha	ave you had	with	
	None	Some	A Lot
10. Sleeping	2	1	0
11. Hurting or aching in any part of your bo	dy 2	1	0
12. Getting tired easily	2	1	0
13. Feeling depressed or sad	2	1	0
14. Nervousness	2	1	0
During the past week: How often did you 15. Socialize with other people (talk or visit	None	Some	A Lot
with friends or relatives)	0	1	2
16. Take part in social, religious, or recreati activities (meetings, church, movies,	on		
sports, parties)	0	1	2
During the past week: How often did you 17. Stay in your home, a nursing home, or h because of sickness, injury, or other hea	None Iospital lth	1-4 Days	5-7 Days
problem	2	1	0

## MANUAL SCORING FOR THE DUKE HEALTH PROFILE

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\* <u>Raw Score</u> ≈ last digit of the numeral adjacent to the blank checked by the respondent for each item. For example, if the second blank is checked for item 10 (blank numeral = 101), then the raw score is "1", because 1 is the last digit of 101.

<u>Final Score</u> is calculated from the raw scores as shown and entered into the box for each scale. For physical health, mental health, social health, general health, self-esteem, and perceived health, 100 indicates the best health status, and 0 indicates the worst health status. For anxiety, depression, anxiety-depression, pain, and disability, 100 indicates the worst health status and 0 indicates the best health status.

Missing Values: If one or more responses is missing within one of the eleven scales, a score cannot be calculated for that particular scale.

#### Appendix J

#### Family APGAR Scale

The following questions have been designed to help us better understand you and your family. You should feel free to ask questions about any item in the questionnaire. Please try to answer all questions

Family is the individual(s) with whom you usually live. If you live alone, consider family as those with whom you now have the strongest emotional ties.

For each question, check only one box

	Almost always	Some of the time	Never
I am satisfied that I can turn to my family for help when something is troubling me	2 🗌	1	0 🗌
I am satisfied with the way my family talks over things with me and shares problems with me	2 🗌	1	0 🗌
I am satisfied that my family accepts and supports my wishes to take on new activities or directions	2 🗌	1 🗌	0 🗌
I am satisfied with the way my family expresses affection and responds to my emotion, such as anger, sorrow, or love	2 🗌	1	0 🗌
I am satisfied with the way my family and I share time together	2 🗌	1	0 🗌

#### Appendix K

#### Family Hardiness Index

Directions:

Please read each statement below and decide to what degree each describes your family. Is the statement false (0), mostly false (1), mostly true (2), or true (3) about your family? Circle a number 0 to 3 to match your feelings about each statement. Please respond to each and every statement.

					_
In our family	False	Mostly False	Mostly True	True	-
1. Trouble results from mistakes we make	0	1	2	3	R
2. It is not wise to plan ahead and hope because things do not turn out anyway	0	1	2	3	®
3. Our work and effort are not appreciated no matter how hard we try and work	0	1	2	3	R
4. In the long run, the bad things that happen to us are balanced by the good things that happen	0	1	2	3	
5.We have a sense of being strong even when we face big problems	0	1	2	3	
6.Many times I feel I can trust that even in difficult times things will work out	0	1	2	3	
7. While we don't always agree, we can count on each other to stand by us in times of need	0	1	2	3	
8. We do not feel we can survive if another problem hits us	0	1	2	3	R
9. We believe that things will work out for the better if we work together as a family	0	1	2	3	
10. Life seems dull and meaningless	0	1	2	3	R
11.We strive together and help each other no matter what	0	1	2	3	1
12. When our family plans activities we try new and exciting things	0	1	2	3	
13. We listen to each others' problems, hurts and fears	0	1	2	3	1
14. We tend to do the same things over and over it's boring	0	1	2	3	R
15. We seem to encourage each other to try new things and experiences	0	1	2	3	
16. It is better to stay at home than go out and do things with others	0	1	2	3	R
17. Being active and learning new things are encouraged	0	1	2	3	1
18. We work together to solve problems	0	1	2	3	
19. Most of the unhappy things that happen are due to bad luck	0	1	2	3	ß
20. We realize our lives are controlled by accidents and luck	0	1	2	3	R
The ® symbol is for computer use only	-	· · · · · · · · · · · ·	Total		-

