Parental Stress in Mothers of Boys with Duchenne Muscular Dystrophy

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Objective To examine parental stress in mothers of boys with Duchenne muscular dystrophy (DMD). **Method** Stress and its predictors were examined in mothers of boys with DMD (n = 112). Comparisons were made with mothers of healthy children (n = 800), children with cerebral palsy (CP; n = 28), siblings of boys with DMD (n = 46), and longitudinally (n = 16). **Results** The presence of problem child behaviors consistently predicted maternal stress. Stress related to child behavior was higher in the DMD versus the normative group. No differences in stress were found in the DMD versus CP groups. Stress related to boys with DMD versus siblings was not significantly different. Over time, maternal stress related to child variables diminished. **Conclusion** Stress in mothers of boys with DMD is elevated, possibly due to increased problem behaviors, particularly in social interactions, rather than due to the physical demands of the disease alone.

Key words Duchenne muscular dystrophy; parental stress; family functioning.

Duchenne muscular dystrophy (DMD), the most severe of the many forms of muscular dystrophy, occurs with an incidence of about 30 per 100,000 live born males (Mendel, Griggs, & Ptacek, 1998). It is an X-linked developmental disorder that causes progressive muscle weakness. The gene involved normally codes for a protein called dystrophin, which localizes to muscle, and dystrophin-like products, which localize to the central nervous system. In children with DMD, these products are missing due to a deletion in the gene. As the genetic deletion that causes DMD is located on the x chromosome, boys have the more severe, fatal form of the disease. Among boys with DMD, a wide range of intellectual functioning is observed. While most boys with DMD function within the normal limits of intelligence, about 19% of the DMD population is mentally retarded (as compared with 2–3% in the general population) and the mean IQ of those affected is 85. The extent of cognitive involvement is variable across individuals but is not associated with physical severity and does not appear to be progressive.

The course of the physical impairment is progressive. Initially, boys with DMD appear to be developing normally. At ages 2 to 3, slight motoric impairments appear, and the child is perceived as being somewhat clumsy. Difficulties such as falling or having trouble climbing stairs become increasingly apparent, and diagnosis usually occurs around age 5. As muscles continue to weaken, the boys begin to walk stiffly, with abdomens protruding to compensate for weakening leg muscles, and increased physical support is required. By age 12, boys with DMD usually require a wheelchair. Medical management of DMD involves protracted corticosteroid therapy in order to slow the progression of muscle weakness, though in many boys with DMD these medications may also result in weight gain, immunosuppression, and glucose intolerance, among other undesired effects. Daily physical therapy, a set of exercises overseen and performed by the primary caregiver, is also prescribed to maintain optimal muscle function. During childhood, surgical intervention is likely, to elongate tendons in the legs. After prolonged wheelchair use, boys with DMD may also undergo surgery to correct spine curvature that negatively impacts pulmonary function. Typically, efforts are focused upon improving quality of life and delaying the course of the disease, as there is currently

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no cure for DMD. Death generally occurs by the time the boy reaches his early 20s, usually due to respiratory or cardiac failure resulting from extreme muscle weakness.

DMD, which is both chronic and terminal, may be characterized as a "complex chronic condition" in that it involves specialized and time-consuming care, even when the terminal phase lies years in the future (Gravelle, 1997). As such, it may be expected to have effects on the family similar to both chronic and terminal illnesses. DMD poses stressors in terms of daily care requirements, such as negotiating wheelchair transportation and meeting recommended physical therapy requirements. In addition, as with other complex chronic illnesses, many psychological adjustments become necessary, such as facing separation and loss, experiencing and expressing emotions (including anger, guilt, sadness, loss of control, resentment of increased demands), and changing values, expectations, roles, and responsibilities (Copeland, 1988).

It may be hypothesized that the physical, emotional, and logistical issues associated with DMD and other complex chronic illnesses would lead to impaired family functioning, particularly with regard to parental stress. However, the existing research on the level of parental stress in families of children with chronic illness and developmental disabilities is equivocal. Some studies indicate that parents of ill children experience stress levels comparable to those of parents of healthy children, for example in parents of children with spina bifida (Spaulding & Morgan, 1986), cystic fibrosis (Walker, Ford, & Donald, 1987), and kidney disease (Soliday, Kool, & Lande, 2000). Other studies found elevated levels of reported stress in parents of children with cystic fibrosis and congenital heart disease (Goldberg, Morris, Simmons, Fowler, & Levison, 1990), spina bifida (Holmbeck et al., 1997), Down syndrome (Roach, Orsmond, & Barratt, 1999), juvenile rheumatoid arthritis (Manuel, 2001), renal disease, and Duchenne muscular dystrophy (Holroyd & Guthrie, 1986). In particular, parents of boys with DMD reported higher levels of stress than parents of children with cystic fibrosis or renal disease, though all groups of children with a chronic disease reported more stress than parents of healthy controls, and in patterns consistent with the care requirements of their child's disease (Holroyd & Guthrie, 1986).

Prior research has examined a variety of factors related to stress in parents of chronically ill children. In a study of children with developmental disabilities, family variables, particularly family resources, were found to predict stress, and impairment in child social skills had a stronger relationship to parental stress than any other aspect of functioning (i.e., motor, cognitive, communication, adaptive behavior; Smith, Oliver, & Innocenti, 2001). In children born at low birth weight, developmental status and parent-child relationship have been found to contribute significantly to parenting stress (Robson, 1997). In a study of parents of children with cerebral palsy, the strongest predictor of maternal stress was child behavior problems (Mobarak, Khan, Munir, Zaman, & McConachie, 2000). These studies suggest that interpersonal or behavioral variables are a source of parental stress, beyond aspects of daily care or medical concerns. To date, no such examination of factors of stress has been reported with a large sample of parents of children with DMD.

The present study sought to further elucidate aspects of parental stress in families of boys with DMD. Generally, prior studies of stress in parents of children with a chronic illness indicated that mothers of disabled and chronically ill children report greater parental stress and overall distress (Beckman, 1993; Manuel, 2001; Pelchat et al., 1999; Saviolo-Negrin et al., 1999). We therefore focused upon stress levels reported by mothers because, as primary caregiver, they may be at particular risk for increased stress.

