Factors Associated with Parenting Stress in Mothers of Children with Fragile X Syndrome

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ABSTRACT. Whereas previous research has demonstrated elevated levels of parenting stress in parents of children with general developmental disability, there has been little investigation of stress in parents of children specifically affected by the common neurogenetic disorder fragile X syndrome (FraX). This study elucidates stress profiles in mothers of children with FraX and delineates the contribution of child characteristics, home environment, and maternal psychological functioning to specific dimensions of parental stress. Data on child, home, and family characteristics were collected from 75 families with a child affected by FraX. These characteristics were entered into multiple regression analyses with a domain or subscale of the Parenting Stress Index as the dependent variable in each analysis. The results demonstrated that aspects of child behavior, family cohesion, household income, and maternal psychopathology differentially correlate with specific dimensions of parenting stress. Determining the relative contribution of factors associated with stress will assist in the development of interventions to improve parental well-being in mothers of children with FraX. *J Dev Behav Pediatr 24:267–275, 2003.* Index terms: *fragile X syndrome, maternal stress.*

It has been well documented that parents of children with developmental delay are at risk for increased levels of psychological stress when compared with parents of typically developing children.[1–8] Although there have been

progressive changes in the distribution of parenting responsibilities between mothers and fathers,[9] mothers continue to carry the disproportionate burden in raising a disabled child, thereby being more inclined to experience stress related to child care.[10–18] In response to heightened stress, mothers of children with developmental delay often demonstrate depression, anxiety, health problems, social isolation, and low self-esteem.[3,5,11,18–22]

Whereas several studies have examined how parents are affected by the challenges of raising a child with nonspecific developmental delay, there has been little investigation

of parenting stress in mothers of children affected by fragile X syndrome (FraX), one of the most common genetic causes of developmental disability. FraX is caused by a mutation in a single gene on the X chromosome and is the most commonly inherited form of mental retardation with an incidence of approximately 1 in 2000 to 1 in 4000.[23] Male children with the full mutation typically have mental retardation with particular cognitive deficits in arithmetic reasoning, visual spatial abilities, processing sequential information, working memory, and attention.[24–26] Affected individuals often experience high rates of behavioral and emotional problems including difficulties with peer social interactions, gaze avoidance, social anxiety, hyperactivity, and stereotypical behavior.[27–31] Female children display similar cognitive difficulties and behavioral problems, but the degree of impairment is typically less severe because of the presence of an intact gene on the second X chromosome.

Given that FraX is associated with a predisposition for a particular cognitive and behavioral phenotype, it is of potential importance to examine the causes of parenting stress in mothers of children within this distinct population. Previous research has shown that, in comparison with the normative sample for the Parenting Stress Index (PSI),[32] parents of children with FraX experience high levels of parenting stress,[33] but, to our knowledge, the factors influencing stress have never been investigated. Delineating the relative contribution of factors associated with parenting stress will assist in the development of more specific intervention programs designed to improve adaptation in mothers of children with FraX.

This study elucidates stress profiles in mothers of children with FraX and evaluates the relative contribution of child characteristics, the home environment, and maternal psychological functioning to overall parenting stress and specific dimensions of parenting stress. We hypothesized that, when compared with the normative sample for the PSI,[32] mothers of children with FraX would report higher levels of total stress, more feelings of isolation, difficulties with self-perceptions of parenting competence, and difficulties accepting the child. We further hypothesized that the following aspects of the child, family, and mother would be associated with maternal stress: (1) child behavioral problems, (2) intelligence of the child, (3) child age, (4) family cohesion, (5) family income, and (6) maternal psychological well-being.

METHODS

Subjects

A total of 120 families participated in a study of children and adolescents with fragile X syndrome (FraX). Families were recruited from an existing fragile X registry, the Stanford Child Psychiatry Department research website, and the National Fragile X Foundation, as well as through referrals from other researchers, clinicians, and families. To eliminate potential confounding effects of having more than one child with FraX contributing to parenting stress characteristics, families with only one child in the house-

hold diagnosed with the full mutation were included in the current analyses. To further remove confounds within the dimension of parenting stress, families were included in the current analyses only if all other children in the family were healthy and typically developing.

