To the Graduate Council:

I am submitting herewith a dissertation written by A. Graham Parker entitled "An Analysis of the Coping Strategies Used by Males With Duchenne Muscular Dystrophy." I have examined the final copy of this dissertation for form and content and recommend that it be accepted in partial fulfillment of the requirements for the degree of Doctor of Philosophy with a major in Education.

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An Analysis of the Coping Strategies Used by Males With Duchenne Muscular Dystrophy

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A. Graham Parker

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DEDICATION

This dissertation is
dedicated to

John

for all the years
of love and support.

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ABSTRACT

The perceived stressors and coping strategies of 15 males ages 9 through 19 with Duchenne Muscular Dystrophy (DMD) were investigated. Through a semi-structured interview process, subjects were asked to identify the stressful situations they encounter. These stressful situations were categorized semantically by independent raters.

Subjects were also asked to rate the severity and frequency of each perceived stressor on a 5-point Likert-type scale. Finally, subjects were asked to report the coping strategies utilized to deal with the stressful situations. Coping strategies were categorized in terms of active or avoidant styles.

Subjects were divided into three groups for comparison: 9 to 12 year olds, 13 to 15 year olds, and 16 to 19 year olds. Five variables were investigated to determine the relationships between age of subject, type of stressor, severity of stressor, frequency of stressor, and coping strategy.

Results indicated that this sample of males with DMD identified six primary sources of stress related to their disease. Younger subjects were more concerned with the physical implications of their disease, while older subjects voiced greater concern about psychosocial issues. Specifically, results supported the research question

that older subjects would be more concerned than younger subjects with the implications of their disease for a shortened life span. No substantial age differences were found in terms of types of coping strategies reported. Overall, however, active coping strategies were reported more than avoidant strategies. In addition, there was evidence of the use cognitive approaches to coping in all three age groups. Within the total sample, a moderate correlation was noted between severity and frequency of a stressor.

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CHAPTER 1

INTRODUCTION

Approximately twenty years ago Mattsson (1977) defined a chronic illness as "a disorder with a protracted course which can be progressive and fatal, or associated with a relatively normal life span despite impaired physical or mental functioning" (p. 183). Mattsson reported that 30% to 40% of children up to age 18 have one or more long-term disorder. This figure, in keeping with his broad definition, includes children with sensory impairment, mental retardation, speech disorders, learning disorders, and behavior problems. More recently, Midence (1994) states that between 10% and 20% of children suffer from chronic disease. The author uses the terms "chronic disease" and "chronic illness" interchangeably. They are operationally defined as "conditions that extend beyond three months, often for life, and cannot be cured" (p. 311). This narrower definition provided by Midence refers more specifically to children with medical conditions. Examples of chronic medical conditions include asthma, hemophilia, sickle cell anemia, epilepsy, cystic fibrosis, and muscular dystrophy.

Compared with "healthy" children, children with chronic illness, particularly those who manifest a physical disability, are perceived to be at greater risk for psychosocial adjustment difficulties (Cadman, Boyle,

Szatmari, and Offord, 1987; Eiser, 1990; Stein & Jessop, 1984). Presumably, the presence of a chronic illness, with or without accompanying physical disability is stressful.

Definition of Terms

Matheny, Aycock, and McCarthy (1993) use the terms "stress" and "stress response" interchangeably. The authors emphasize the physiological and psychological adjustments made by the individual in the face of demanding events, situations, or thoughts. Negative physical and emotional outcomes represent the symptoms of experiencing the stress response. According to Eiser (1990) "stress" may be viewed as the occurrence of a problem situation requiring some decision-making process for appropriate action. For children with chronic illness, such problem situations include frequent hospitalizations, invasive medical procedures, uncomfortable daily regimens, and restriction of physical activity. Cohen and Lazarus (1979) suggest that one's cognitive appraisal of the perceived stressful encounter plays a central role in adaptation and management of the impact of stressful events. By "appraisal" the authors refer to the evaluation of the significance of an event for one's well-being (primary appraisal), and the evaluation of one's coping resources and options (secondary appraisal).

A child's developmental level determines, in part, the types of cognitive appraisals made. Preschool children, for

instance, are more likely to view disease as "punishment" for misbehavior (Schowalter, 1970). The parent takes most of the responsibility of coping for the young pre-logical child. The physical presence of and reassurance by the caregiver is most crucial at this age (Natterson and Knudson, 1960). The universality and permanence of death are comprehensible to the preadolescent child, however (Schowalter, 1970). Older children and adolescents, in particular, are able to understand the idea of disease and body malfunction. Moreover, they are able to exert a greater amount of personal control over their situation (Bibace and Walsh, as cited in LaMontagne, 1987). Thus, effective adaptation to stress, or "coping", appears to increase with age.

Cohen and Lazarus (1979) define coping as "efforts, both action-oriented and intrapsychic, to manage environmental and internal demands, and conflicts among them, which tax or exceed a person's resources" (p. 219). In this framework, coping appears to be a process rather than an outcome. It can be further characterized as both "cognitive and behavioral efforts made by individuals to master, tolerate, or reduce" (Folkman and Lazarus, p.223, as cited in Curry and Russ, 1985) stressful demands. Coping appears to be a constellation of interpersonal and cognitive problem-solving acts (Cohen and Lazarus, 1979). Definitions of stress, appraisal, and coping have originated in the

adult literature and have been subsequently applied to the pediatric literature (Compas, 1987). Similarly, the conceptualization of issues related to psychosocial adjustment of chronically ill individuals has proceeded from adult to pediatric models. Given the prevalence of children with chronic disease, and the presumed stress encountered by the affected child, it is important to identify the strategies used by chronically ill children to cope with their disease.

Review of the Literature

Theories and Models of Children's Coping

Rutter (1983) identifies a number of individual differences that influence the child's ability to cope with stress. Age, for example, is identified as important, with older children, in general, more able to employ problem-solving strategies. Sex differences are noted, with prepubertal boys described as "more vulnerable" (p. 19) to stress events than their female peers. Intellectual ability tends to mediate the stress response, with the "tendency for children of above average intelligence to have rather low rates of psychiatric disorder" (p. 22). One could argue that deficits in effective coping strategies are related to psychiatric difficulties such as depression and anxiety.

Active Versus Avoidant Coping Styles

One conceptualization of coping styles found in both the adult and pediatric literature is that of "active"

versus "avoidant". Curry and Russ (1985) define active coping as the seeking of information in the face of stress which is intended to mediate the stress response. Peterson (1989) defines active coping as a willingness to seek information relevant to the stressor, and initiation of some strategy, such as relaxation or imagery, to exercise control during a stressful event. Avoidant coping, on the other hand, is defined as attempts to deny the presence of a stressor or as attempts to distract oneself from a stressor (Suls and Fletcher, 1985).

Among "healthy" children and adolescents (ages 5 through 18) a number of studies suggest that the use of active coping strategies for social and academic stressors is associated with more desirable outcomes than is the use of avoidant strategies (Peterson, 1989). Overall, the author notes that, among healthy children, active coping increases with age. The use of avoidant strategies, in contrast, has been associated with increased levels of self-reported anxiety in youngsters of the same age (Peterson, 1989). Thus, it may be that chronically ill children utilize more avoidant strategies, while chronically ill adolescents utilize more active strategies.

Suls and Fletcher (1985) completed a meta-analysis of the efficacy of various coping strategies. The investigators reviewed over forty studies with adult subjects. Specifically, they investigated under what

conditions "avoidant" and "nonavoidant" styles appeared effective in reducing such conditions as pain, stress, and anxiety. The authors identified as avoidant those behaviors or cognitions that reflected denial, distraction, or repression. Nonavoidant responses to stress were characterized by vigilance, attention, and monitoring of sensory input. They further differentiated whether a specific strategy produced desirable results for the short-term versus the long-term. Results of the investigation suggested no strong overall effects favoring the use of either avoidant or nonavoidant strategies in stressful situations. However, avoidant strategies appeared to be more useful in the short-term. It was suggested that the avoidant style may be functional in the short-term until the person is capable of applying problem-focused strategies to cope with an unpleasant reality (p. 252). Given these results, it may be that children with chronic illness initially utilize an avoidant strategy in coping with their condition, and later employ active strategies as they are forced to cope with an inevitable outcome. Unfortunately, no such meta-analysis exists in the children's coping literature.

Emotion-Focused Versus Problem-Focused Coping Styles

In addition to the active versus avoidant coping dimension, the emotion-focused versus problem-focused dimension is also described in the literature. Emotion-

focused coping strategies are defined as efforts by the individual to alter their appraisal of threatening conditions, and, thereby reduce their emotional distress. Problem-focused coping strategies are defined as efforts to directly alter the stress situation itself. (Rutter, 1983). There is some indication that the use of problem-focused coping strategies yields more desirable outcomes than emotion-focused strategies (Compas et al., 1988).

Compas (1987) reviewed the pediatric literature on coping and identified seven major lines of research: (1) attachment and separation during infancy; (2) social support; (3) interpersonal cognitive problem solving; (4) coping in achievement contexts; (5) Type A and B behavior patterns; (6) styles of repression and sensitization, and (7) resilience or invulnerability to stress. Compas concurs with Cohen and Lazarus (1979) and Folkman and Lazarus, as cited by Curry and Russ (1985), that coping is an effortful response to stress. During childhood, coping is affected by both personal and environmental factors. Compas suggests that "both problem-focused and emotion-focused coping are important in successful adaptation to stress" (p. 399). The use of a particular strategy depends upon the situation, and "effective coping is likely to be characterized by flexibility and change" (p. 399). Compas advocates the development of appropriate coping measures suitable for children and adolescents, as well as longitudinal studies to

clarify whether, and to what degree, coping styles change with the individual's cognitive, social, and emotional development.

Compas, Malcarne, and Fondacaro (1988) reported that the investigation of the relationship between cognitive appraisals and coping was lacking in the pediatric coping literature. They investigated 130 subjects, ages 10 to 14, with regard to two types of stressors. Each youngster described one stressful interpersonal and one stressful academic event, and then rated the perceived amount of control over the event on a Likert-type scale (1= complete control; 5= no control). The subjects generated a list of all the possible ways in which the stressful events could have been handled, and then identified the strategy they actually used. Responses were classified as either problem-focused or emotion-focused by the investigators. Emotional and behavioral problems were assessed with the Child Behavior Checklist (CBC). Subject's mothers completed the parent version, while the subjects completed the Youth Self-Report. Results of the multivariate analysis (MANOVA) indicated that students who were less adept at generating problem-focused solutions experienced more adjustment problems, per the CBC. The girls in this study used more emotion-focused coping strategies than the boys with regard to academic coping. The generation and use of emotion-focused strategies increased with age of subject, while the

generation and use of problem-focused coping remained consistent. With regard to appraisals, subjects perceived the causes of academic stressors as more controllable than the causes of social stressors. Subjects also generated more problem-focused alternatives for coping with academic stress.

Coping With Acute Medical Situations

In the early 1970s Cohen and Lazarus (1973) assessed the role of individual coping processes in the mediation of outcomes from surgical procedures. While this study did not utilize children as subjects, the authors explored the idea of coping dispositions in the face of acute health-related stress. Surgical patients were classified along the "avoidance" versus "vigilance" dimension utilizing the coper-avoider Sentence Completion Test (SCT) and the modified Repression-Sensitization Scale (R-S). The subjects' self-reported level of anxiety was identified on a 1 to 10 scale prior to the surgical procedure as well. Twenty-three percent of the subjects were identified as "avoiders", 16% as "vigilant", and 61% as reflecting a mix of both coping styles. Outcomes were measured with a standard Recovery Index, which identifies number of days in the hospital post-surgery, number of minor post-surgical complications, number of pain medications requested, and number of negative psychological reactions. Results indicated that the avoidant and mixed groups recovered

somewhat faster than the vigilant patients. The authors concluded that an avoidant orientation may be an effective mode of coping in a medical situation in which the likelihood or threat of death is small.