Evaluation of maternal stress in DMD would benefit from further attention to several issues. Firstly, stress in mothers of boys with DMD has rarely been examined, with the exception of a study of a modest sample (n = 16) compared with other illness groups (Holroyd & Guthrie, 1986). A study of stress in a larger sample of mothers of boys with DMD may provide a more complete depiction of stress experienced in this population. In particular, examination of particular variables associated with higher maternal stress is warranted. Secondly, comparison of stress in mothers of boys with DMD with that in a group of mothers of children with a chronic, nonterminal illness would elucidate the unique experience of mothers of boys with DMD. We therefore sought to examine maternal stress in mothers of boys with DMD as compared with mothers of children with cerebral palsy, a nonprogressive, nonterminal illness that, like DMD, involves motor impairment. A third aim in the present study was to understand maternal stress contextually in families of boys with DMD. We therefore compared stress related to child characteristics in boys with DMD with that of their siblings. Lastly, we sought to investigate maternal stress over time, as this progressive disease worsens. We hypothesized that as the disease worsens, maternal stress will increase.

Method

Participants

Part 1. Boys with DMD. Data for 127 boys with DMD and their mothers were initially included in these analyses. All participants were enrolled in an ongoing study of cog-

nitive skills in boys with DMD, who were recruited through the Duchenne Muscular Dystrophy Clinical Trials study as well as by private physicians associated with Muscular Dystrophy Association clinics (Columbia Presbyterian Medical Center and Albert Einstein Medical Center, New York, NY; Scottish Rite Children's Medical Center, Atlanta, GA; Newington Children's Hospital, Newington, CT) and through announcements and mailings by the Muscular Dystrophy Association and the Parent Project. All children with DMD were male, between 4 and 17 years of age, in otherwise good general health, spoke English, and were willing to participate. Diagnosis of DMD was based on clinical onset of progressive weakness before 5 years of age, elevated serum creatine kinase levels, and either molecular assessment of the mutation in the DMD gene or a muscle biopsy that was deficient in dystrophin and compatible with DMD. In six families, more than one boy met criteria for inclusion; for these families, no more than two eligible sons with DMD were included. At the time of testing, 58 boys (45.7%) were in wheelchairs and 69 boys (54.3%) were still walking. Findings on cognitive, behavioral, and psychosocial aspects of these children have been reported previously (Hinton, De Vivo, Nereo, Goldstein, & Stern, 2000; Hinton, De Vivo, Nereo, Goldstein, & Stern, 2001; Hinton & Fee, 2000; Nereo & Hinton, 2003).

DMD vs. CP. In the second set of analyses, data for the DMD group were compared with data for a group of children diagnosed with cerebral palsy (CP) and their mothers. The 42 children with CP were between 6 and 8 years old at the time of participation. These children were enrolled in an ongoing study examining the school-age cognitive outcome of children born at very low birth weight (i.e., <1500 g; Arad et al., 2002). They were drawn from a sample of children enrolled at birth in the DEN (Developmental Epidemiology Network) cohort (Leviton et al., 1997). Each child received a standardized and reliable neurological evaluation (Chiriboga, Kairam, & Kline, 1993) from an experienced pediatric neurologist to diagnose whether the child had CP. The CP comparison group was selected because effects of the physical disability were somewhat controlled, as both diseases are associated with motor impairment. Twenty-two children with CP were male and 20 were female. Only one child was included from each participating family.

Part 3. Boys with DMD vs. Siblings. Where possible, one healthy sibling without DMD was also recruited for each participant with DMD. Inclusion criteria were: age 4 to 17 years old; age within 5 years of the boy with DMD; good general health; English speaking; and willingness to participate. Where more than one sibling participant was available, preference was given first to male gender and then to closeness of age. A total of 60 siblings met these criteria, participated, and had complete data for the present analyses. All sibling pairs were from separate families. Thirty-one siblings were male and 29 were female. Thirty-seven siblings were older than their brother with DMD (21 male and 16 female) and 22 siblings were younger (10 male and 12 female). One female sibling was a fraternal twin.

Part 4. Longitudinal Analysis of Boys with DMD. Twenty-eight families from the larger DMD sample have participated in an ongoing longitudinal study, where follow-up data are collected every 2 years. In addition to the initial inclusion criteria, follow-up study inclusion criteria are: age 10 years or younger at first assessment, geographical accessibility, and willingness to continue participation. Data from three time points have been collected for 19 participants thus far and were included for analysis to determine whether there has been any change in parental stress over time.

Mother and child participant characteristics for each set of analyses are summarized in Table I.

Procedure

Procedures for the collection of data included in the present study were approved by the home institution's institutional review board. Prior to data collection, mothers provided written informed consent and children provided verbal assent. All families were seen either in an office in the medical center in their area or in a quiet room in their homes when travel was inconvenient. Parents completed questionnaires while their children were being tested and they mailed them to the investigator in cases where additional time was needed.

Measures

Maternal Stress. Mothers of all participants completed the Parenting Stress Index–Short Form (PSI-SF; Abidin, 1995). This self-report measure was developed from the perspective that the stress a parent experiences is a function of characteristics of both the child and the parent, as well as their unique style of interaction. Accordingly, its 36 items compose the following three subscales: parental distress (emotional distress in the parenting role), parent-child dysfunctional interaction (problematic parent-child interactions), and difficult child (problematic child behavior or demands). A total raw score greater than 90 indicates elevated stress, according to the measure's authors, as it falls above the 90th percentile in the normative group. In addition, a defensive responding scale is computed based upon items commonly endorsed by all parents, in or-

Table I. Child and Mother Participant Characteristics

Analyses	Child Participants	% Male	% White	% in Wheelchair	Child Age, years *M*±*SD*	PPVT SS *M*±*SD*	PSI-SF Total *M*±*SD*	Mother Age, years *M*±*SD*	Mother Modal Education
DMD	127 boys with DMD	100.0	87.4	45.7	9.61±2.87	100.01±22.08	81.22±20.18	38.16±5.49	HSG
DMD vs. CP	112 boys with DMD	100.0	86.6	45.5	9.43±2.81	99.04±22.58	81.28±20.18	37.75±5.43	HSG
	28 children with CP	52.4	53.6	0.0	6.43±0.63	92.93±17.51	65.49±20.83	36.75±5.61	HSG
DMD vs. siblings	46 boys with DMD	100.0	90.0	32.0	8.85±2.41	99.47±20.53	87.33±19.59	37.90±5.30	HSG
	46 healthy siblings	43.5	90.0	0.0	10.07±3.2	109.94±17.39	79.93±17.87		
Longitudinal DMD	16 boys with DMD (Time 1)	100.0	81.3	18.8	7.87±1.41	102.38±17.07	86.88±12.31	34.21±4.63	HSG
	16 boys with DMD (Time 2)			68.8	9.94±1.44		81.13±21.86		
	16 boys with DMD (Time 3)			87.5	12.81±1.56		80.81±12.91		