In addition, families were included only if the mother had the FraX *premutation* as opposed to the FraX full mutation. Because it has been found that females with the full mutation have a variable phenotype often exhibiting a range of learning disabilities and psychological problems,[34,35] exclusion of mothers with the full mutation ensured that stress levels were not influenced by lack of coping mechanisms caused by cognitive deficits or pre-existing psychiatric conditions associated with the full mutation.

After removing all confounds, 75 families were included in the current analyses; 56 of these families had a male child with FraX and 19 families had a female child with FraX. All children were aged between 6 and 17 years (10.90 ± 2.64 years). Of the children in the sample, 92% were white, 4% were Hispanic, 1.3% were African American, 1.3% were Asian, and 1.3% were multi-ethnic. Families in 26 states and Canada, across urban, suburban, and rural areas, were represented in the sample. Mothers were aged between 30 and 52 years (40.61 ± 4.71 years). Within the sample, 82.7% of mothers were married and 17.3% were single. The highest level of education for the mother was as follows: 4.0% partial high school, 17.3% high school diploma, 1.3% technical college, 33.3% partial college, 30.7% college degree, 10.7% graduate degree, and 2.7% unreported. Of all mothers in the sample, 65.3% were employed and 34.7% were homemakers.

Procedures

To determine a family's eligibility, previous results of fragile X testing were requested. DNA testing for the *FMR1* mutation was performed on all probands and previously untested family members. The diagnoses of children with FraX, their siblings, and mothers were confirmed by Southern Blot DNA analysis. Southern blot analyses were performed as detailed by Taylor et al[36] at Kimball Genetics, Inc. (Denver, CO).

Mothers of children with FraX responded to a demographics questionnaire, the Parenting Stress Index (PSI), Child Behavior Checklist (CBCL), Symptom Checklist-90-R (SCL-90-R), and Family Environment Scale (FES). Children and adolescents with FraX were administered the complete Wechsler Intelligence Scale for Children (WISC)-III. Test administration and questionnaire completion were accomplished in the family household. All assessors were trained in adherence to a standardized protocol and supervised by a licensed psychologist.

Measures

Parenting Stress Index. The PSI[32] is a 120-item self-report questionnaire used to determine the quality and magnitude of stress experienced by a parent in relation to his or her child. The Child Domain includes questions targeting how the child's behavior impacts the parent, and the Parent Domain incorporates questions about the parent's

ability to cope with the task of parenting. The Total Domain provides a composite score including both Child and Parent Domains. The PSI, renormed in 1990, is a standardized measure with sound psychometric properties. The internal consistency reliabilities of the PSI Domains, as measured by the Cronbach alpha statistic, are the following: Parent Domain = .93, Child Domain = .90, and Total Domain = .95. Percentile scores, derived from the frequency distribution of the normative data, are used to interpret responses on the PSI, with the "mean" as 50% and normal range scores within the 15th to 80th percentiles. A high score on a subscale indicates a higher level of stress reported by the parent (i.e., a high score on the Competence subscale is associated with more stress related to feelings of parenting competence). According to the PSI manual, parents whose scores are considered high (i.e., ≥85th percentile) would typically be considered candidates for referral or professional services.

Subscale scores within each domain provide detailed information about the sources of stress. The Child Domain is composed of the following subscales: adaptability, acceptability, demandingness, mood, distractibility/hyperactivity, and reinforcement given to the parent. The Parent Domain is composed of the following: depression, attachment, restriction of role, sense of competence, social isolation, relationship with spouse, and parental health. Based on an exhaustive review of the literature, a priori hypotheses targeted three subscales of interest as specific dimensions of maternal stress: (1) Isolation (Parent Domain), (2) Competence (Parent Domain), and (3) Acceptability (Child Domain).