The stress experienced and coping strategies implemented by children undergoing acute medical procedures has also been investigated. Curry and Russ (1985) evaluated 48 children, ages 8 to 10, undergoing dental procedures. Subjects were observed during treatment and interviewed following treatment with regard to their coping efforts. Nine functional cognitive and behavioral classifications of coping strategies were derived: (1) information-seeking; (2) support-seeking; (3) direct efforts to maintain control; (4) reality-oriented working through; (5) positive cognitive restructuring; (6) defensive reappraisal; (7) behavior-regulating coping cognitions; (8) emotion-regulating coping cognitions; and (9) diversionary thinking. These nine categories include strategies reflective of both the active versus avoidant and emotion-focused versus problem-focused dichotomies. Every child in the sample used at least two cognitive and one behavioral coping response. The average child utilized a variety of strategies; however, older children were noted to use more cognitive versus behavioral strategies.

Zastowny, Kirschenbaum, and Meng (1986) reported the importance of coping skills training in the adaptation to

stressors, including hospitalization for surgery. They assessed children ages 6 to 10 preparing for surgery. Thirty-three parent-child dyads were divided into three groups; (1) information only; (2) information and anxiety reduction training; and (3) information, anxiety reduction training, and coping skills intervention. Coping skills training was seen as advantageous for assisting parents in modeling coping behaviors, increasing reinforcement of the child for relaxed responding, and increasing the child's perception of control. The investigators assessed the child's behavior before, during, and following surgery via behavioral diaries, rating scales, and questionnaires. Results of the training were positive. The coping skills intervention (group 3) helped children adapt to hospitalization better than anxiety reduction (group 2) and information only (group 1) interventions. Only the coping skills group exhibited fewer problem behaviors than the information group both before and after hospitalization.

Peterson (1989) summarized eight studies which evaluated the coping responses of children undergoing acute medical procedures. Observations, interviews, rating scales, and physiological measures were utilized to collect behavioral and cognitive data. Studies classified coping responses along the dimensions of active versus avoidant, internal versus external locus of control, and emotion-focused versus problem-focused. Results of the overview

included: less defensive children played with medically relevant toys and had lower levels of self-reported anxiety following hospitalization (Knight, et al., as cited in Peterson, 1989); children with active coping styles were more internal in their locus of control (LaMontagne, as cited in Peterson, 1989); and active coping, as evidenced by seeking information, decreased child distress prior to surgery (Peterson and Toler, as cited in Peterson, 1989).

Coping With Chronic Illness

With regard to chronic illness and treatment, Cohen and Lazarus (1979) identify six "threats" with which the chronically ill individual must cope: (1) to life and fear of dying; (2) to bodily integrity; (3) to self-concept and future plans; (4) to emotional equilibrium; (5) to the fulfillment of customary social roles and activities; and (6) to the need to adjust to a new physical or social environment (p. 229). In addition, Cohen and Lazarus (1979) identify five "adaptive tasks" of the chronically ill individual: (1) reduction of harmful environmental conditions; (2) tolerance or adjustment to negative events and realities; (3) maintenance of a positive self-image; (4) maintenance of emotional equilibrium; and (5) continuation of satisfying relationships with others (p. 232). According to Mattsson (1977), the appropriate release and control of emotions is essential for a child's coping: "The expression of anxious, sad, impatient, and angry feelings at times of

exacerbations, and of confidence and guarded optimism during periods of clinical quiescence, is characteristic of well-adapted children with chronic illness" (p. 191). Three other factors determine adaptation to chronic illness, according to Moos and Tsu (1977): (1) background and personal characteristics, such as age, cognitive development, and previous coping experiences; (2) illness-related factors, such as type of symptoms, rate of onset, and general disease progression; and (3) features of the general physical and social environment, including quality of sensory stimulation and relationships with significant others.

Early Findings

Natterson and Knudson (1960) investigated the stress responses of 33 hospitalized children diagnosed with cancer, leukemia, and various blood diseases. The investigators utilized a number of hospital personnel, including physicians, nurses, and occupational therapists, to observe pediatric inpatients during hospital activities. Activities observed were not limited to medical procedures, per se, but also included meal times and free play. Children under age five were noted to have the most severe reaction when separated from their mothers. Children ages 5 to 10 reacted most strongly to medical procedures, while children ages 6 to 12 experienced the most severe distress following the death of another child on the unit. The authors concluded

that death awareness in their chronically ill population was age-related. Unfortunately, the authors did not use a standardized measure of their construct of "stress". Nor did they utilize a standardized behavioral observation or rating system to assess children's reactions. Staff observations were recorded anecdotally, with no procedure to insure inter-rater reliability.

Pless and Roghmann (1971) summarized the findings of three epidemiological surveys relating to the psychological and social consequences of chronic physical disorders. The first, the National Survey of Health and Development, followed more than 5,000 children born in England, Wales, and Scotland during the first week of March, 1946. Respiratory dysfunction was rated as the most prevalent disorder. The Isle of Wight, England study represented the second survey. The educational, emotional, and physical disabilities of over 3,000 children, ages 9 to 11, were assessed. Children with neurological problems were most prevalent, with respiratory disease a close second. The third study described was the Rochester Child Health Survey, which consisted of a one percent probability sample of all children under 18 years of age living in Monroe County, New York. Again, respiratory disease represented the most frequently occurring medical condition. In all three studies, "ill" children were compared to a comparable sample of "healthy" children.

With regard to educational achievement, reading skills were specifically targeted. The data suggested that reading underachievement was related to disease severity. Studies differ, however, in terms of suspected causality. The Isle of Wight study showed no significant difference between number of school absences in ill versus healthy children. However, according to the National Survey, ill children were absent from school significantly more often than healthy controls.

Behavioral and psychological maladjustment were assessed in all three studies via rating scales and behavioral symptom questionnaires. Parents, teachers, and the children themselves served as informants/raters. The data suggested that "chronically ill children are, compared with healthy children, more frequently truant, more often troublesome in school, and more often socially isolated" (p. 355). The Isle of Wight study showed that the rate of psychiatric disorders was 17% in chronically ill children, as compared to 7% of healthy children. According to the National Survey, "the frequency of emotional symptoms is seen to be consistently related to the duration of the disorder" (p. 356). Overall, the authors concluded that approximately 30% of chronically ill children "may be expected to be handicapped by secondary social and psychologic maladjustments" (p. 357).

In a more recent epidemiological study (Cadman et al., 1987) over 3,000 children and adolescents were evaluated with the Survey Diagnostic Instrument (SDI). This instrument incorporates items from the Child Behavior Checklist (CBC). Parent, teacher, and subject self-report versions were utilized. Subjects fell into one of three categories: (1) physically healthy, (2) chronically ill without a physical disability, and (3) chronically ill with one or more physical disability. Results of the SDI were used to classify children as psychiatrically disordered, per the Diagnostic and Statistical Manual of Mental Disorders, third edition (DSM-III). Results of the SDI indicated that chronically ill children without physical disability had lower risk of psychiatric disorder than ill children with a physical disability. Isolation was more typical of children with both a chronic illness and a physical disability. Both chronically ill children with and without disability were at higher risk for psychiatric problems than healthy control subjects. Diagnoses of "neurosis" and "attention deficit/hyperactivity disorder" were the most prevalent disorders in children with combined illness and physical disability. Thus, the Cadman et al. study is in keeping with the three earlier epidemiological studies, identifying chronically ill children, particularly those with a physical disability, as more at risk for psychosocial difficulties.

Zelter, L., Kellerman, J., Ellenberg, L. & Rigler, D. (1980) compared 345 healthy adolescents to 168 adolescents with chronic illness, including diabetes, cystic fibrosis, cancer, and cardiac disease. Subjects in the "healthy" group did include adolescents with mild, acute health concerns, such as sinus infections, colds, and minor surgery. Utilizing an author-designed Illness Impact Questionnaire (IIQ), all subjects rated their functioning in a number of areas along a four point Likert-type scale. Areas assessed included relationships within the family, school and peer activities, independence, future orientation, and effects of treatments. Total IIQ scores did not differ significantly between healthy and chronically ill adolescents, on the whole. However, significant differences were noted when specific "illness" groups were compared with one another. For example, cancer and rheumatologic patients expressed greater total impact of disease than did cardiac and diabetic patients. Chronically ill adolescents reported greater school and treatment-related disruption than healthy adolescents who underwent minor, acute medical intervention. Both healthy and ill adolescents were similar in their positive outlook on the future. Results of this study suggest chronically ill children may cope adequately with their disease and should not necessarily be characterized as "disturbed" or "maladjusted".

Emergence of Standardized Measurements

Beginning in the mid-1980s, the literature on children's coping with chronic disease is characterized by utilization of more standardized measurement instruments. Stein and Jessop (1984) assessed the psychological adjustment of 81 chronically ill children with the Personal Adjustment and Role Skills Scale-II (PARS-II). Subjects, ages five and older, were diagnosed with a number of chronic conditions, including respiratory disease (49%), central nervous system disorders (28%), and endocrine disorders (16%). Severity of illness was measured by a standardized morbidity measure, (ex., number of days in bed in the past 2 weeks), as well as by the Functional Status Measure. The latter instrument assesses the child's developmental capacity to perform a variety of activities in the domains of communication, mobility, eating, etc. Bivariate correlations were used to assess the relationships between psychological adjustment and each morbidity measure. Results indicated that only the number of days absent from school within the past 2 weeks was correlated with poor psychological adjustment. In general, higher functional status scores were related to higher psychological adjustment scores on the PARS-II.

Two hundred fifty-five chronically ill/disabled children and adolescents, ages 6 to 18, were studied to evaluate the constancy of behavior problems (Breslau and

Marshall, 1985). Subjects were diagnosed with various disorders, including cystic fibrosis, cerebral palsy, and spina bifida. Mothers of affected subjects completed the Psychiatric Screening Inventory (PSI) in 1978 (T1) and again 5 years later. This instrument contains seven subscales, including self-destructive tendencies, mentation problems, conflicts with parents, and isolation. Stability of scores over time was assessed with Cronbach's Alpha, a measure of internal consistency reliability. Moderate stability of behaviors was found for four subscales- mentation problems, conflict with parents, regressive-anxiety, and isolation. Low stability over time was found for the remaining three subscales- self-destructive tendencies, fighting, and delinquency. Chronically ill/disabled children showed statistically significant improvement on the regressive-anxiety subscale as they grew older.

Coping Strategies

Only recently are researchers beginning to identify the coping strategies used by chronically ill children. Spirito, Stark, and Tye (1994) compared chronically ill and acutely ill children with regard to their use of active versus avoidant coping strategies. Subjects in both groups (chronics vs. acutes) ranged in age from 7 to 17. Using a retrospective procedure, subjects were asked to describe a distressing situation encountered in the hospital. They

categorized their feelings (e.g.,anxious, angry, sad) regarding the situation, and further assessed the degree of emotional response on a 5 point Likert-type scale. Three distinct groups of stressors were classified by the authors: (1) specific aspects, such as bad food, (2) illness-related concerns, and (3) pain-related concerns. Selection of problems differed with age. Younger children appeared more distressed by pain, while adolescents identified problems related to their diagnosis as more distressing. Results indicated that acutely ill or injured children were more likely than chronically ill children to use avoidant coping strategies. Acutely injured adolescents were more likely than chronics to use self-blame strategies. Adolescents were more likely to use cognitive strategies than were younger children.