PPVT SS = Peabody Picture Vocabulary Test Standard Score. PSI-SF Total = total score on the Parenting Stress Index–Short Form. DMD = Duchenne muscular dystrophy. CP = cerebral palsy. HSG = high school graduate.
For first DMD analyses, characteristics of all participants are described. For DMD vs. CP, DMD vs. siblings, and longitudinal DMD analyses, only characteristics of the nondefensive responders included in analyses are described.

der to determine whether the respondent's answers shall be considered valid. A score lower than 11 on the defensive responding scale is considered "defensive" and the PSI-SF protocol's validity is therefore questionable. The PSI-SF was developed based upon the full-length Parenting Stress Index, using factor analysis of the scale's original 100 items to determine the three primary subscales. Test-retest reliability of the PSI-SF total score and the subscales ranges from .68 to .85. Internal consistency (alpha) for the short form total score and subscales ranges from .80 to .91 (Abidin, 1990).

Estimated Verbal IQ. Boys with DMD and their siblings completed the Peabody Picture Vocabulary Test–Revised (PPVT-R; Dunn & Dunn, 1981), an individually administered test of receptive vocabulary that yields age-referenced standard scores (M = 100, SD = 15). PPVT-R scores have been shown to have a correlation of .70 with the Wechsler Intelligence Scales for Children–Revised, Full Scale IQ Score; with the Verbal IQ Score the correlation is .69 (Dunn & Dunn, 1997). Children with CP, as participants of a different protocol, were administered the PPVT–Third Edition (Dunn & Dunn, 1997). This test presents the same task as the PPVT-R, and also yields age-referenced standard scores.

Child Behavior Problems. Mothers completed the Child Behavior Checklist (CBCL; Achenbach, 1991). The CBCL consists of 108 problem behaviors rated by parents with regard to frequency in their child (0 = *never* to 2 = *very much*). The CBCL yields T scores (M = 50, SD = 10) for eight subscales of behavior and personality, factors of internalization and externalization, and a total score. These T scores are derived from a comparison of the individual's score with the appropriate normative group, based upon gender and age. Test-retest reliability for the total problem behavior score is .95 (Achenbach, 1991).

Data Analysis

For the different groups, child gender and ethnicity group percentages, mean age, PPVT standard score, and PSI-SF total score were calculated. Mean mother age and education were also determined. Child and mother participant characteristics are summarized in Table I. Descriptives are for those participants included in primary analyses for each part of the investigation; prior to most analyses, those participants with scores less than 11 on the defensive responding PSI-SF subscale were excluded as recommended by the measure's author (as further described in the Results section).

Part 1. Boys with DMD. To compare stress level in mothers of boys with DMD with that of the normative sample reported by the scale's authors (n = 800 parents of children at well-child clinic visits), one-sample t tests compared mean subscale scores with normative means, with alpha set at .01. For these first analyses, all participants were included, as potential defensive responders were included in the normative sample as well. For the next set of analyses, participants with defensive responding subscale scores less than 11 were excluded. Using independent t tests, excluded (defensive) responders were compared with included (nondefensive) responders with regard to child age, estimated verbal IQ, and mother age, to ensure similarity of groups. Families with two boys with DMD (6 families) were compared with those with one boy with DMD (100 families) on total PSI-SF scores to evaluate whether a significant difference in stress existed.

Linear regression analyses were conducted on data

from nondefensive participants to determine the relative contributions of other variables to parental stress. For these analyses the following variables were entered into the equation simultaneously: child age, wheelchair use, estimated verbal IQ, and total behavior problems; mother age and highest level of education; and number of parental figures and siblings in the home, with each of the three PSI-SF subscales as the dependent variable. Alpha was set at .05.

A particular behavioral profile among boys with DMD has been reported by this group elsewhere (Hinton & Fee, 2000), and problem child behaviors, as measured by the CBCL, were found to significantly contribute to parental stress across three subscales in Part 1 of our research. Therefore, further analyses were conducted to examine levels of problem behaviors in stressed versus nonstressed mothers. All mothers were assigned to one of these subgroups (stressed or nonstressed) based upon whether the total PSI-SF score fell above or below the 90th percentile (raw score ≥ 91), the clinical cut score recommended by the scale's author (Abidin, 1995). To establish that the two groups (stressed vs. nonstressed) did not differ on other variables likely to contribute to maternal stress, *t* tests were conducted to compare child age, child estimated verbal IQ, and mother's age, and a chi-square analysis was conducted to compare the boys' use of a wheelchair. Alpha was set at.05 to protect against a type II error. CBCL subscale *T* scores (withdrawn, somatic complaints, anxious/depressed, social problems, thought problems, attention problems, delinquent behavior, aggressive behavior) were compared in the two subgroups using a MANOVA (multivariate general linear model). Alpha was set at .01.

Part 2. DMD vs. CP. As in the DMD group, participants in the CP group with defensive responding scores less than 11 were excluded. To ensure similarity of groups, excluded (defensive) responders were compared with included (nondefensive) responders with regard to child age, estimated verbal IQ, and mother age. Independent *t* tests were then calculated for child age, PPVT standard scores, and mother age to examine similarity of CP and DMD groups on these variables. Next, an ANCOVA (analysis of covariance) was conducted comparing CP and DMD group mean scores on each of the three PSI-SF subscales, with age and gender entered as covariates. Alpha was set at .01.

Part 3. Boys with DMD vs. Siblings. Participants with a defensive responding score less than 11 on the PSI-SF for the boy with DMD or his sibling, or both, were excluded. Excluded (defensive) responders were compared with included (nondefensive) responders to ensure similarity of groups. Paired *t* tests were calculated for age and PPVT standard scores to examine similarity of the boys with DMD and their siblings. Socioeconomic and background variables were controlled for because participants were from the same family and household. Paired *t* tests were then conducted with regard to the PSI-SF parent-child dysfunctional interaction and difficult child subscales. (The parental distress subscale, which examines parental characteristics, was not examined for these analyses, as the respondent was the same person). Alpha was set at .01.