#### Appendix L

#### FAMILY ASSESSMENT DEVICE Brown University/Rhode Island Hospital Family Research Program

#### **INSTRUCTIONS:**

This booklet contains a number of statements about families. Please read each statement carefully, and decide how well it describes your own family. You should answer according to how you see your family.

For each statement there are four (4) possible responses:

Strongly Agree (SA)	Check SA if you feel that the statement describes your family very accurately.
Agree (A)	Check A if you feel that the statement describes your family for the most part.
Disagree (D)	Check D if you feel that the statement does not describe your family for the most part.
Strongly Disagree (SD)	Check SD if you feel that the statement does not describe you family at all.

These four responses will appear below each statement like this:

41. We are not satisfied with anything short of perfection.

\_\_\_\_\_ SA \_\_\_\_ A \_\_\_\_ D \_\_\_\_ SD

The answer spaces for statement 41 would look like this. For each statement in the booklet, there is an answer space below. Do not pay attention to the blanks at the far right-hand side of each space. They are for office use only.

Try not to spend too much time thinking about each statement, but respond as quickly and as honestly as you can. If you have trouble with one, answer with your first reaction. Please be sure to answer <u>every</u> statement and mark all your answers in the <u>space provided below</u> each statement.

FAMILY ASSESSMENT DEVICE	ID
Brown University/Rhode Island Hospital Family Research Program	Page 1
Planning family activities is difficult because we misunderstand eac	sh other
SA A D SD	
2. We resolve most everyday problems around the house.	
SAADSD	·
3. When someone is upset the others know why.	
SAADSD	
4. When you ask comeans to do compating you have to shack that the	
4. When you ask someone to do something, you have to check that they	
5. If someone is in trouble, the others become too involved.	
SA A D SD	
6. In times of crisis we can turn to each other for support.	
SAADSD	<u></u>
7. We don't know what to do when an emergency comes up.	
SAADSD	
$\mathbf{x}_{\mathbf{x}}$ . We competimes run out of things that we need	
SA A D SD	
9. We are reluctant to show our affection for each other.	
SAADSD	
10. We make sure members meet their family responsibilities.	
SAADSD	
11. We cannot talk to each other shout the sedners we feel	
11. We cannot tark to each other about the sadness we reel.	

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FAMILY ASSE	SSMENT DEVICE				ID	
<b>Brown Universit</b>	ty/Rhode Island Hos	pital Family Re	search Program			Page 2
12.	We usually act on	our decisions r	egarding problem	15.		
	SA	A	D	SD		
13.	You only get the in	nterest of others	when something	s is important to them	).	
	SA	A	D	SD	-	
14.	You can't tell how	a person is fee	ling from what th	ney are saying.		
	SA	A	D	SD	-	
15.	Family tasks don't	get spread aro	und enough.			
	SA	A	D	SD	-	
16.	Individuals are acc	epted for what	they are.			
	SA	A	D	SD	-	
17.	You can easily get	away with brea	aking the rules.			
	SA	A	D	SD	-	
18.	People come right	out and say thi	ngs instead of hir	ting at them.		
	SA	A	D	SD	-	<u> </u>
19.	Some of us just do	n't respond em	otionally.			
	SA	A	D	SD	-	
20.	We know what to a	do in an emerge	лсу.			
	SA	A	D	SD		
21.	We avoid discussi	ng our fears and	concerns.			
	SA	A	D	SD	-	
27	It is difficult to tal	k to each other	about tender feel	inos		
£2.	SA	A	D	SD		
					-	