Mediating Factors

Mediating factors, such as severity of illness and family functioning have also only recently begun to be assessed by researchers. Eiser (1990) notes that the "deficit model" of chronic illness is being slowly replaced with the conceptualization that ill children and their families are "ordinary people in exceptional circumstances" (p. 85). With regard to an illness's effect on a child, Eiser notes that the types of mental health problems prevalent in any specific disorder remain largely unknown. Midence (1994) concurs with Eiser regarding the need for

further investigation of the interaction of intrapersonal, interpersonal, and social-ecological factors as mediators in the coping response.

Coping With Duchenne Muscular Dystrophy

Within the broad topic of children's coping with chronic disease, the population of boys with Duchenne Muscular Dystrophy (DMD) has been largely ignored. DMD is one of many muscular dystrophies. Emery (1994) defines muscular dystrophies as a "group of genetic disorders with progressive muscle wasting and weakness where the disease primarily involves muscle in which there are characteristic microscopic changes" (p. 21).

Duchenne Muscular Dystrophy is the most common form of dystrophy. It is named for the French neurologist who is credited with first describing the disease in 1861 (Emery, 1994). DMD is inherited as an X-linked recessive gene. Thus, the mother is the carrier, though she typically manifests no sign of the disease herself (Madorsky, Radford, and Neumann, 1984). Progression of this disease is fairly consistent across affected male children. Difficulty with balancing and walking is often noted before age two (Emery, 1994). Increased weakness of the hips and shoulders is evidenced by school age, with most boys requiring a wheelchair by age ten (Madorsky, et al., 1984). After childhood, the rate of progression of the disease is usually less obvious; however, most muscle groups eventually become

affected (Emery, 1994). Additional physical complications of DMD include "contractures", such as the turning inward of a foot, and "scoliosis", or curvature of the spine. This latter phenomenon can cause compression of a lung, resulting in compromised respiratory functioning. (Emery, 1994). The expected life span of males with DMD is between twenty and thirty years, with death most commonly associated with respiratory or cardiac failure (Madorsky et al., 1984). Presently, there is no cure for DMD. Treatment emphasizes the promotion of good health in general, the prevention of physical deformities, and the preservation of respiratory function (Emery, 1994).

Cognitive/Adjustment Issues and DMD

With regard to the psychosocial adjustment of males with DMD, scant empirical studies exist. Psychological research with this population has tended to focus on intellectual versus emotional functioning. The little information that is available remains inconclusive. A number of years ago Sherwin and McCully (1959) studied 15 boys with DMD, ages 10 through 14. The subject's verbal intelligence test scores ranged from 90 (Average) to 130 (Superior). Motor tasks were eliminated from study, given the nature of the disease. The authors concluded that boys with DMD showed cognitive development in keeping with population estimates, and that cognitive skills do not deteriorate as the disease progresses.

Sherwin and McCully also included "clinical" evaluation of their subjects via psychiatric interview and projective assessment. They concluded that the subjects "showed relatively little overt anxiety or depression, as such, in relation to the illness" (p. 61). Coping strategies tended to fluctuate between extremes of "passive nonparticipation" to "sudden impulsive aggressiveness" (p. 61). Excessive use of fantasy was identified via projective instruments. Not surprisingly, many of the subject's fantasies reflected themes of mobility and freedom. With the exception of the intelligence test data in which mean, standard deviation, and range were reported, results and interpretation of data appear largely subjective.

A few years after the Sherwin and McCully study, Zellweger and Hanson (1967) evaluated 38 boys with DMD, ranging in age from two to eighteen years. Assessment of intelligence was completed with the Wechsler Intelligence Scale for Children (WISC) and the Stanford-Binet Intelligence Scale (SB). Functional classification of all subjects was made on a one to ten scale, with "one" indicating the ability to climb stairs independently, and "ten" denoting bed existence. The authors used a cut-off score of 80 on the intelligence instruments to determine mental retardation. They reported that 16 of their 38 subjects (42%) reflected IQs below this cut-off. Consistent with the previous findings of Sherwin and McCully (1959),

Zellweger and Hanson concluded that the age of the affected boy did not predict his IQ score. Nor did the IQ of the affected boy decline with age and progression of disease. Unfortunately, these authors used an incorrect classification of mental retardation, overidentifying subjects as cognitively impaired.

Lincoln and Staples (1977) evaluated 68 children and adults with various neuromuscular diseases, including DMD. All subjects were given a battery of cognitive tests, including the Wechsler Intelligence Scale for Children (WISC) and the Wechsler Adult Intelligence Scale (WAIS). These researchers found "the incidence of subnormality in Duchenne dystrophies was shown to be no higher than might be found in a sample of the general population" (p. 214). Psychosocial variables were not addressed.

Lovelace (1983) described cognitive and personality factors of patients seen in his clinic. Three dystrophies--Myotonic dystrophy, Charcot-Marie-Tooth syndrome, and DMD--were reviewed. Lovelace stated that 20% of the DMD patients in his clinic "have subnormal intelligence" (p. 7). With regard to personality, males with DMD were characterized by the author as having "profound problems with self-image" (p. 8). Older patients, in particular, were described as "depressed" and "paranoid" (p. 8). These classifications were reflective of clinical judgement, however, and were not substantiated with a formal assessment instrument.

Leibowitz and Dubowitz (1983) studied both cognitive and emotional variables in their assessment of 57 boys with DMD, ages 3 to 13. The instrument of intelligence was not identified; however, the reference to "Verbal" and "Performance" scales suggests the Wechsler. Emotional functioning was assessed with the Rutter Behavior Questionnaire. Both parent and teacher versions were completed for each subject. Results of this study indicated subject's IQ scores fell "about 1 standard deviation below the mean, the majority functioning better on performance than on verbal tasks" (p. 17). In keeping with previous investigations, the authors did not find an IQ decline with age or progression of disease. With regard to emotional/behavioral issues, the authors concluded that "a high rate of emotional disturbance" (p. 18) is associated with DMD. Parents reported "neurotic" behavior at home, while teachers reported both "antisocial" and neurotic behavior in the school environment. No statistical data were displayed in this study to corroborate the conclusions provided.

In the survey study conducted by Madorsky et al. (1984), patients with DMD, parents, and professional health care workers related their needs in managing the psychosocial aspects of dying associated with DMD. Survey participants included 32 patients twelve to twenty-one years, 75 parents of affected males, and 43 professionals.

Physicians, allied health professionals (e.g., physical therapists), mental health professionals, and hospital administrators comprised the latter group. The specifically designed survey asked study participants to rate on a Likert-type scale the importance and availability of medical and psychosocial services. According to the respondent's ratings, 55% of parents and 53% of patients indicated life expectancy information was important. The desire for information regarding death was not related to the age of the patient. The professionals perceived counseling and support services as being more available to families than did the patients or parents themselves.

Recent Conceptualization of Coping with DMD

The concept of "coping with muscular dystrophy" is a recent phenomenon in the psychological literature. Emery (1994) specifically utilizes the term "coping process" (p. 64) to identify the psychological and emotional reactions subsequent to the diagnosis of a serious disease such as muscular dystrophy. He recognizes five stages: 1) shock and denial, 2) anxiety, 3) anger and guilt, 4) depression, and 5) psychological homeostasis. Attainment of the fifth stage, asserts Emery, is dubious, as the affected individual most likely always feels "a sense of loss and deprivation throughout life" (p. 66).

To summarize, it appears that sound empirical studies regarding the ways males with DMD cope with their chronic

and progressive illness do not exist. The available information regarding psychological aspects of DMD is inconclusive. Some investigations have found subnormal intelligence, while others have found no greater presence of cognitive deficiency than would be found in the general population. With regard to psychosocial adjustment, some studies have suggested that boys with DMD have emotional difficulties in a greater proportion to their non-affected peers. However, inadequate methodologies and statistical applications have characterized the data thus far. Studies utilizing reliable and valid measures of coping with this population have not been published to date.

Coping With Cystic Fibrosis

Another population of chronically ill individuals with demographics similar to DMD is the group of children and young adults with Cystic Fibrosis (CF). Like DMD, CF is a genetic disorder. Specifically, it is the most common genetic disorder affecting Caucasians, with a reported incidence rate of 1 in 2,000 births (Thompson, Hodges, and Hamlett, 1990). Cystic fibrosis is manifested by progressive decline of respiratory function, leading to a shortened life expectancy. The lungs, trachea, stomach, and pancreas are affected by the disease. Accompanying the disease are various complications, including shortened stature, decreased fertility in females, and sterility in males (Boyle, di Sant'Agnese, Sack, Millican, and Kulczychi,

1976). Uncomfortable daily regimens to loosen and expectorate phlegm are necessary, along with sleeping under a "mist tent" to enhance breathing capacity (Boyle et al., 1976). There is no cure for CF at present. However, with advances in medical treatment, many patients with CF are surviving into their late twenty's (Thompson, Gustafson, Hamlett, and Spock, 1992).

Adjustment Issues and CF

As with DMD, the CF literature on psychosocial functioning contains projective assessments and interviews. Unlike DMD, however, there has been a greater attempt in the recent CF literature to employ objective, norm-referenced measurement of psychological variables and appropriate statistical analysis. In the 1970s Tropauer, Franz, and Dilgard (1977) and Boyle et al. (1976) relied on psychiatric interviews and projective instruments such as the House-Tree-Person test and the Rorschach to assess adjustment among individuals with CF. With regard to the former study, the authors concluded that children with CF often manifest "chronically unexpressed feelings of anxiety, hostility or despair" (p. 215). Four main sources of stress for the CF patients were identified in the latter study, including: 1) altered physical appearance, 2) strained interpersonal relationships, 3) conflicts in upbringing, and 4) increased awareness of the future.

More recently, Thompson et al. (1990) compared 63 children with CF to 116 psychiatrically referred children and 177 nonreferred children from a community-based sample. The participants completed the Child Assessment Schedule (CAS), a semi-structured interview. Section one consists of questions regarding school, friends, and worries in which DSM-III items are embedded. The child is next asked in section two to discuss the onset and duration of positive symptoms identified in section one. Section three is an ongoing activity in which the interviewer records behavioral observations of the child with regard to 53 targeted behaviors. The authors reported "satisfactory test-retest and inter-rater reliability was demonstrated for presence or absence of diagnosis as well as for the diagnostically related symptom scores for conduct, depression, and anxiety disorders" (p. 749). Results of the CAS interviews were analyzed with an overall multivariate analysis of variance (MANOVA). The CF group did not differ from the nonreferred group with regard to total CAS score. The CF group and the nonreferred group endorsed significantly fewer problem items than the psychiatrically referred group in terms of total CAS score. However, the CF group and the psychiatrically referred group endorsed more problem items than the nonreferred group in the content areas of "worries", "self-image", and "separation anxiety".