Part 4. Longitudinal Analysis of Boys with DMD. Maternal stress was examined across three time points, approximately 2 years apart (Time 1 to Time 2, $M = 25.81$ months; Time 2 to Time 3, $M = 28.55$ months), using a general linear model (GLM) doubly multivariate repeated measures design. Participants with defensive responding scores less than 11 at one, two, or all three time points were excluded. The within-subject factor of time consisted of three levels (the three time points at which data were collected). At each of these levels, the measures were the three subscales of the PSI-SF. Alpha was set at .05. For subscales for which significant differences were detected over time, contrasts were utilized in the equation to establish where the differences lay (Time 1–2, Time 2–3, and/or Time 1–3).

Results

Part 1. Boys with DMD

To determine how parental stress in parents of boys with DMD compared with a normative sample, one-sample *t* tests were conducted comparing the PSI-SF subscale scores of all 127 mothers of boys with DMD to norm mean subscale scores. The mean DMD parental distress score (26.77) was not significantly different from the normative sample's mean (normative $M = 26.4$; $t = 0.56$, $df = 125$, *ns*). Mean DMD parent-child dysfunctional interaction score ($M = 23.98$) was significantly greater than that of the normative sample (normative $M = 18.7$; $t = 8.51$, $df = 125$, $p = .000$). Likewise, mean DMD difficult child score ($M = 30.64$) was also significantly greater than that of the normative sample (normative $M = 26$; $t = 5.65$, $df = 125$, $p = .000$). These data are summarized in Table II. Further, 33% of the mothers of boys with DMD ($n = 42$) had PSI-SF total scores greater than or equal to the 90th percentile (the clinical cut score), as compared with 10% in the normative sample (Abidin, 1995).

Next, defensive responders ($n = 15$) were excluded, resulting in a sample size of 112. Comparisons in defensive versus nondefensive responders of child age, estimated IQ, and mother age ($t = 1.86$, 1.49, and 1.98, respectively; $df = 124$, *ns*) and wheelchair use ($X^2 = .04$, *ns*) did not indicate significant differences across groups. Comparison of total maternal stress in families with two boys with DMD

Table II. Parental Stress in Mothers of Boys with DMD vs. Normative Sample

PSI-SF Subscale	DMD ($n = 127$) $M \pm SD$	Norm ($n = 800$) $M \pm SD$	t Value	df
Parental distress	26.77±7.37	26.4±7.2	0.56*	125
Parent-child dysfunctional interaction	23.98±6.97	18.7±4.8	8.51**	125
Difficult child	30.64±9.23	26.0±6.7	5.65**	125

PSI-SF = Parental Stress Index–Short Form. DMD = Duchenne muscular dystrophy.
*$p > .05$.
**$p = .000$.

and in families with one boy with DMD revealed no significant difference ($t = -0.78$, $df = 108$, *ns*); all families with nondefensive protocols were therefore included in remaining analyses.

Linear regression analyses were then conducted to examine the effects of the following variables on each aspect of maternal stress in the DMD group: child age, estimated verbal IQ, wheelchair use, total child behavior problems, mother age and highest level of education, and number of parental figures and siblings in the home. Significant contributors to the parental distress subscale were estimated verbal IQ and child behavior problems ($\beta = -.24$ and .36; $p = .03$ and .001, respectively). As a group, the independent variables included in the regression equation significantly predicted 21% of the variance for parental distress, $F(8, 82) = 2.76$; $p = .009$. With regard to parent-child dysfunctional interaction, only child behavior problems ($\beta = .53$; $p = .000$) was a significant contributor, though wheelchair use approached significance ($\beta = -.22$; $p = .06$). The set of independent variables included in the regression equation for parent-child dysfunctional interaction significantly predicted 38% of the variance, $F(8, 81) = 6.26$; $p = .000$. With regard to the difficult child subscale, wheelchair use and child behavior problems were significant ($\beta = -.22$ and .65; $p = .03$ and .000, respectively). Together, the independent variables in this regression equation significantly predicted 55% of the variance for the difficult child scores, $F(8, 81) = 12.11$; $p = .000$. These data are summarized in Table III.

Results of analyses conducted to compare variables likely to covary with levels of maternal stress indicated no differences between mothers who reported high stress (PSI-SF total score ≥90th percentile) versus those who reported low stress (PSI-SF total score <90th percentile). These included child age ($t = 0.66$, $df = 110$, *ns*), child estimated verbal IQ ($t = 1.11$, $df = 110$, *ns*), mother age ($t = -0.44$, $df = 98$, *ns*), and child wheelchair use ($X^2 = .43$, *ns*). Comparisons of the eight CBCL subscales were then conducted in these subgroups of stressed versus nonstressed mothers. The omnibus F test indicated a significant difference between groups, $F(8, 99) = 4.28$, $p = .000$. Tests of between-subjects effects revealed that mothers reporting higher stress levels also reported significantly higher subscale scores regarding each aspect of their child's behavior. The nonstressed subgroup means on all CBCL subscales were less than 67, the clinical cut score for the measure recommended by the author. The stressed subgroup means on all CBCL subscales were also less than 67, with the exception of the mean social problems subscale score, which was 66.6. These results are summarized in Table IV.

Part 2. DMD vs. CP

Defensive responders were excluded from both groups, resulting in 112 participants with DMD and 28 participants with CP. In the CP group, the excluded (defensive) responders were compared with included (nondefensive) responders with regard to child age, estimated verbal IQ, gender, and mother age; these tests indicated no significant differences (child age, PPVT, and mother age: $t = -0.34$, 1.32, and 0.50, respectively, $df = 40$, *ns;* gender $X^2 = .76$, *ns*).

To examine the similarity between the DMD and CP groups, t tests comparing child age and estimated verbal IQ were conducted. As anticipated, the children with CP were significantly younger than the boys with DMD ($t = 10.28$, $df = 138$, $p = .000$). Estimated verbal IQ and mother age did not differ significantly between groups ($t = 1.33$ and .86, respectively, $df = 138$, *ns*).

ANCOVAs comparing DMD and CP groups' PSI-SF subscale scores were conducted, with age and gender entered as covariates. None of these group scores differed significantly at the alpha level established (.01), though the difficult child subscale comparison approached significance, reflecting the DMD group's higher mean score, $F(3, 1) = 2.91$, $p = .04$. These data are summarized in Table V.