versi	ly/Rhode Island Hospital Family Research Program				Page
					-
23.	We have trouble m	ecting our bills.			
	SA	A	D	SD	
24.	After our family tri	es to solve a probl	em, we usually d	liscuss whether it worl	ked or not.
	SA	A	D	SD	
25	We are too self-cen	tered.			
23.	SA	A	D	SĎ	
26	W	timen to cook other			
26.	SA	A	D	SD	
27.	We have no clear e	A	D	SD	
28.	We do not show ou SA	r love for each oth A	er. D	SD	
29.	We talk to people d	lirectly rather than A	through go-betw D	reens. SD	
				02	<u> </u>
30.	Each of us has part	icular duties and r	esponsibilities.	SD	
	SA	A	D	5D	<u></u>
31.	There are lots of ba	d feelings in the fa	mily.		
	SA	A	D	SD	
32.	We have rules abou	t hitting people.			
	SA	A	D	SD	
22	We get involved wi	th each other only	when something	interest us.	
55.					

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vn Ur	niversity/Rhode Is	sland Hospital Fa	mily Research Pı	ogram	Pa
34.	There's little tin	ne to explore per	sonal interests.		
	SA	A	D	SD	
26	WI	1	_		
33.	SA	say what we mea	n. D	SD	
36	We feel accente	d for what we are	*		
50.	SA	A	D	SD	
37.	We show interest	st in each other w	hen we can get s	omething out of it person	nally.
	SA	A	D	SD	
38.	We resolve mos	t emotional upset	s that come up.		
	SA	A	D	SD	
39.	Tenderness take	s second place to	other things in o	ur family.	
	SA	A	D	SD	
40.	We discuss who	is to do househo	ld jobs.		
	SA	A	D	SD	
41.	Making decision	is is a problem fo	r our family.		
	SA	A	D	SD	
42.	Our family sho	ws interest in eac	h other only whe	n they can get something	gout of it.
	SA	A	D	SD	
43.	We are frank wi	ith each other.			
	SA	A	D	SD	
44.	We don't hold to	o any rules or sta	ndards.		
	SA	A	D	SD	

<b>V</b> 25	ESSMENT DEVICE				
niver	sity/Khode Island Ho	spital ramity Res	earch Frogram		rag
45.	If people are asked	to do something, t	they need remindi	ng.	
	SA	A	D	SD	
46.	We are able to mak	e decisions about	how to solve pro	blems.	
	SA	A	D	SD	
		1 24 1			
47.	If the rules are brok	en, we don't know	w what to expect.	SD.	
	5A	A	D	5D	
48.	Anything goes in ou	or family.			
	SA	A	D	SD	
49.	We express tendern	ess.			
	SA	A	D	SD	
50.	We confront proble	ms involving feeli	ngs.		
• • • •	SA	A	D	SD	<u> </u>
51	We don't get along	well together.			
	SA	A	D	SD	
БÒ	157- Jan 24 4-11-4-	-h athan			
52.	we don't talk to ead	A chi ouner when we	are angry.	SD	
	SA	A	U	50	
53.	We are generally di	ssatisfied with the	family duties as	signed to us.	
	SA	A	D	SD	
54.	Even though we me	an well, we intruc	le too much into o	each others lives.	
	SA	A	D	SD	
	There are rules abo	ut dangerous situ:	ations.		
55.					

FAMILY A Brown Uni	ASSESSMENT DEVIC versity/Rhode Island I	ID Page 6			
56.	We confide in each other.				
	SA	A	D	SD	
57.	We cry openly.				
	SA	A	D	SD	
58.	We don't have reaso	mable transport.			
	SA	A	D	SD	· · · · · · · · · · · · · · · · · · ·
59.	When we don't like what someone has done, we tell them.				
	SA	A	D	SD	
60.	We try to think of different ways to solve problems.				
	SA	A	D	SD	