Thompson et al. (1992) evaluated forty-five CF children with regard to parent and self-reported behavior problems. The Child Assessment Schedule interview was completed with the children. A number of objective instruments were utilized by the children and their parents. The children completed the Children's Health Locus of Control Scales and the Self-Perception Profile for Children. These instruments are purported to measure health expectations and self-esteem, respectively. Parents completed the Symptom Checklist 90-Revised (SCL-90-R) to assess psychological distress. The Missouri Children's Behavior Checklist (MCBC) was completed by parents as a measure of their child's adjustment. (Psychometric properties of the instruments were reported by the authors). Analysis of variance was utilized on obtained scores. Findings suggested that poorer adjustment scores on the Child Assessment Schedule and the Missouri Children's Behavior Checklist were related to poorer general self-worth scores on the Self-Perception Profile for Children. Good and poor adjustment groups, per the MCBC and the CAS, did not differ in terms of their health locus of control orientation. Maternal distress scores derived from the SCL-90-R did not differ for good or poor adjustment groups, as identified by the MCBC and the CAS.

CF and DMD have similar features with regard to biological etiology and the presumed stress caused by the

illness. The interview format has been utilized throughout the literature to obtain information concerning coping with both acute and chronic medical situations. Studies on adjustment to CF and DMD have used interviews as well. However, specific sources of stress have been identified in the CF, but not the DMD literature.

Statement of the Problem

Children and adolescents who are chronically ill must cope with a number of stressors (Cohen and Lazarus, 1979; Mattsson, 1979; Natterson and Knudson, 1960; Spirito et al. 1994; Tropauer et al., 1977; Zelter et al., 1980). The general children's coping literature utilizes the active versus avoidant and problem-focused versus emotion-focused dimensions to conceptualize the process. However, only one study to date (Spirito et al., 1994) has conceptualized coping in chronically ill children and adolescents in terms of the active versus avoidant dimension. No study has been found which identifies chronically ill children and adolescents along the problem-focused versus emotion-focused dimension. Instead, existing studies have tended to classify subjects with regard to presence or absence of some behavioral or psychiatric phenomenon, such as "anxiety" or "neurosis". The bias tends to favor the presumption that chronically ill pediatric patients are at greater risk for psychosocial difficulties than their "healthy peers".

Continued investigation is needed to identify differences in and implementation of coping strategies with these subjects. Spirito et al. (1994) note that the literature is particularly deficient with regard to assessing changes in coping over the course of an illness. Current studies tend to "lump" subjects with various disorders together as well. Thus, measures of stress, coping, and adjustment have not been employed differentially with respect to specific populations of ill children and adolescents. The most important disease-related and intrapersonal-related factors for successful coping with chronic disease have yet to be clearly identified.

With regard to coping with a specific chronic disease, males with DMD have received minimal attention in the pediatric literature. However, specific stressors associated with CF, and psychological adjustment to CF have been identified in the literature. DMD and CF share certain demographic features: both are genetic, both are diagnosed at an early age, both are progressive, and both are fatal. It is expected that some similarities between the two groups in terms of sources of stress and the coping process might be identified. No such investigation has been conducted for boys with DMD. Nor has the literature to date investigated the coping strategies utilized by boys with DMD. Therefore, the CF literature serves as a model for the current study.

The current study is an exploratory investigation. The purposes are threefold: (1) to identify by age the stressors encountered by boys with DMD; (2) to identify by age the coping strategies employed by boys with DMD to manage the identified stressors; and (3) to evaluate the relationship between the severity and the frequency of identified stressors.

CHAPTER II

METHODOLOGY

In the following sections, a description of the subject selection process, the data collection procedure, and an overview of the methodology will be provided. Detailed explanation of the methodology was submitted to the Institutional Review Board prior to data collection (see Appendix A for Form B).

Subjects

The final sample for the current study consisted of 15 males, ages 9 to 19, with the diagnosis of Duchenne Muscular Dystrophy (DMD). All subjects were affiliated with the clinics provided by the Muscular Dystrophy Association of Greater East Tennessee. This organization assists patients in the areas surrounding Chattanooga and Knoxville, Tennessee. Additionally, MDA patients residing in North Georgia are generally included in the Chattanooga catchment area. MDA provides free monthly clinics in both Chattanooga and Knoxville. Medical management and ongoing therapy issues are addressed at the monthly clinics through individual contact with physicians and physical therapists.

The MDA patient services coordinator utilized the organization's computer data base to assist the investigator in identification of subjects. Within the Chattanooga area, 19 males with DMD are regular clinic participants. Of these 19, four males were below the age cutoff and 5 males were

above the age cutoff. Of the ten remaining, all were students and known to the patient services coordinator to be performing adequately at school. School performance was utilized as an indicator of cognitive functioning, versus formal assessment with an intelligence test.

Within the Knoxville area, 16 males with DMD are regular clinic participants. Two patients fell below the age cutoff and 5 patients were above the cutoff. Of the nine remaining, all were students and were known to the patient services coordinator to be performing adequately at school. Therefore, a total of 19 males were identified for participation. Letters of informed consent were mailed to the parents of these patients. The return rate was 79% (see Appendix B for sample selection data).

Originally, a minimum age of 10 and a maximum age of 18 were selected for inclusion. However, because of small sample size, two 9 year old subjects and 2 nineteen year old subjects were included. The two youngest subjects presented with no obvious cognitive deficits and appeared well able to participate in the self-report format. As previously noted, no formal intellectual assessment was administered, but all subjects appeared within normal intellectual limits. All subjects were students, with the two oldest participants attending a local junior college. All subjects resided with their parents.

Procedures

The investigator provided the letters of informed consent and self-addressed stamped envelopes to the MDA patient services coordinator. The coordinator subsequently mailed the information to the parents of the prospective subjects, asking permission for their sons to participate in the study (see Appendix C for sample informed consent letter). Using this procedure, the investigator only knew the identity of those males who would actually participate. When informed consent letters were returned, the investigator obtained phone numbers of parents from the MDA patient services coordinator. Parents were subsequently contacted by the investigator by phone to schedule interview appointments. During the phone conversation, parents were prompted to ask any additional questions regarding the purposes and procedures of the study.

The data collection procedure was consistent with qualitative research methodology. The method of data collection was a semi-structured interview format. Interviews were conducted individually, with suggested locations being either in subjects' homes or at the monthly muscular dystrophy clinic (see Appendix D for sample interview recording form). Parents were given the option of locations. Ten interviews were conducted in the subject's home; three interviews were conducted at the monthly MDA

clinic; two interviews were conducted at the subject's school.

At the time of the interview, subjects were told that they were assisting the investigator with a study. The purposes of the study, as noted in the "Statement of the Problem" section, were explained. Subjects were given a consent form to sign at the time of the actual interview, and a witness was present for verification (see Appendix E for sample subject consent form). The subject's age was the only demographic or identifying information recorded on the interview form. In six cases, parents were present at the time of the interview. However, they remained in another portion of the room and did not directly respond or interject. Each subject received a number (e.g., Subject #1). Numbers were recorded on a log sheet in chronological order of the interview, along with subject age and location of interview.

Measures

Part I

The semi-structured interview form was developed by the investigator. The interview protocol is described below. Instructions to subjects, research questions to be asked, data recording, and data sorting procedures are described.

Simplified operational definitions of "stressor" and "stress symptoms" (in accordance with Matheny et al., 1993) were given to subjects. "A stressor is a situation, event,

or thought that causes one to experience stress. This stress or stress response represents your adjustment to problems. Stress symptoms are your reactions - the negative things you think or feel because of stress". Examples of common stressors and stress symptoms relevant to the pediatric/adolescent experience were provided for clarification (Matheny et al., 1993):

Examples of Stressors	Examples of Stress Symptoms
Arguments with parents	Depression
Violence at school	Fear

Subjects were then asked to focus on and identify the disease-related stressors with which they must cope: "Please tell me the hardest things about having muscular dystrophy. What bothers you the most?" Each subject identified at least one stressor, and no limit was imposed. Responses were recorded verbatim on the interview form. A separate form was utilized for each stressor named.

Part II

Subjects were then asked to consider the severity of the identified stressor(s) on a 5-point Likert-type scale: "How much stress does _____ cause you?" Anchors were provided and read to each subject. The investigator circled the endorsed responses on the interview form. Severity ratings were as follows:

1 = Very little

2 = Some

3 = A lot

4 = Very much

5 = An extreme amount

Subjects were then asked to consider and rate the frequency of the perceived stressor(s) on a 5-point Likert-type scale: "How often do you have to handle _____ ?" Anchors were provided and read to each subject. The investigator circled the endorsed response on the interview form. Frequency ratings were as follows:

1 = rarely, with the stressor occurring less than once per month

2 = sometimes, with the stressor occurring at least once per month

3 = often, with the stressor occurring at least once per week

4 = frequently, with the stressor occurring daily

5 = constantly, with the stressor occurring more than one time daily

Part III

The final portion of the interview consisted of asking subjects to identify the coping strategies they employ to manage the identified stressors. A simplified operational definition of coping was given to the subjects, in keeping with the Folkman and Lazarus (1980) model: "Coping refers to what people do to handle the stressors in their lives. Some people may think about something to feel better, or

some people may do something to feel better". Subjects were then asked to reflect upon the stressors previously identified and to report how they typically coped with them. For example, the investigator queried a 15 year old subject: "When you have trouble finding an accessible bathroom in public, how do you handle it?" Subjects could give more than one coping strategy per stressor if they wished. Responses were recorded verbatim on the interview form.

Data Sorting Procedure

After all subjects had been interviewed, interview recording forms were given to two independent doctoral-level psychologists for sorting. The areas of concern previously identified from the cystic fibrosis literature served as the basis for the sorting procedure. The investigator provided the sorters with the following instructions: "Please sort the identified stressors into the following semantic categories: (1) concern regarding physical appearance, (2) concern regarding interpersonal relationships, (3) concern regarding conflicts in upbringing, and (4) concern regarding increased awareness of the future". The sorters were given large index cards labeled with the four semantic categories to assist in organization. Inter-rater reliability for the sorting of reported stressors was calculated by the investigator using the formula: Agreements - Disagreements/Agreements (A - D / A). Results indicated good inter-rater reliability, with $\underline{r} = .89$. (Only three

responses were disputed). The category name of the stressor (e.g., "concern regarding physical appearance") was written on the interview form by the investigator following the calculation of reliability. Those responses which did not fall under the four established categories were set aside and subsequently thematized by the investigator. Thus, the investigator served as a third rater. Alternative categories of stressors were identified by the investigator. Their category names were subsequently indicated on the interview forms.

Coping responses were then evaluated and sorted by the independent psychologists as either active or avoidant. Sorters were given the definitions of "active" and "avoidant" coping responses in accordance with the definitions of Curry and Russ (1985) and Suls and Fletcher (1985). Degree of inter-rater reliability was calculated by the investigator using the formula: Agreements - Disagreements/ Agreements (A -D / A). Results indicated good inter-rater reliability, with $\underline{r}$ = .97. (Only one response was disputed). Identification of coping responses as either active or avoidant was recorded on the interview form by the investigator following sorting. The investigator served as the third rater for the one response on which the two independent raters did not agree.

Separate interview forms were used for each identified stressor, and subjects did not identify the same number of

stressors. Data analysis was not based on subject-to-subject comparison, but rather on age-related differences.

Research Questions

Three research questions were investigated in this study: 1) Does the perception of a stressor differ as a function of age? Older subjects were believed to be more likely than younger subjects to be concerned with fatality issues. Stating the converse, younger subjects were believed to be more concerned with day-to-day adaptation than the long-term outcome of their disease; 2) Does the coping strategy employed by a subject differ as a function of age? Older subjects were believed to be more likely than younger subjects to utilize active strategies. Stating the converse, younger subjects were believed to be more avoidant in their coping style; and 3) Is there a positive correlation between the perceived severity and the perceived frequency of a stressor? Those stressors perceived as most difficult to cope with were predicted to be those that occurred with the most frequency.