Part 3. Boys with DMD vs. Siblings

After defensive responders were excluded, 46 sibling pairs remained. The excluded boys with DMD were compared with included boys with DMD with regard to child age and estimated verbal IQ; these tests indicated no significant differences (age and PPVT: $t = 0.58$ and 1.24, respectively, *ns*).

Next, the 46 boys with DMD were compared with their siblings using paired t tests. No significant differ-

Table III. Summary of Regression Analyses for Variables Predicting Parental Stress in Mothers of Boys with DMD

PSI-SF Subscale	Variables	B	SE	Beta	*t*
Equation 1: Parental distress	Child age	0.08	0.30	–.04	–0.27*
	Wheelchair use	0.51	1.61	.04	0.32*
	PPVT	–0.07	0.03	–.24	–2.23***
	CBCL	0.19	0.06	.36	3.40****
	Mother age	0.21	0.14	.18	1.55*
	Mother education	0.52	0.65	.10	0.80*
	No. of parents	0.92	2.67	.04	0.35*
	No. of children	–0.71	0.60	–.12	–1.19*
Equation 2: Parent-child dysfunctional interaction	Child age	0.38	0.28	.16	1.34*
	Wheelchair use	–2.94	1.56	–.22	–1.88**
	PPVT	–0.04	0.03	–.15	–1.55*
	CBCL	0.30	0.05	.53	5.61*****
	Mother age	0.06	0.13	.05	0.47*
	Mother education	0.18	0.61	.03	0.29*
	No. of parents	–0.75	2.51	–.03	–0.30*
	No. of children	–0.01	0.56	–.002	–0.02*
Equation 3: Difficult child	Child age	0.57	0.31	.17	1.81**
	Wheelchair use	–3.78	1.70	–.22	–2.23***
	PPVT	–0.04	0.03	–.11	–1.27*
	CBCL	0.49	0.06	.65	8.10*****
	Mother age	–0.04	0.15	–.02	–0.26*
	Mother education	0.44	0.68	.06	0.64*
	No. of parents	–2.39	2.82	–.07	–0.85*
	No. of children	–0.24	0.64	–.03	–0.38*

DMD = Duchenne muscular dystrophy. PSI-SF = Parenting Stress Index–Short Form. PPVT = Peabody Picture Vocabulary Test Standard Score. CBCL = Child Behavior Checklist total *T* score. No. of parents = number of parents in the home. No. of children = number of children in the home. For Equation 1, R^2 = .212; F = 2.76; p = .009; Equation 2, R^2 = 38; F = 6.26; p = .000; Equation 3, R^2 = .545; F = 12.11; p = .000.

*p > .05.

**p < .10.

***p < .05.

****p = .001.

*****p = .000.

Table IV. Problem Child Behaviors Reported by Stressed versus Nonstressed Mothers of Boys with DMD

CBCL Factor/Subscale	Nonstressed mothers (n = 68) $M \pm SD$	Stressed mothers (n = 40) $M \pm SD$	*F*	R^2
Withdrawn	56.94±7.12	62.03±8.15	11.52**	.098
Somatic complaints	55.12±6.52	60.10±8.16	12.17**	.103
Anxious/depressed	55.02±6.12	63.08±9.66	28.23***	.210
Social problems	59.71±8.55	66.60±8.59	16.32***	.133
Thought problems	56.04±6.78	60.50±8.90	8.59*	.075
Attention problems	57.78±8.12	63.30±8.41	11.34**	.097
Delinquent behavior	52.41±4.69	55.05±5.05	7.52*	.066
Aggressive behavior	52.84±5.58	59.53±9.44	21.49***	.169

DMD = Duchenne muscular dystrophy. CBCL = Child Behavior Checklist. Mean scores are on a *T* scale (M = 50, SD = 10).

*p < .01.

**p = .001.

***p = .000.

Table V. Maternal Stress in DMD versus CP Groups

PSI-SF Subscale	DMD ($n = 112$) $M \pm SD$	CP ($n = 28$) $M \pm SD$	F	df
Parental distress	28.15±6.55	25.64±7.57	1.62*	3, 1
Parent-child dysfunctional interaction	24.62±6.93	22.67±7.77	0.86*	3, 1
Difficult child	31.71±8.93	25.96±10.1	32.91**	3, 1

DMD = Duchenne muscular dystrophy. CP = cerebral palsy. PSI-SF = Parental Stress Index–Short Form. Gender and age were covariates in the analysis. Findings are *ns* at .01 level.

*$p > .05$.

**$p < .05$.

ences between groups with regard to age were found, though siblings were slightly older ($t = -2.57$, *ns*) and had a significantly higher estimated verbal IQ ($t = -3.68$, $p = .001$). In analyses of maternal stress, neither the parent-child dysfunctional interaction nor the difficult child subscales of the PSI-SF were found to differ significantly between groups ($t = 1.21$ and 2.20; $p > .05$ and $p = .034$, respectively), though the difficult child subscale comparison approached significance at the alpha level set (.01). These data are summarized in Table VI.

Part 4. Longitudinal Analyses of Boys with DMD

Three participants were excluded for defensive responding subscale scores less than 11 at one, two, or all three time points, resulting in 16 participants. A GLM doubly multivariate repeated measures design was used to analyze PSI-SF subscale scores over time. Neither parental distress nor parent-child dysfunctional interaction subscale scores varied significantly over time, $F(32, 2) = .972$ and .592, respectively, *ns*. However, scores on the difficult child subscale did significantly change, $F(32, 2) = 4.26$; $p = .024$. Specifically, mean scores of the difficult child subscale decreased over time. These data are summarized in Table VII. Contrasts in the GLM equation for the difficult child subscale revealed that the significant decrease in stress occurred between Time 1 and Time 3, $F(15, 1) = 9.54$, $p = .007$. No significant difference was detected between Times 1 and 2, or between Times 2 and 3, $F(15, 1) = 2.37$ and 1.87, respectively, *ns*.

Discussion

The present study sought an in-depth investigation of parental stress among mothers of boys with DMD. To that end, an examination of stress in mothers of boys with DMD was conducted (a) as compared with a normative sample, (b) in relation to demographic, behavioral, and medical variables, (c) in comparison with another sample of mothers of children with a nonprogressive, nonterminal illness involving motor impairment (CP), (d) in comparison with stress related to siblings of boys with DMD, and (e) across three data collection points, each approximately 2 years apart.