The Isolation subscale (alpha .82) measures the degree to which a parent feels socially isolated from peers, relatives, and other emotional support systems.[32] The Competence subscale (alpha .83) measures how competent a parent feels with the tasks of parenting, with high scores indicating that the respondent does not find the role of parenting as reinforcing as they expected.[37] Finally, the Acceptability subscale (alpha .79) measures the coping skills of parents when they are faced with the knowledge that their child does not fulfill parental expectations.[32] We hypothesized that these subscales capture the mother's ability to cope with the task of parenting and the effect that parenting a child with a disability may have on the mother. These subscales and the Parent Domain score were used as dependent variables in our analyses of parental stress.

Child Behavior Checklist. The CBCL[38] is a standardized and widely used instrument that rates behavioral problems in children and adolescents. Parents report the presence and severity of behavioral problems in the following domains: withdrawn behavior, social problems, anxiety and depression, somatic complaints, attention problems, thought problems, aggressive behavior, and delinquent behavior. The CBCL also yields three composite scores: Total Behavioral Problems, Externalizing Behavioral Problems, and Internalizing Behavioral Problems. The Total Behavioral Problems scale was used as an independent variable in this study. The mean for this measure is 50, with a standard deviation of 10. Test-retest reliability in a 7-day interval for the Total Problems scale is .93.

Wechsler Intelligence Scale for Children-III. The WISC-III is a standardized intellectual assessment for children aged 6 to 17 years yielding Verbal, Performance, and Full-Scale IQ (FSIQ).[39] The FSIQ score was used in the present analyses as a predictor of parental stress. The IQ scores have a mean of 100 with a standard deviation of 10, and reliability coefficients for the FSIQ are very high, ranging between .94 and .97, varying by age.

Family Environment Scale-Cohesion Subscale. The Cohesion subscale of the FES[40] assesses the degree of commitment, support, and help that family members provide for one another. Although the FES is composed of 10 subscales measuring the social environment of families, the Cohesion subscale best represents the relationship dimensions of greatest interest in this study and was used as an independent variable. This measure was originally developed as a true-false self-report questionnaire. Mothers in our study had difficulty responding to the items on a dichotomous scale, and it was therefore modified to a Likert response scale from 0 to 5. The internal consistency reported by Moos and Moos[40] for the true-false Cohesion subscale is .78. Although there are no separate estimates of internal reliability for the Likert format, the internal subscale reliability should be higher when the Likert response option is used than when a true-false option is used (Rudolf Moos, personal communication, 2003).

Family Adjusted Income. Annual household income was adjusted for regional differences in housing and cost of living. Parent report of gross annual household income was divided by the median household income in the family's area as defined by the home's ZIP code. The ZIP code median income was determined by Decisionmark Corporation (Cedar Rapids, IA) and based on the 1990 United States Census data and the Census Bureau 1998 estimates and 2003 projections. These data were obtained from the website www.homes.com. We limited the demographic variables included in our model to avoid multicollinearity among our predictor variables. Adjusted income offered more statistical power than the other demographic variables because it was measured on a continuous scale and had a normal distribution. The other demographic variables were measured on ordinal or dichotomous scales, and some had skewed distributions.

Symptom Checklist-90-R. The SCL-90-R[41] is a standardized self-report measure of psychopathological symptoms. The 90 questions are clustered into the following symptom dimensions: somatization, obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, and psychoticism. The Global Severity Index (GSI) is generated as an indicator of total psychiatric disturbance because it combines information concerning both the intensity of distress and the number of symptoms reported from all the symptom dimensions. Because the GSI provides the best gauge of the current level and depth of a psychiatric condition, this score was used as a predictor in our analyses. The normed mean score for the SCL-90-R GSI is a T score of 50, with a standard deviation of 10. Internal consistency of the symptom dimensions included in the GSI ranges from .77 to .90[42,43], and test-retest reliability of 1 week ranges from .80 to .90.[42]

Statistical Analyses

Descriptive statistics were calculated for the PSI Child, Parent, and Total Domains, and for the subscales selected a priori for our analyses. Multiple regression analyses were conducted to determine the degree to which child characteristics, home environment, and maternal psychopathology were associated with different dimensions of parenting stress.