Data Analysis

The research design for this study was primarily qualitative and exploratory in nature. There was no random assignment to experimental or control groups, as would be the case in experimental research. The investigator's primary interest was in the subject's perception of his disease, as reflected in his own words. The basis for the

investigation of stressors in the current population was found in the cystic fibrosis literature. A secondary interest involved the identification of coping strategies used by males with Duchenne muscular dystrophy.

Subjects for this study constituted a naturally occurring group, consisting essentially of all available males ages 9 through 19 with DMD in the geographical area. (For purposes of this study, "geographical area" was defined as a 150 mile radius from Chattanooga, Tennessee).

CHAPTER III

RESULTS

The following sections describe both the descriptive and qualitative analyses of five variables: 1) type of stressor; 2) perceived severity of a stressor; 3) perceived frequency of a stressor; 4) type of coping strategy; and 5) age of subject.

Part I - Descriptive

Research Question One: Influence of Age on Stressor

The first research question to be answered concerned the nature of the relationship between age and perceived stressor in a population of males with DMD. It was presumed that older subjects would be more concerned than younger subjects with fatality issues. Younger subjects, on the other hand, were predicted to be more concerned with day-to-day adaptation than long-term outcome.

The interview and data sorting processes were described in Chapter II. The data set obtained from DMD males revealed the following six categories of stressors in order of frequency: 1) problems with mobility (e.g., inability to walk); 2) increased awareness of the future (e.g., knowledge of premature death); 3) concern regarding interpersonal relationships (e.g., difficulty initiating conversation with females); 4) daily living issues (e.g., needing help with toileting); 5) perceptions of being singled out (e.g., inability to participate in a particular activity at

recess); and 6) concern regarding physical appearance (e.g., being stared at in the wheelchair). Table 1 lists the types of stressors categorized in this study, along with the number of endorsements for each category. Table 2 lists the mean number of stressors by age group. Means for the three age groups were as follows: 9 through 11 = 1.7; 13 through 15 = 2.2; 16 through 19 = 2.3. Thus, group means were fairly close, and no one subject overly represented the data set.

This first research question addressing age and the six different types of perceived stressor was answered via calculation of percentages of responses. The specific question to be answered was: Does age appear to influence the types of stressors identified by boys with Duchenne muscular dystrophy? In this study, age has three artificially imposed levels - 9 through 12 years, 13 through 15 years, and 16 through 19 years. Six levels of the stressor variable were initially identified in the sorting process.

Table 3 depicts the number of endorsements by age group for each stressor. The investigator collapsed the original six types of stressors into two broader domains -- "physical issues" and "psychosocial issues" -- to make descriptive information more concise. The physical issues domain consisted of responses from "problems with mobility" and "daily living issues." Examples of stressors in this domain

Table 1

Types of Stressors in Order of Frequency[a]

Types	# of Responses
Problems With Mobility	11
Increased Awareness of the Future	7
Concern Regarding Interpersonal Relationships	4
Daily Living Issues	4
Perceptions of Being Singled Out	3
Concern Regarding Physical Appearance	1
Total Responses =	30

[a]Inter-rater reliability for categorization of stressors =.89

Table 2

Mean Number of Stressors by Age Group

Ages 9 - 12	Ages 13 - 15	Ages 16 - 19
1	3	4
4	2	2
2	2	1
1	3	
1	1	M = 2.3
2	M = 2.2	
1		
M = 1.7		

Table 3

Original Data Set: Number of Endorsements by Age for Each Stressor

	MO	AF	IR	DL	SO	PA[a]
9 - 12	n = 7	n = 0	n = 0	n = 3	n = 1	n = 1
13 - 15	n = 3	n = 3	n = 2	n = 1	n = 1	n = 0
16 - 19	n = 1	n = 4	n = 2	n = 0	n = 1	n = 0
Total n:	11	7	4	4	3	1

[a]MO = Problems with Mobility
AF = Increased Awareness of the Future
IR = Concern Regarding Interpersonal Relationships
DL = Concern Regarding Daily Living
SO = Perception of Being Singled Out
PA = Concern Regarding Physical Appearance

included inability to ambulate, and requiring assistance with dressing. The psychosocial issues domain consisted of responses from "increased awareness of the future", "concern regarding interpersonal relationships", "perceptions of being singled out", and "concern regarding physical appearance." Examples of stressors in this domain included fear of choking and embarrassment caused by staring from others.

Table 4 depicts the percentages of responses in each age group utilizing the physical and psychosocial themes. Results indicated that responses from the oldest two age groups reflected more concern about psychosocial stressors than those from the youngest subjects. Males 16 through 19 years (6 responses) and males in the 13 to 15 year old category (7 responses) reported more psychosocial themes than males 9 through 12 (2 responses). Conversely, the youngest subjects, 9 to 12 years, reported more concern regarding physical/daily living stressors (10 responses) than 13 to 15 year olds (4 responses) and 16 to 19 year olds (1 response).

Research Question Two: Influence of Age on Coping Style

This research question was answered in two parts. In part one, descriptive information was derived by calculating the percentages of responses for the entire sample which fell in either the active or avoidant coping domain.

Table 4

Percentages of Stressor Responses by Age

	Physical	Psychosocial[a]
Ages 9 - 12 n = 7	84% (10 responses)	16% (2 responses)
Ages 13 - 15 n = 5	36% (4 responses)	64% (7 responses)
Ages 16 -19 n = 3	14% (1 response)	86% (6 responses)

[a]Physical = Problems with Mobility, Daily Living Issues

Psychosocial = Increased Awareness of the Future, Concern Regarding Interpersonal Relationships, Perceptions of Being Singled Out, Concern Regarding Physical Appearance

The second part of the question considered age differences in reported coping strategies. It was expected that older subjects would identify active coping strategies more frequently than younger subjects. Age consisted of three levels-- 9 through 12, 13 through 15, and 16 through 19. Coping responses consisted of either active or avoidant.

In part one of the second research question, percentages of active and avoidant coping responses were obtained for the entire sample (N = 30 responses). Results indicated that 77% of responses fell into the active coping domain, while 23% of responses fell into the avoidant domain. As a group, subjects identified active coping strategies more frequently in dealing with disease-related stressors.

Part two of this question compared types of coping by the three previously established age groups (i.e., 9 through 12 years, 13 through 15 years, and 16 through 19 years). Table 5 depicts percentages of responses in active versus avoidant coping domains. Results indicated that responses of boys ages 13 to 15 reflected the greatest disparity between reported active and avoidant coping strategies. In this age group, 91% of responses (n = 10) corresponded to the active domain, while 9% of responses (n = 1) corresponded to the avoidant domain. Less disparity was noted between the responses of the youngest and oldest age

Table 5

Percentages of Coping Responses by Age

	Active	Avoidant	
Ages 9 - 12 n = 7	67% (8 responses)	33% (4 responses)	40% (12)
Ages 13 - 15 n = 5	91% (10 responses)	9% (1 response)	37% (11)
Ages 16 - 19 n = 3	71% (5 responses)	29% (2 responses)	23% (7)
Totals	77% (23)	23% (7)	

groups. Sixty-seven percent of the responses (n = 8) from the 9 to 12 year olds corresponded to the active coping domain, whereas 33% of responses (n = 4) corresponded to the avoidant domain. Among 16 to 19 year olds, 71% of responses (n = 5) corresponded to the active coping domain, while 29% of responses (n = 2) corresponded to the avoidant domain. In general, participants in this study used more active coping strategies (77%) than avoidant coping strategies (23%).

Research Question Three: Correlation Between Frequency and Severity of a Stressor

Research question three concerned the relationship between severity and frequency of perceived stressors. It was predicted that those stressors identified as the most stressful would also be rated as occurring with the greatest frequency. Ratings for stressfulness ranged from one to five: 1 = a little ; 2 = somewhat; 3 = a lot; 4 = very; 5 = extremely. Ratings for frequency ranged from one to five as well: 1 = rarely; 2 = sometimes; 3 = often; 4 = frequently; 5 = constantly. A Pearson product-moment correlation was run between the two variables.

Figure 1 depicts the correlations between severity and frequency of stressors for the entire sample and also by age groups. A moderate correlation (r =. 46) was obtained between the variables of severity and frequency for the entire sample. The mean frequency rating was 3.0 ("often"),

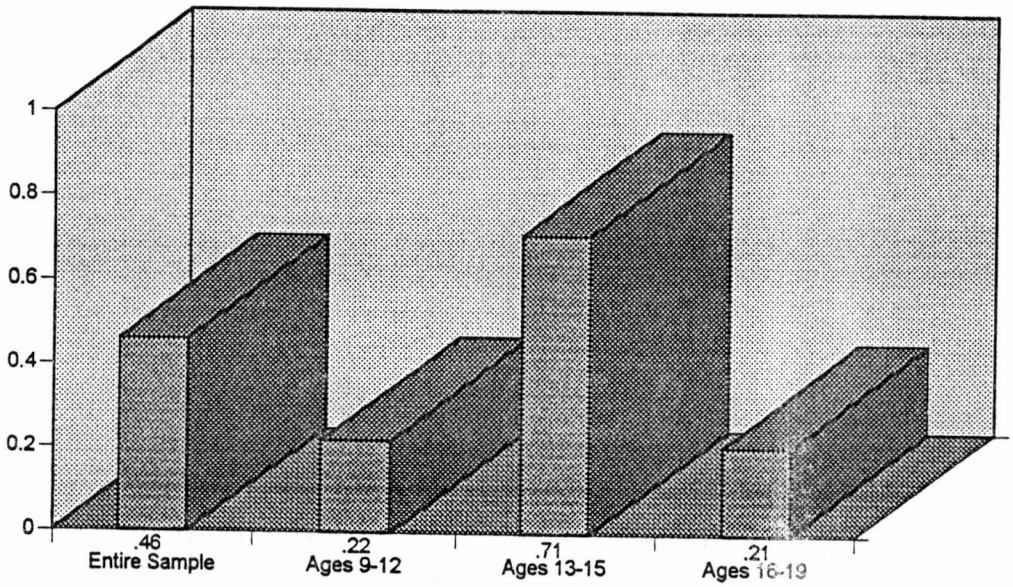

Figure 1

Correlations Between Severity and Frequency of Stressors

with a standard deviation of 1.0. The mean severity rating was 3.2 ("a lot"), with a standard deviation of 1.3. For the entire sample, it appears that the stressors reported are occurring at least once per week, and are moderately stressful.

Separate Pearson product-moment correlations between severity and frequency of perceived stressors by age group were then calculated. For the 9-12 age group, a low positive correlation (r = .22) was obtained. The mean frequency rating for this group was 3.4 (between "often" and "frequently"), with a standard deviation of 1.0. The mean severity rating for this group was 3.5 (between "a lot" and "very"), with a standard deviation of 1.3. For this group, reported stressors are occurring at least once per week, and are at least moderately stressful.

For the 13-15 age group, a strong positive correlation (r = .71) was obtained. The mean frequency rating for this age group was 2.7 (between "sometimes" and "often"), with a standard deviation of 1.0. The mean severity rating for this group was also 2.7 (between "somewhat" and "a lot"), with a mean of 1.4. For this group, the identified stressors are occurring less than once per week, but generally produce "a lot" of stress.

For the 16-19 age group, a rather low positive correlation (r = .21) was obtained. The mean frequency rating for this age group was 3.0 ("often"), with a standard

deviation of .81. The mean severity rating for this group was 3.5 (between "a lot" and "very"), with a standard deviation of .97. For this group, the identified stressors are occurring at least once per week, and are producing more than "a lot" of stress.