As compared with the normative sample, mothers of boys with DMD did not report greater parental distress (stress related primarily to parent variables), yet did report greater stress related to their interactions with their sons and to their sons' behavior (parent-child and child variables). These results suggest that the mothers' interactions with their sons and the behaviors of these boys with DMD are more stressful than those of the normative group, composed of parents recruited from well-child clinics. Similarly, 33% of the mothers of boys with DMD reported stress greater than the clinical cut score established by the PSI-SF's author, as compared with the normative group's 10%. These findings indicate that, indeed, mothers of boys with DMD experience greater stress than a healthy normative group and that in particular the stress is related to their children, in that their children's behaviors and interactions with them are more stressful than is the experience of raising healthy children for other mothers. Research from this group (Hinton & Fee, 2000) indicates that social skills are impaired in boys with DMD and suggests that this impairment is a part of the phenotype, rather than a reaction to the illness. Together, these findings suggest that decreased social awareness and competency in boys with DMD present particular difficulty for their caregivers. Beyond potential physical, financial, emo-

Table VI. Maternal Stress Related to Boys with DMD versus Their Siblings

PSI-SF Subscale	DMD ($n = 46$) $M \pm SD$	Siblings ($n = 46$) $M \pm SD$	t
Parent-child dysfunctional interaction	25.39±8.20	23.77±7.81	1.21*
Difficult child	31.88±8.91	27.98±8.02	2.20**

DMD = Duchenne muscular dystrophy. PSI-SF = Parental Stress Index–Short Form. Findings are *ns* at .01 level.

*$p > .05$.

**$p < .05$.

Table VII. Repeated Measures Analysis of Parental Stress in Mothers of Boys with DMD

PSI-SF subscale	Time, $M \pm SD$	Univariate $F(2, 32)$
Parental distress	1, 28.63±6.64	0.97*
	2, 30.50±5.89	
	3, 29.06±4.75	
Parent-child dysfunctional interaction	1, 25.00±4.90	0.59*
	2, 23.88±5.95	
	3, 23.56±5.98	
Difficult child	1, 33.25±4.73	4.26**
	2, 31.00±7.56	
	3, 28.18±6.06	

DMD = Duchenne muscular dystrophy. PSI-SF = Parental Stress Index–Short Form.
*$p > .05$.
**$p < .05$.

tional, or logistical strains presented by DMD, social problems of boys with DMD (such as not getting along with others, being teased, acting young for one's age, being clingy, or not being liked by others, all items from the CBCL social problems subscale) may be problem behaviors that make mother-child interactions difficult.

In further support of the relationship between poor social skills in boys with DMD and maternal stress, we found that predictors of maternal stress were variables related to the child. In particular, for each of the three domains of maternal stress (parental distress, parent-child dysfunctional interactions, difficult child), child problem behaviors were a significant contributing factor. Further, the child's low estimated verbal IQ contributed significantly to the parental distress subscale, and wheelchair use contributed significantly to the difficult child subscale. Perhaps surprisingly, other family variables expected to contribute to maternal stress level (number of parents and number of children in the home) were not found to make significant contributions. These analyses suggest that maternal stress hinges largely on the child's behavioral and intellectual functioning as well as practical aspects of caring for him, rather than on maternal variables tested.

Further, mothers reporting higher stress reported significantly greater problem behaviors on the eight subscales of the CBCL (withdrawn, somatic complaints, anxious/depressed, social problems, thought problems, attention problems, delinquent behavior, aggressive behavior). Overall, then, greater problem behaviors were found in the boys of the more highly stressed mothers. It may be that these characteristics mutually affect one another: Children of stressed mothers may exhibit worsening behavior; worsening behavior may increase maternal stress. Increased support or services targeting one or both of these issues may help to improve both mother and child functioning and inhibit this reciprocal interaction.

While highly stressed mothers reported generally increased CBCL factor and subscale scores, across subgroups (high vs. low stress) mean scores for each of the eight CBCL subscales were below the clinical cut score, with one notable exception. The subgroups of mothers reporting high stress also had a mean CBCL social problems subscale *T* score of 67 (the clinical cut score), suggesting that the level of impairment of social skills was at a clinically significant level for these boys with DMD. In fact, post hoc analyses indicate that this area of the child's functioning is a significant factor in the parent-child and child characteristics of maternal stress. Mothers of the subgroup of boys with DMD with a CBCL social problems *T* score greater than or equal to the clinical cut score reported significantly higher parent-child dysfunctional interaction and difficult child subscale scores than mothers reporting fewer social problems in their boys ($t = -3.47$ and -4.28; $p = .002$ and .000, respectively). The social impairment associated with DMD, then, has repercussions for family functioning, as well as for the boy's own adjustment, as previously shown. Further, these findings are similar to those in an investigation of parental stress in families of children with developmental disabilities, which showed that personal or social skills predicted stress, while adaptive behavior, cognition, communication, and motor skills did not (Floyd & Gallagher, 1997). The present findings suggest that an intervention for boys with DMD targeting social functioning and interactions with caregivers may contribute to decreasing parental stress and improving family functioning.

Mothers of boys with DMD did not report significantly greater stress as compared with mothers of children with CP. However, stress related to child variables was slightly elevated in the DMD group, though not at a significant level ($p = .04$, greater than alpha of .01 established for the analysis). Taken together, the data suggest that mothers of boys with DMD, while experiencing greater

overall stress than mothers of healthy children, do not necessarily experience greater levels of stress than mothers of children with CP. Further, as with comparisons of the DMD and the normative groups, maternal distress in the DMD group was not significantly different when compared with the CP group. These preliminary findings suggest that to the extent that maternal stress in these groups does vary, it may be a function of child variables. Future investigation into maternal stress across pediatric medical illnesses would help elucidate this issue, as the present data are inconclusive. In particular, such studies should examine the role of behavioral variables that may differ across disease type, in order to guide clinical interventions which may help minimize maternal stress and improve family functioning.