#### Appendix M Information Sheet for the Subjects

Researcher

Jih-Yuan Chen, PhD. Candidate, MSN, RN. Doctoral Student Hahn School of Nursing and Health Science University of San Diego (858) 278-4201 jihc@sandiego.edu Faculty Advisor Susan L. Instone, DNSc, RN, CPNP Hahn School of Nursing and Health Science University of San Diego 5998 Alcala Park San Diego, Ca 92110 (619) 260-4549 sinstone@sandiego.edu

#### Purpose and Explanation

Jih-Yuan Chen, a doctoral candidate at the University of San Diego, is doing a research study to learn more about how Taiwanese families with a child having Duchenne muscular dystrophy (DMD) function. As a parent of a child with DMD, you are being asked to participate. Ms. Chen will collect and analyze the data in this study. Dr. Instone serves as the faculty advisor. Ms. Chen may consult with Dr. Instone and the other members of her dissertation committee from the University of San Diego regarding data analysis.

#### Procedures

If you agree to participate in the study, the following will occur

- 1. You will be called by Ms. Chen and asked to participate voluntarily in the study.
- 2. You will be given a chance to ask questions about this research study before you are asked to sign the consent form.
- 3. You will be sent the questionnaires by mail. These include the Demographic Sheet (including family and medical characteristics, child's upper and lower extremities functioning, and child's daily activity), the Duke Health Profile-17 items, Family APGAR Scale-5 items, Family Hardiness Index-20 items, and Family Assessment Device-60 items. It will take about one hour to complete these questionnaires.
- 4. Please answer the questions within 2 weeks at a convenient time. The researcher will be available by phone or face to face if you need help answering the questions. If for any reason you do not wish to answer the question, you may stop at any time.
- 5. Your participation is voluntary and may be terminated at any time for any reason.
- 6. Any information shared with Ms Chen will not be shared with any others. All information will be confidential.

#### **Risks/Discomforts**

You have been informed that participation in this study may involve few emotional risks. If counseling or support is needed, you may call social worker Miss Hui-Fong Wang 07-380-1000 at the Taiwan Muscular Dystrophy Association.

#### Benefits

You will receive no benefit from participating in this study. Ms Chen may achieve a better understanding of what factors influence family functioning in families with a child having a DMD. In addition, the study may contribute knowledge to the development of intervention that will help to promote better functioning.

#### Appendix N

#### Consent Form

Project: Functioning Among Taiwanese Families with a Child Having Duchenne Muscular Dystrophy

Researcher: Jih-Yuan Chen, RN, MSN

Ms. Chen is an Associate Professor at the College of Nursing of Kaohsiung Medical University, and a doctoral student conducting research for a dissertation at the University of San Diego, USA.

The purpose of the study is to investigate the factors associated with functioning among families with a child having Duchenne Muscular Dystrophy. If you agree to be in the study, you will complete four written questionnaires: Family Hardiness Index (FHI), Family APGAR, Family Assessment Device (FAD), Duke Health Profile (Duke), and Demographic sheet. These questionnaires are expected to take one hour to complete.

You will be free to telephone the researcher: Ms. Jih-Yuan Chen 07-3233778 (Taiwan) or 002-1-858-278-4201 (USA), and Dr. Susan Instone 002-1-619-260-4549 (USA) with any questions you may have. Your name will not appear on any of these questionnaires. Furthermore, all information provided in the questionnaires will be treated in a confidential manner. All data will be locked in a file cabinet with access only by the investigator. All data will be destroyed in five years.

This study will not provide any direct benefits to you, but the results of the study may influence the quality of life of others families in the future. You have been informed that participation in this study may involve few emotional risks. If counseling or support is needed, you may call social worker Miss Hui-Fong Wang 07-380-1000 at the Taiwan Muscular Dystrophy Association. You may decide to withdraw at any time, and your child's medical care will not be affected in any way if you decide to withdraw.

Completing the questionnaires will take about one hour. I have been given the opportunity to ask whatever questions I desire.

Signature of Participant

Date

Location

Signature of Researcher

Date