Part II - Qualitative Analysis

The data collection and sorting procedures previously described in Chapter II represent the initial qualitative component of this study. Secondly, the investigator subsequently reviewed and thematized all responses which further contributed to the qualitative analysis.

Table 6 displays a frequency count of active and avoidant coping strategies by age. The top portion of table 6 identifies the number of active strategies by age, with three distinct strategies identified by the investigator: 1) cognitive reframing; 2) behavioral persistence; and 3) cognitive/behavioral distraction. Cognitive reframing refers to the subject's purposeful attempt to redefine his situation in order to reduce the emotional impact of the stressor. Behavioral persistence is simply the subject's continued efforts to pursue a desired outcome despite obstacles. Cognitive/behavioral distraction represents the subject's purposeful attempt to focus attention on an alternative thought or behavior to minimize the emotional impact of the stressor.

Table 6

Frequency Count of Coping Strategies by Age

Active Strategies

	Cognitive Reframing	Behavioral Persistence	Cog/Behav Distraction
9 - 12	2	4	2
13 - 15	3	6	1
16 - 19	3	2	0
Totals	8	12	3

Avoidant Strategies

	Passive Retreat	Social Withdrawal	Task Avoidance	Cognitive Avoidance
9 - 12	1	1	1	1
13 - 15	0	0	0	1
16 - 19	0	1	0	1
Totals	1	2	1	3

The lower portion of Table 6 identifies the number of avoidant coping strategies by age. Four distinct strategies were identified: 1) passive retreat; 2) social withdrawal; 3) task avoidance; and 4) cognitive avoidance. Passive retreat refers to the subject's withdrawal from a problem situation in wake he has determined he cannot effect a positive outcome. Social withdrawal is the subject's purposeful removal of himself from a social to a solitary environment. Task avoidance refers to the subject's conscious effort to avoid that task or situation perceived as stressful. Cognitive avoidance refers to the subject's conscious mental effort to avoid thinking about a problem situation.

Tables 7 and 8 display examples of active and avoidant comments, respectively, made by subjects which correspond to the strategies previously described. Within the active coping domain, there appears to be a slight trend for behavioral persistence to be used more frequently by 9 to 12 and 13 to 15 year olds than either cognitive reframing or cognitive/behavioral distraction. Among the oldest group, cognitive reframing and behavioral persistence appeared to be used with approximately the same frequency. No exemplars of cognitive/behavioral distraction were noted among the 16 to 19 year olds.

Fewer total responses were available for analysis within the avoidant domain. However, one trend noted was

Table 7

Illustrative Comments Reflecting Active Coping

Age	Comments	Strategies
19	I'm religious-I pray a lot. I also look at it like, "I'm healthier than a lot of kids with this disease."	Cognitive Reframing
14	I'll have an assistant at the high school, but I'll try to do everything as independently as possible.	Behavioral Persistence
15	When I can't find an accessible toilet out in public, I just have to keep trying.	Behavioral Persistence
15	When I get home, I do something to take my mind off of it (referring to problems with decreased mobility at school).	Cognitive/ Behavioral Distraction
11	When people stare at me, I think, they're not staring because they hate me, they're just curious.	Cognitive Reframing

Table 8

Illustrative Comments Reflecting Avoidant Coping

Age	Comments	Strategies
9	I'll let Billy and Joe in front of me because I can't run.	Passive Retreat
10	When I worry about not being able to walk, I like to be alone.	Social Withdrawal
10	I don't always go to my physical therapist. Sometimes I tell my mom I don't want to go.	Task Avoidance
10	I try not to think about it when I need help with little things.	Cognitive Avoidance
19	I'm shy. I worry about talking to girls. I usually just don't talk to them.	Task Avoidance

for the youngest group to report strategies corresponding to all four categories. A second trend among avoidant responses was the use of cognitive avoidance strategies among all three age groups.

CHAPTER IV

DISCUSSION

As previously noted in Chapter I, there are millions of children and adolescents in the United States diagnosed with a chronic illness (Midence, 1994). Despite these numbers, there is little research to address the existence of disease-specific stressors, and the types of mental health problems encountered by these groups remains largely unknown (Eiser, 1990). The population of individuals with cystic fibrosis (CF) serves as an exception, in that efforts have been made to identify and understand the unique psychosocial challenges of this group (Thompson et al., 1990).

The current study attempted to explore the relationship between perceived stressors and coping strategies in a sample of males with Duchenne Muscular Dystrophy (DMD). Inherent in this investigation was the issue of age; specifically, the degree to which age mediated the types of stressors perceived and the types of coping strategies reported.

Part I - Descriptive

Research Question One: Influence of Age on Stressor

Four main sources of stress have been identified in the CF literature, and served as the basis for the current analysis of DMD subjects: 1) concern with physical appearance; 2) concern with interpersonal relationships; 3) conflicts in upbringing; and 4) increased awareness of the

future (Tropauer et al., 1976). In the current study identification of stressors by participants was explored, in addition to examining possible age effects related to stressors. Specifically, descriptive data were obtained to address the question of whether age appeared to influence the types of stressors identified.

The subjects in the current study expressed concerns which were categorized into six areas, consistent with issues identified from previous research (Cohen & Lazarus, 1979; Tropauer et al., 1976). Responses from DMD subjects revealed the following sources of stress: 1) problems with mobility; 2) daily living issues; 3) increased awareness of the future; 4) concern with interpersonal relationships; 5) perceptions ofbeing singled out; and 6) concern with physical appearance. Thus, the categories of stressors identified by DMD subjects bore some similarity to categories of stressors identified by CF subjects.

Perceptions of physical appearance, perceptions of social relationships, and awareness of future implications of the disease were noted by both chronically ill groups. However, problems with mobility, daily living issues, and perceptions of being singled out were unique to the DMD group. The implication from this comparison is that disease-related stressors vary between groups of individuals with chronic illness. The exploratory results of this study provided some insight into the specific disease-related

concerns of males with DMD and distinguished their concerns from another chronically ill sample.

Identification of "increased awareness of the future", "concern regarding interpersonal relationships", and "concern regarding physical appearance" was consistent with the types of stressors identified by the chronically ill cystic fibrosis population. There was no support in the current study, however, for the stressor "conflicts in upbringing" as noted by the group with cystic fibrosis. Three categories of stressors, "problems with mobility", "daily living issues", and "perceptions of being singled out" emerged as specific to the DMD group.

With regard to age and type of stressor perceived, age differences were observed when percentages were calculated. Results were consistent with the researcher's presumptions that older subjects would report more psychosocial concerns than younger subjects, and would specifically express concern regarding fatality. As predicted, younger subjects' responses reflected more concern about physical limitations and day-to-day functioning.

As predicted, the 9 to 12 year old subjects' responses reflected more concern with the physical implications of their disease on a day-to-day basis, such as inability to run or participate in sports. As noted in Table 1 and more specifically in Appendix I, problems with mobility were overwhelmingly rated as the primary concern of 9 to 12 year

olds. Secondly, concerns regarding daily living were reported by this younger group. In contrast, mobility and daily living issues appeared essentially unimportant to the oldest age group, despite the fact that the physical progression of their disease was the greatest and, therefore, the most limiting.

Concern Regarding Premature Death

The 13 to 15 year old group and the 16 to 19 year old group were more focused on psychosocial issues than the youngest group. This appears consistent with normal development. One would expect younger children to be more externally focused, while adolescents have developed a greater capacity for cognitive rumination (Cowan 1978). Adolescents possess the ability for abstract thought, and can entertain possibilities which are yet to exist. Therefore, it is not surprising that concern with future disease complications and awareness of a shortened life-span were noted by subjects in the two adolescent groups. In contrast, none of the subjects in the youngest age group expressed concern regarding the implications of their disease for a premature death.

Among the 13 to 15 year old group, two of the five subjects (subjects # 1 and 4) specifically noted fear of premature death. Both were aged 15. This represents 40% of the respondents for this age group. Subject # 1 stated that thoughts of death produce "a lot" of stress and occur "at

least once per month", according to the Likert scales provided. However, he further named two other categories of stressors, which indicates that thoughts of death are not his sole source of stress.

Subject # 4 noted that the thought of death is "a little" stressful, but that it occurs at least once per month, according to the Likert scales provided. Only one other stressor, which was related to mobility, was noted by this subject.

The oldest age group consisted of only three subjects. Two of the three subjects -- roughly 66% -- reported thoughts of death as sources of stress. Subject # 3, age 19, appeared very focused on future implications of disease in that three of his four identified stressors reflected themes of death. His fourth identified stressor involved interaction with young women. He specifically noted fear of choking, and was aware that patients with DMD frequently die as a result of respiratory arrest. He ranked his concerns as "very" and "extremely" stressful, with occurrences ranging from "at least once per month" to "at least once per week", according to the Likert scales provided.

Subject # 10, also age 19, rated thoughts of death as "very" stressful and occurring "daily". He identified only one other source of stress -- interacting with girls. Thus, in both of the oldest subjects, thoughts of death and concern with opposite sex relationships coexisted. Sadly,

it appears that these subjects have the normal developmental drive for a relationship, but may have some ambivalence toward social approach because of their disease and the eventuality of premature death.

Research Question Two: Influence of Age On Coping Style

This question considered the type of coping style, active or avoidant, most frequently reported by the participants with DMD. In part one, the entire sample was considered; active styles were clearly reported as most frequently utilized relative to avoidant coping styles. However, when examining type of coping style by age in part two, age differences were noted.

The 13 to 15 year olds reported substantially more use of active strategies. The disparity between active and avoidant strategies was less pronounced with the youngest and the oldest age groups, although the trend to use active more than avoidant clearly existed. Thus, the current study did not fully support specific developmental influences with regard to coping strategy. Results are somewhat in contrast to what was expected.

According to the literature, active coping among "normal" children and adolescents increases with age (Peterson, 1989). In general, active strategies are associated with better adaptation than avoidant strategies as well. Thus, using "normal" youngsters as a frame of

reference, it appears that subjects in the current study somewhat resemble their non-diseased peers, in that active coping was used predominantly more than avoidant coping at all age levels. Moreover, the trend was for the oldest two groups to report active strategies more frequently than the youngest subjects.

According to the literature, effective coping tends to increase with age (Rutter, 1983). Active coping has been considered more effective than avoidant coping in a variety of contexts that specifically involve long-term situations. While no specific assessment of "effectiveness" was completed in this study, it may be suggested that the prevalence of active coping strategies is preferable to avoidant coping strategies, given the long-term nature of the disease.

As Suls and Fletcher (1985) note in their meta-analysis of the adult coping literature, avoidant strategies may be more functional in the short-term until the individual is able to apply more problem-focused strategies to deal with an unpleasant long-term reality. Thus, the adoption of active coping strategies may signal the individual's gradual acceptance of an unavoidable situation (i.e., premature death).

However, results from this study do not completely support more use of active strategies with increasing age. It was the 13 to 15 year old responses that reflected more

active strategies. This may be due to other developmental factors which are superimposed on the overall developmental sequence. That is, this particular age group of mid-adolescence may be more overwhelmed by stressors in general.

Another possible explanation for this finding is that the imposed age levels are narrow and may be susceptible to the influence of inherent developmental factors associated with adolescence. In their study of over 500 adolescents with multiple chronic illnesses, Zelter et al. (1980) noted that dividing adolescent age ranges sometimes confounds results due to overlapping developmental issues.

Research Question Three: Correlation Between Frequency and Severity of A Stressor

Severity and frequency of a perceived stressor were predicted to be positively correlated. Specifically, it was presumed that subjects who identified stressors as particularly difficult to manage would also identify those stressors as occurring with greater frequency.