Maternal stress related to parent-child interactions and to difficult child behaviors in boys with DMD was also compared with these variables related to the siblings of the boys with DMD. In these parent-child and child characteristics, mothers reported no difference in boys with DMD as compared with siblings. That is, mothers did not experience greater stress in interacting with their sons with DMD, or perceive boys with DMD to be more difficult to raise than healthy siblings. This subsample of mothers of boys with DMD ($n = 46$) also reported greater stress in boys with DMD as compared with a healthy normative sample (DMD total $M = 86.33$; normative total $M = 71.00$; $t = 5.22$, $df = 44$, $p = .000$). It may be that the experience of having a chronically ill child has more global effects, in that the additional stress leads to lower overall tolerance in these mothers, and that parenting skills and coping are generally poorer as a result.

A post hoc one-sample t test to analyze PSI-SF subscale scores of siblings revealed that parental distress and difficult child characteristics are no different than in the measure's normative group ($t = 1.41$ and 1.60, respectively, *ns*), though parent-child dysfunctional interactions were significantly higher ($t = 4.31$, $p = .000$). While the DMD group was all male, the sibling group included both males and females. Post hoc paired t tests comparing boys with DMD and male siblings only ($n = 20$) revealed no difference in maternal stress with regard to parent-child interactions and child variables ($t = -0.62$ and 0.57, $df = 18$ and 17, respectively, *ns*), supporting the conclusion that maternal stress related to DMD and sibling groups do not differ significantly regardless of the effects of gender. Taken together, these data suggest that mothers of boys with DMD do not report greater difficulty in raising them as compared with their siblings in part because parenting both children creates stress, rather than because the stress level is not of consequence. These findings are consistent with the comparisons conducted to examine maternal stress in families with one versus more than one boy with DMD, which revealed no significant differences. Increased stress may therefore be experienced more broadly, rather than in increments related specifically to the boy with DMD or to the number of boys with DMD in the family.

Finally, stress was examined over three time points, each approximately 2 years apart. Over this span, stress with regard to parent or parent-child characteristics did not change significantly. However, stress related to child characteristics alone (the difficult child PSI-SF subscale) decreased significantly over this period. This finding was especially notable, as it was expected that stress would increase over time as the disease progressed and care demands increased. At the start of data collection, 81.2% of boys with DMD were walking, but by Time 3, 87.5% were in wheelchairs. Despite the worsening symptoms in their child, mothers were perhaps learning to cope and adjust to offset the negative effects. Our data may reflect the previously reported findings that stress is generally greatest at the time of diagnosis and decreases with later adjustment (Folkman, Lazarus, Gruen, & DeLongis, 1986), and that increased resolution regarding a child's diagnosis (i.e., acceptance and appropriate coping) is related to lower levels of parental stress (Sheeran, Marvin, & Pianta, 1997). These findings suggest that coping style and adjustment are more salient to psychological outcome than disease severity. It may also be that these mothers of boys with DMD have access to and make use of formal support services and their informal support system to a degree that allows for improved functioning over time. Our findings also indicate that boys with DMD exhibit social skills impairments, and maternal stress that improved over time was specifically related to child variables. Boys with DMD, then, may also learn improved social skills over the years, thereby helping to minimize maternal stress. It should also be noted that the significant decrease found in stress related to child variables occurred over the span of 4 years. Perhaps support services at the time of diagnosis would aid these families in coping with disease-related stress and contributing factors (such as delayed social skills in boys with DMD) more quickly. An alternative explanation for the decrease in stress found in our longitudinal sample is that these mothers—a smaller, willing, and able subset of the total group studied—are those who are coping better, and who may therefore continue to participate in the research. Finally, it must be emphasized that these findings are based upon a small sample and should be interpreted with caution. Future studies should examine stress longitudinally in a larger sample, taking into account support resources available and utilized.

Together, these findings support the notion that maternal stress is related to poor social skills exhibited by some boys with DMD, rather than the requirements of caretaking alone. Across the four parts of the study, disease progression and disability did not alone account for parental stress, as expected. Rather, behavior was consistently found to contribute to stress related to difficult parent-child interactions and to the child himself. Further, mothers of boys with DMD reported more stress than those of children with CP, suggesting that disease demands alone do not account for stress experienced. Finally, as stress was found to diminish over time as the disease progressed, rather than to increase as was hypothesized, it appeared that mothers learned to cope with the burdens associated with the disease as well as problematic social interactions with their sons.

However, these findings should be considered within the context of the limitations of the present study. For example, our data included only information from the mothers of boys with DMD, whereas a more complete picture of family functioning would include information about paternal stress and sibling functioning. The present study also did not incorporate additional variables that may have an impact upon maternal stress, such as resources available and utilized by each family (e.g., support or therapeutic services). Additionally, the findings are limited by the incomplete measurement of functional impairment, measured by use of a wheelchair at time of assessment. Further, assessment of maternal stress and child problem behaviors were both reported by the mothers of boys with DMD, suggesting a potential confound.

In sum, the present study does provide initial information about maternal stress in families of boys with DMD, a sample that has received little attention in the pediatric psychology literature. The investigation indicates that mothers of boys with DMD do experience significantly greater stress than mothers of healthy children, suggesting a need for increased services to help families cope with the effects of this disease. This stress is perhaps not experienced as being related to the child with DMD alone, as stress reported with regard to the siblings of boys with DMD was, in the main, not significantly lower. Mothers of boys with greatest social impairment reported the highest levels of maternal stress, suggesting a need for interventions targeting improved social interactions in boys with DMD and decreasing stress in their mothers. Further, it may be that the problematic social interactions and associated behaviors found in many boys in the DMD group accounted for maternal stress to a greater degree than did the physical characteristics and demands of the illness. Finally, stress related to child characteristics in mothers of boys with DMD decreases over time, suggesting a need for further study, in a larger sample, of supports and services which aid mothers in coping with this progressively worsening illness and facilitate timely adjustment to illness demands. These data suggest that interventions aimed at improved social interactions in boys with DMD, and providing support and strategies for mothers caring for these boys, would likely minimize the level of maternal stress experienced in families of boys with DMD.

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References

Abidin, R. (1990). *The Parental Stress Index: Short Form*. Charlottesville, VA: Pediatric Psychology Press.

Abidin, R. (1995). *The Parenting Stress Index–Short Form*. Odessa, FL: Psychological Assessment Resources.

Achenbach, T. M. (1991). *Manual for the Child Behavior Checklist/4–18 and 1991 profile*. Burlington: University of Vermont, Department of Psychiatry.