When a Pearson product-moment correlation for the entire sample was computed, a moderate positive correlation between severity and frequency was obtained. Thus, the hypothesis regarding the relationship between frequency of a stressor's occurrence and perceived severity was supported.

When considering the influence of age, it is noted that only the correlation from the 13 to 15 year old group was significant. Thus, particularly in the youngest and oldest

age groups, participants did not necessarily endorse a strong relationship between severity and frequency of a stressor. This suggests that a problem, such as awareness of the future, is quite distressing to the subject, but thoughts about this issue may be somewhat infrequent. Likewise, a problem situation such as difficulty with independent toileting may occur often, but may not necessarily be perceived as extremely difficult to manage.

The absence of a strong relationship across age groups between severity and frequency of a stressor suggests variability in subject's perceptions. In addition, the overall moderate correlation between severity and frequency of a stressor suggests that two distinct constructs were measured.

Part II - Qualitative Analysis

The qualitative analysis included categorization of the coping processes exemplified by participant responses. Three "active" strategies and four "avoidant" strategies which reflected coping constructs from previous literature (Curry & Russ, 1985) were identified. The active strategies represented an effortful attempt to minimize the emotional impact of the stressor (Cohen & Lazarus, 1979). The avoidant strategies reflected effortful attempts to deny the reality of the current situation. Examples of active and avoidant coping behaviors and cognitions reported by the participants were displayed in Tables 7 and 8.

When considering the influence of age, it was expected that older, rather than younger, subjects would predominantly report the use of cognitive strategies, whether in the active or avoidant category. This prediction was supported by the research by Curry and Russ (1985) with children in a medical setting. This research suggested that older children use more cognitive versus behavioral strategies for coping. However, the comments displayed in Table 6 provide evidence of the use of cognitive strategies at all age levels. As noted in both Tables 7 and 8, even subjects within the youngest age group verbalized the use of cognitive reframing techniques to mediate the impact of a stressor.

This finding, which is in contrast to other research (Peterson, 1989) as well as expectations, may be accounted for by individual characteristics of participants. Specifically, males in this particular sample may be brighter than their average peers. While males who were performing below grade expectations were excluded from this study, no controls on upper limits of cognitive ability were in place. Therefore, the range of the functioning level of the sample is not known other than "at least average." It is possible, then, that the youngest subjects may have been exceptionally bright and developmentally more mature, as exemplified by the use of cognitive coping strategies.

Contributions

The contributions of the current study are threefold. First, research addressing the psychosocial adjustment of children and adolescents diagnosed with a chronic illness is limited. This is particularly true of males diagnosed with DMD. Despite the rather high profile nature of muscular dystrophy in general (i.e., yearly telethon and sponsorship by Jerry Lewis), little has been done to identify the concerns or psychosocial adaptation of this population. By providing information regarding the perceived stressors and coping strategies of males with DMD, it is hoped that families, mental health practitioners, medical practitioners and educators will have greater understanding of this population.

The second contribution of this study involves the selection of methodology. The qualitative design of the study allowed subjects to express their concerns directly via an interview format. While there was structure imposed by the directive for subjects to name a stressor associated with their disease, subjects were essentially free to name any concern. Thus, no a priori agenda existed in the interview process.

The literature regarding coping with chronic illness has tended to use parents or teachers as raters in the evaluation of the youngster's behavior or emotional adaptation. Standardized instrumentation will dictate

outcomes to some extent. For example, when rating a youngster on a specific psychological instrument, it is more likely that some form of "pathology" will be identified by the very nature of the instrument given. This study was successful in allowing subjects to conceptualize their thoughts and feelings without prompting them with items on a standardized instrument.

Finally, the current investigation sheds a more positive light on individuals with a disability. Many researchers, educators and clinicians are biased in the belief that disability equals dysfunction. There is a tendency to pathologize differences, and this may be even more likely in a pediatric population. The current results help to "normalize" those with a disability by focusing on coping, and demonstrate that youngsters with a disability may react very similarly to their peers when confronted with a problem situation. In keeping with their non-disabled peers, subjects in the current reflected a tendency to utilize active strategies more as they aged. Moreover, the subjects in the current study identified both cognitive and behavioral strategies to cope with stressors. This suggests that, despite disability, males with DMD are flexible in the resources they bring to bear in a stressful situation. As suggested by Eiser (1990), individuals with disabilities are ordinary people in extraordinary situations.

Clinical Implications

Results of this study generally reflect a positive view of the psychosocial status of males with DMD. While no formal personality or mood instrument was administered to the subjects, results suggest that males with DMD are not generally "disturbed", as previously suggested by Leibowitz and Dubowitz (1983). Again, the focus is on coping and not on identifying pathology. Nor do current results support Lovelace's (1983) conclusion that males with DMD are "depressed" and "paranoid". In contrast, subjects in this study appeared well able to assess their situation and verbalize their concerns accordingly. The majority of responses suggest that the subjects employ effortful coping in response to their disease. As previously noted, these subjects are similar developmentally to their "healthy" peers. Thus, while previous studies have suggested that chronically ill youngsters with and without accompanying physical disability have more psychosocial difficulties than "healthy controls" (Cadman et al., 1987), no obvious pathology was detected in this sample. It is possible, therefore, that Zelter et al. (1987) are accurate in suggesting that chronically ill children may cope adequately.

Limitations

The primary limitation of this study is the small sample size, which has implications for external validity.

Thus, while data trends were demonstrated, specific generalizations about results cannot be made due to the small number of participants. Also, the study was based on a small sample size with a relatively wide age range, 9 years to 19 years. Given that the periods of childhood and adolescence represent marked psychosocial and cognitive change, inherent variability would be expected.

However, external validity was not the investigator's primary interest. The results reflect some association with another group of chronically ill individuals, but also identify some distinctive characteristics as well. Moreover, the results suggest some degree of developmental similarity to "normal" children and adolescents who are coping with stress.

Another limitation of the study is the inability to determine whether the coping strategies identified by subjects in the retrospective format are the ones actually implemented in the event of an actual stressor. It is quite possible that a subject may inadvertently have described a coping strategy that he does not consistently employ when faced with disease-related concerns. Moreover, the methodological procedure dictated the classification of coping responses as either active or avoidant. It is quite possible that "mixed" styles exist in this population, with situational variables dictating the individual's use of one type of strategy versus another. No one individual would be

expected to use one type of strategy exclusively. The individual may tend to use one approach more often than another. When specific responses were analyzed, however, only three subjects giving multiple stressors and coping strategies had responses falling in both the active and avoidant domains.

A final limitation of the current study is the absence of some type of outcome measure to further clarify the degree to which the coping strategies reported by subjects are effective in reducing the impact of perceived stressors.

Future Research

Future research should continue to explore the psychosocial variability within groups of chronically ill individuals. Efforts should be made to emphasize the strengths of those with a disability, rather than designing studies to support the "deficit model".

With regard to DMD, specific "environmental" factors (such as family support and quality of educational services), and "interpersonal" factors (such as temperament and stage of disease process) should be analyzed to determine their impact on the coping process. Such an analysis would yield information regarding the conditions under which effective coping takes place. Longitudinal research would also be beneficial for patients, families,

and clinicians by shedding additional light on the changes in coping responses over the course of the disease.

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APPENDICES

APPENDIX A

FORM B

IRB#_____

Date Received in ORC_____

THE UNIVERSITY OF TENNESSEE, KNOXVILLE

Application for Review of Research Involving Human Subjects

I. IDENTIFICATION OF PROJECT Date 4-26-96

Principal Investigator and Co-PI:

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There is no CO-PI for this study.

Title of Project: An Analysis of the Coping Strategies Used by Males With Duchenne Muscular Dystrophy

Department: Psychoeducational Studies Unit

Starting date: Upon IRB approval

Estimated completion date: May, 1997

External Funding Agency and Identification Number: not applicable

Grant submission deadline: not applicable

I. OBJECTIVES OF PROJECT

(1) To identify the perceived stressors encountered by males

with Duchenne muscular dystrophy:

(2) To identify the coping strategies employed by males with Duchenne muscular dystrophy:

(3) To determine what, if any, age differences exist in perceived stressors and coping strategies employed.

III. DESCRIPTION OF SUBJECTS

Working in conjunction with the Greater East Tennessee Muscular Dystrophy Association, the PI will attempt to obtain 50 males, ages 10 to 18, with the diagnosis of Duchenne muscular dystrophy (DMD). DMD, which is the most common form of dystrophy, only affects males. The disease is characterized by progressive muscle wasting and weakness. Progression of the disease is fairly consistent across affected males. Difficulty with balancing and walking is typically noted before age two. Increased weakness of the hips and shoulders is evidenced by school age, with most boys requiring a wheelchair by age 10. After childhood, the rate of progression of the disease is usually less obvious.

Mr. Lee Willard, Patient Services Coordinator for Greater East Tennessee MDA, will assist the PI in identification of subjects and the obtaining of parental consent. He will identify, through the computer data base, all males with DMD within the required age range. The PI will provide Mr. Willard with self-addressed stamped envelopes and informed consent/parental permission forms. The PI will assume all postage costs. Pending IRB approval, the informed consent letters will be mailed out to prospective subjects by Mr. Willard. The PI will contact those parents who return the signed form and subsequently arrange an interview time. Subject consent forms will be administered in person at the time of the interview.

IV. METHODS OR PROCEDURES

Following receipt of consent forms, the PI will contact parents of prospective subjects to arrange for an interview. Individual semi-structured interviews will be conducted with subjects either in their homes or at the muscular dystrophy summer camp held in July. Interview are expected to take less than one hour. Parents will be allowed to be present during the interview if they desire to do so.

Part I

Subjects will be told that they are assisting the PI with a research project to find out what stressors they encounter and what strategies they employ to cope with the stressors. They will be told they may drop out of the study at any time. Subjects will be given a simplified operational definition of stress in accordance with Eiser (1990), which emphasizes the idea of stress as a problem situation requiring a decision-making process. Examples of stressful situations will be provided to the subjects for clarification. In order not to bias the subjects toward a restricted response set, examples representative of a variety of domains will be given:

1. School-related
   a. not understanding an assignment
   b. failing a test

2. Interpersonally-related
   a. being teased or called names
   b. being punished by a parent

3. Disease-related
   a. needing help with clothing in the bathroom following toileting
   b. having a surgical procedure

Subjects will then be asked to identify the stressful situations with which they must cope. Each subject must identify at least one stressful situation, but no limit will be imposed. Responses will be recorded verbatim on the interview form. Subjects will then be asked to consider the severity of the identified stressor on a 5-point Likert-type scale. Anchors will be shown and read to each participant. The investigator will record the endorsed responses on the interview form. Severity ratings are as follows:

1 = A little
2 = Some
3 = A lot
4 = Very
5 = Extremely

Subjects will then be asked to rate the frequency of the perceived stressful situation on a 5-point Likert-type scale. Anchors will be shown and read to the participants. The PI will record the endorsed responses on the interview form. Frequency ratings are as follows:

1 = Rarely, with the situation occurring less than once per month

2 = Sometimes, with the situation occurring at least once per month
3 = Often, with the situation occurring at least once per week
4 = Frequently, with the situation occurring daily
5 = Constantly, with the situation occurring more than one time daily

Part II

The final portion of the interview will consist of asking subjects to identify the coping strategies they employ to manage the identified stressful situation. A simplified operational definition of coping will be given to the subjects, in keeping with the Folkman and Lazarus (1980) model: "Coping refers to what people do to handle the stressful situations in their lives. Some people may think about something to feel better, or some people may do something to feel better". Subjects will then be asked to reflect upon the stressors previously identified and report how they typically cope with them. Responses will be recorded verbatim. For clarification purposes, examples from the three previously identified domains will be given to the subjects:

1. School-related
   a. not understanding an assignment
      example: close the book and talk to another student
   b. failing a test
      example: decide to study harder next time

2. Interpersonally-related
   a. being teased or called names by somebody
      example: pretend to be sick to stay home, away from the person who teases
   b. being punished by a parent
      example: apologize and ask mom or dad for a second chance
3. Disease-related
   a. needing help with clothing in the bathroom after toileting
      example: hold my bladder for a long time so I don't have to ask for help
   b. having a surgical procedure
      example: breathe deeply when they stick me with a needle because breathing helps me relax

In summary, the procedure involves having subjects identify the stressors they encounter, rating the frequency and severity of the stressors, and identifying the coping strategies they use to manage the stressors. All subjects are treated in the same fashion in this quasi-experimental

design. There is no control or comparison group.

V. SPECIFIC RISKS AND PROTECTION MEASURES

Subjects in this study have a progressive degenerative disease with which they must deal daily. Given this, subjects routinely discuss disease issues with family members, teachers, and peers. Participation in this study is anticipated to present no greater risk of harm than is encountered in daily life or during a routine psychological evaluation. Thus, it meets the definition of "Minimal Risk Research". Subjects will be told they may drop out of the study at any time. Further, parents may withdraw their consent at any time. Parents may also be present during the interview process. Parent written consent and subject written consent will be obtained. In the unlikely event that a subject becomes distressed during the interview process, the interview will be terminated immediately and the parent will be contacted. If requested by parent or subject, additional supportive counseling will be available through the MDA support group, at no cost to the parent. Alternatively, individual counseling will be available if needed through two doctoral level psychologists at Siskin Hospital. These individual services will also be provided at no cost to the parent.

The Patient Services Coordinator, Mr. Willard, will know the identity of all subjects in this study, as he will mail out informed consent letters. However, it should be noted that Mr. Willard, by the nature of his position, has ongoing contact with the subjects and their families apart from this investigation. He assists them in obtaining equipment, medical consultation, and psychosocial support. He further organizes and attends the annual MDA summer camp, along with assisting in the annual MDA telethon. He will not have access to the completed interview forms revealing identifying information. However, the PI has agreed to share the results of the study with Mr. Willard to assist with the overall role of MDA. All identifying information will be concealed.

Given the method of data collection, the identity of all subjects will be known to the PI as well. However, only the PI will hold the subject's identity and the raw data as written on the interview form. Interview forms will be kept in a locked file cabinet in the PI's office (number 1), located on the second floor of Siskin Hospital, Psychology Department, Suite 2. Each interview form will be coded with a number, and the information obtained from the interview will be entered into the computer. The subjects' names will not be entered into the computer to insure confidentiality. Interview forms will be destroyed following data entry.

VI. BENEFITS VS.RISKS

Participation in this study is considered to put the child at no greater than minimal risk. The PI will not be introducing any new information to the subject in the form of the interview (i.e., subjects are already aware of their disease and resultant stressors). Moreover, by virtue of dealing with their disease on a daily basis, these subjects are accustomed to discussion with parents, teachers, and peers. In the unlikely event that a subject becomes distressed during the interview, the interview will be immediately terminated and the subject's parent (if not already present) will be contacted. As previously noted, additional psychosocial support is available through the MDA support group or through individual treatment with either of two psychologists.

With regard to the potential benefits of this study, the results are expected to assist professionals and families by increasing their awareness of the stressors associated with muscular dystrophy. The process may enlighten parents with regard to their son's psychosocial needs. In addition, information will be gained to assist with coping with a disability. Finally, participation in the study may encourage those not previously accessing the MDA support group to do so.

VII. METHOD OF OBTAINING "INFORMED CONSENT" FROM SUBJECTS

See attached parent letter detailing the basic elements of written informed consent.

See attached participant letter to be used to obtain written informed consent from subjects. In accordance with UTK guidelines, written consent will be obtained from subjects, as their ages will range from 10 through 18. Supplementary verbal explanation will be given if needed. A "witness" signature line is included as some subjects may be unable to write legibly, secondary to their disease.

VIII. QUALIFICATIONS OF THE INVESTIGATOR

The PI is a masters level school psychologist and licensed psychological examiner credentialed in the state of Tennessee. She has 12 years experience working with children in both the public education and hospital environments. For the past five years, the PI has worked in a physical rehabilitation hospital. A primary job role has been to assist children and adolescents in coping with a disability. The PI is also a volunteer for the Muscular Dystrophy Association, serving as a support group facilitator for over three years.

IX. ADEQUACY OF FACILITIES TO SUPPORT RESEARCH

The interviews will be conducted either in subject's homes or at the MDA summer camp, in accordance with parental preference. Those parents who wish to be present during the interview will likely elect for home as the interview site. Those interviews occurring at camp will be conducted in a quiet room selected by Mr. Willard, Patient Services Coordinator. The room utilized will be away from other campers, so as to insure privacy and confidentiality. Mr. Willard will bring the subject to the interview room so that other campers will not see the PI or suspect anything negative with regard to the participant.

X. RESPONSIBILITY OF PROJECT DIRECTOR

By the Compliance with the policies established by The University of Tennessee, Knoxville, Institutional Review Board, the principal investigator subscribes to the principles stated in "The Belmont Report" and standards of professional ethics in all research, development, and related activities involving human subjects under the auspices of The University of Tennessee.

a. Approval will be obtained from the Institutional Review Board prior to instituting any change in the research project.

b. Development of any unexpected risks will be reported to the IRB.

c. Signed consent statements will be kept for the duration of the project and for at least three years thereafter.

Principal Investigator

_____
Name

_____
Signature Date

Advisor and Dissertation Committee Chair

_____
Name

_____
Signature Date

XI. **DEPARTMENTAL REVIEW**

Psychoeducational Studies Unit

Head _____
Name

_____
Signature Date

The application described above has been reviewed by the departmental committee and has been approved.

Chair,
Departmental
Review
Committee _____
Name

_____
Signature Date

APPROVED: Steven B. Pulik
Coordinator of Compliances
Office of Research

_____ _____
Signature Date

APPENDIX B

SAMPLE SELECTION DATA

Chattanooga, Tennessee

DMD Clinic Participants	19	
Total below age cutoff	4	21%
Total above age cutoff	5	26%
Total meeting age and cognitive criteria	10	53%
Total return rate	7	70%

Knoxville, Tennessee

DMD Clinic Participants	16	
Total below age cutoff	2	12%
Total above age cutoff	5	31%
Total meeting age and cognitive criteria	9	56%
Total return rate	8	89%

N for total sample = 15
Return rate for total sample = 79%

APPENDIX C

INFORMED CONSENT

Dear Parent:

I am a doctoral student at the University of Tennessee, Knoxville. I am conducting a research project to identify the perceived stressors and coping strategies used by boys with Duchenne Muscular Dystrophy. The procedure for collecting this information will involve an interview with your son which should take less than one hour. I am proposing to conduct these interviews either in your home or at the monthly MDA clinic. The interviews are projected to occur during the months of March and April, 1997. The entire project is expected to be completed by July, 1997.

This study will help determine how boys with muscular dystrophy cope with their disease, and if there are differences in coping depending upon the age of the boy, the severity of the stressor, or the frequency of the stressor. The benefit of this study is that families and professionals may gain more awareness of the issues related to coping with a disability. Participation in this study is anticipated to present no greater risk of harm than is encountered in daily life or during a routine psychological evaluation. In the unlikely event that your son should become distressed during the interview, the interview will be immediately terminated. Also, parents may be present during the interview. Parents may remove their son from the study at any time for any reason, without penalty. Participants may drop out at any time to any reason, without penalty.

The identity of your son will be kept confidential. Only the investigator will have access to the interview forms containing the boys' responses. The interview forms will be kept in a locked filing cabinet in her office in the Psychology Suite, located on the second floor of Siskin Hospital. They will be destroyed as soon as the results are entered into the computer. The computer data will have a code number, not a name, so your son's name will not be accessible. The completed written study will not contain any information to specifically identify your son. Only summary results will be reported, and there will be no reference to your son's individual responses. In addition, at the conclusion of the study, you will be mailed a summary of the results.

If you have questions concerning this study or your son's role in it, please contact me, Graham Parker, at my office:

Siskin Hospital for Physical Rehabilitation
One Siskin Plaza
Chattanooga, TN 37403
(423) 634-1669

I have read and understand the explanation of this study and my son's role in it. I understand that I may withdraw him from the study at any time without penalty. I agree to allow my child to participate.

_____ _____

Child's Name Date

_____

Parent Signature

APPENDIX D

INTERVIEW RECORDING FORM

AGE: _____

Please name a stressful situation related to muscular dystrophy:

_____

_____

_____

Please rate how stressful the situation is, using this scale:

1	2	3	4	5
A Little	Somewhat	A Lot	Very	Extremely

Please rate how often the stressful situation occurs, using this scale:

1	2	3	4	5
Rarely (Less than once per day)	Sometimes (At least once per month)	Often (At least once per week)	Frequently (Daily)	Constantly (More than once per day)

Please explain how you cope with the stressful situation:

_____

_____

_____

APPENDIX E

SUBJECT CONSENT FORM

I understand that Graham Parker is doing a study on boys with Duchenne Muscular Dystrophy. I agree to talk to Graham Parker about the stressful things I have to deal with, and about how I handle those stressful things. If I get upset, I can stop talking with her at any time, for any reason. I understand that I won't be punished in any way if I stop talking with her.

_____ _____

Name Date

_____

Signature

_____ _____

Witness Date

APPENDIX F

LETTER OF EXPLANATION TO MDA BOARD OF DIRECTORS

September 26, 1996

Muscular Dystrophy Association
Board Of Directors

I am completing my doctorate in School Psychology at the University of Tennessee. My dissertation research is focused on children with chronic illness. Specifically, I intend to interview males with Duchenne Muscular Dystrophy, asking them to identify perceived stressors and strategies used to cope with these stressors. Parents will be allowed to sit in during the interview if they wish to do so.

As principal investigator, I will be the only person to collect the data or to know the identity of the subjects. Interview responses will be transcribed and analyzed, but no names will be attached to the transcribed data. Furthermore, no individual subjects will be identified in the completed dissertation.

I have been an employee of the Siskin Hospital Psychological Services department for over five years. My patients have included both adults and children with a number of developmental and acquired disabilities. In addition, I have volunteered as a facilitator for the MDA Support Group in Chattanooga for over three years. Naturally, I have a strong interest in the area of coping with a disability. In particular, I hope to add to the sparse research literature concerning psychosocial aspects of neuromuscular disease.

Sincerely,

_____

A. Graham Parker

VITA

A. Graham Parker was born September 12, 1961 in Chattanooga, Tennessee. After attending elementary and secondary schools in Chattanooga, she attended the University of Tennessee at Chattanooga and obtained a Bachelor of Arts in Psychology in 1982. Ms. Parker obtained a Master of Science in School Psychology in 1985, and a Master of Science in Industrial/Organizational Psychology in 1989. She was licensed in Tennessee as a Psychological Examiner in 1988.

Ms. Parker currently resides and works in Chattanooga. She is a member of the Psychological Services Department at a hospital for physical rehabilitation. She also belongs to the Tennessee Association of School Psychologists and the National Association of School Psychologists.