Arad, I., Durkin, M. S., Hinton, V. J., Kuhn, L., Chiriboga, C., Kuban, K., et al. (2002). Long-term cognitive benefits of antenatal corticosteroids for prematurely born children with cranial ultrasound abnormalities. *American Journal of Obstetrics and Gynecology, 186,* 818–825.

Beckman, P. J. (1993). Comparison of mothers' and fathers' perceptions of the effect of young children with and without disabilities. *American Journal of Mental Retardation, 95,* 585–595.

Chiriboga, C. A., Kairam, R., & Kline, J. (1993). A neurological examination for children: Reliability and utility in Studies of HIV infection. *Pediatric AIDS & HIV: Fetus to Adolescent, 4,* 144–150.

Copeland, D. R. (1988). Stress and the patient's family. In M. L. Russell (Ed.), *Stress management for chronic disease* (pp. 30–48). New York: Pergamon.

Dunn, L. M., & Dunn, L. M. (1981). *The Peabody Pic-*

ture Vocabulary Test–Revised: Manual for forms L and M. Circle Pines, MN: American Guidance Service.

Dunn, L. M., & Dunn, L. M. (1997). *Examiner's manual for the PPVT-III.* Circle Pines, MN: American Guidance Service.

Floyd, F. J., & Gallagher, E. M. (1997). Parental stress, care demands, and use of support services for school-age children with disabilities and behavior problems. *Family Relations, 46,* 359–371.

Folkman, S., Lazarus, R. S., Gruen, R. J., & DeLongis, A. (1986). Appraisal, coping, health status, and psychological symptoms. *Journal of Personality and Social Psychology, 50,* 571–579.

Goldberg, S., Morris, P., Simmons, R. J., Fowler, R. S., & Levison, H. (1990). Chronic illness in infancy and parenting stress: A comparison of three groups of parents. *Journal of Pediatric Psychology, 15,* 347–358.

Gravelle, A. M. (1997). Caring for a child with a progressive illness during the complex chronic phase: Parents' experience of facing adversity. *Journal of Advanced Nursing, 25,* 738–745.

Hinton, V. J., De Vivo, D. C., Nereo, N. E., Goldstein, E., & Stern, Y. (2000). Poor verbal working memory across intellectual level in boys with Duchenne dystrophy. *Neurology, 13,* 2127–2132.

Hinton, V. J., De Vivo, D. C., Nereo, N. E., Goldstein, E., & Stern, Y. (2001). Selective deficits in verbal working memory associated with a known genetic etiology: The neuropsychological profile of Duchenne muscular dystrophy. *Journal of the International Neuropsychological Society, 7,* 45–54.

Hinton, V. J., & Fee, R. (2000). *Evidence of poor social skills among boys with Duchenne muscular dystrophy.* Paper presented at the Gatlinburg Conference on Research and Theory in Mental Retardation and Developmental Disabilities, San Diego, CA.

Holmbeck, G. N., Gorey-Ferguson, L., Hudson, T., Seefeldt, T., Shapera, W., Turner, T., et al. (1997). Maternal, paternal, and marital functioning in families of preadolescents with spina bifida. *Journal of Pediatric Psychology, 22,* 167–181.

Holroyd, J., & Guthrie, D. (1986). Family stress with chronic childhood illness: Cystic fibrosis, neuromuscular disease, and renal disease. *Journal of Clinical Psychology, 42,* 552–561.

Leviton, A., Paneth, N., Susser, M., Reuss, M. L., Allred, E. N., Kuban, K., et al. (1997). Maternal receipt of magnesium sulfate does not seem to reduce the risk of neonatal white matter damage. *Pediatrics, 99,* E2.

Manuel, J. C. (2001). Risk and resistance factors in the adaptation in mothers of children with juvenile rheumatoid arthritis. *Journal of Pediatric Psychology, 26,* 237–246.

Mendel, J. R., Griggs, R. C., & Ptacek, L. J. (1998). Diseases of muscle. In A. S. Fauci, E. Braunwald, K. J. Isselbacher, et al. (Eds.), *Harrison's principles of internal medicine* (14th ed.) (pp. 2473–2483). New York: McGraw-Hill.

Mobarak, R., Khan, N. Z., Munir, S., Zaman, S. S., & McConachie, H. (2000). Predictors of stress in mothers of children with cerebral palsy in Bangladesh. *Journal of Pediatric Psychology, 25,* 427–433.

Nereo, N. E., & Hinton, V. J. (2003). Three Wishes and psychosocial functioning in boys with Duchenne muscular dystrophy. *Journal of Developmental and Behavioral Pediatrics, 24,* 96–103.

Pelchat, D., Ricard, N., Bouchard, J. M., Perreault, M., Saucier, J. F., Berthiaume, M., et al. (1999). Adaptation of parents in relation to their 6-month-old infant's type of disability. *Child: Care, Health and Development, 25,* 377–397.

Roach, M. A., Orsmond, G. I., & Barratt, M. S. (1999). Mothers and fathers of children with Down syndrome: Parental stress and involvement in childcare. *American Journal of Mental Retardation, 104,* 422–436.

Robson, A. L. (1997). Low birth weight and parenting stress during early childhood. *Journal of Pediatric Psychology, 22,* 297–311.

Saviolo-Negrin, N., Cristante, F., Zanon, E., Canclini, M., Stocco, D., & Girolami, A. (1999). Psychological aspects and coping of parents with a haemophilic child: A quantitative approach. *Haemophilia, 5,* 63–68.

Sheeran, T., Marvin, R. S., & Pianta, R. C. (1997). Mothers' resolution of their child's diagnosis and self-reported measures of parenting stress, marital relations, and social support. *Journal of Pediatric Psychology, 22,* 197–212.

Smith, T. B., Oliver, M. N., & Innocenti, M. S. (2001). Parenting stress in families of children with disabilities. *American Journal of Orthopsychiatry, 71,* 257–261.

Soliday, E., Kool, E., & Lande, M. B. (2000). Psychosocial adjustment in children with kidney disease. *Journal of Pediatric Psychology, 25,* 93–103.

Spaulding, B. R., & Morgan, S. B. (1986). Spina bifida children and their parents: A population prone to family dysfunction? *Journal of Pediatric Psychology, 11,* 359–374.

Walker, L. S., Ford, M. B., & Donald, W. D. (1987). Cystic fibrosis and family stress: Effects of age and severity of illness. *Pediatrics, 79,* 239–246.