Four regression models, each entered in one block, were tested using the following PSI raw scores as the dependent variables: Isolation, Parenting Competence, Acceptability, and Parent Domain. The PSI Child and Total Domains were not included as dependent variables in the multiple regression analyses because several questions within these domains pertain to child behavioral problems, creating an overlap between the constructs of Child and Total Domains and the CBCL. The Pearson correlation in our sample between the CBCL and Child PSI Domain score was 0.663 ($p < .001$), and the Pearson correlation between the CBCL and Total PSI Domain score was .573 ($p < .001$). Analyses would therefore be confounded because of the shared variance between the measures.

As noted in the "Measures" section, predictor variables were selected a priori on the basis of an exhaustive review of the literature, statistical limitations imposed because of sample size, and available data. The number of independent variables entered into the model was limited to optimize statistical power and to avoid potential confounds, including collinearity.

In the Isolation, Competence, and Acceptability analyses, six predictor variables were entered including the CBCL total score, WISC-III FSIQ score, child age, family adjusted income, FES-Cohesion, and SCL-90-R-GSI. In the Parent Domain analyses, all predictor variables just mentioned, with the exception of SCL-90-R-GSI, were entered. The GSI was not used as a predictor variable for the Parent Domain Stress score because both the SCL-90-R and the Parent Domain of the PSI incorporate a number of similar questions pertaining to parent psychopathology

(i.e., depression). For all multiple regression analyses, the alpha value for statistical significance was .05 (two-tailed).

RESULTS

Descriptive Statistics

Descriptive statistics of independent and dependent variables are shown in Table 1. The results of the Parenting Stress Index (PSI) domain scores revealed that the mean percentile score of the mothers was in the clinically significant range on the Child Domain scale, in the normal range on the Parent Domain scale, and in the high, but not clinically significant, range on the Total Domain scale ($72.5\% \pm 20.9\%$). The mean Acceptability score was in the high range, and the mean scores for the Competence and Isolation subscales were in the normal range. The inter-correlations among independent and dependent variables are presented in Table 2. A surprising result was the low correlation ($r = .066$) between Global Severity and Acceptability, given the moderate correlation between the Child Behavior Checklist (CBCL) and Global Severity ($r = .350$) and the moderate correlation between CBCL and Acceptability ($r = .320$). A possible explanation for the unanticipated low correlation is that the variance shared between CBCL and Global Severity is independent and unique from variance shared between CBCL and Acceptability. A post hoc regression (entered in one block) with CBCL as the outcome variable, and Acceptability and Global Severity as predictors, was generated. Each predictor was found to contribute independently to the variance in CBCL scores: Global Severity/CBCL ($\beta = .33$, $t = 3.147$, $p = .002$) and Acceptability/CBCL ($\beta = .298$, $t = 2.842$, $p = .006$). The partial correlations with CBCL were Global Severity $r = .348$ and Acceptability $r = .318$.

Multiple Regression Analyses

All independent variables in the multiple regression analyses were entered in one block. The models with Parent

Table 1. Descriptive Statistics of Independent and Dependent Variables

Independent Variables	Mean	SD	Range
Child Age	10.90	2.64	6.03–16.95
Wechsler Full Scale IQ	51.65	15.05	40–103
CBCL Total Problems	59.77	9.17	37–77
Family Cohesion	3.74	0.48	2.44–4.67
Adjusted Household Income[a]	1.56	0.82	0.46–5.41
Global Severity Index	53.43	9.80	30–79
Dependent Variables (PSI)[b]	Percentile Mean	SD	% in High Range
Total Stress	75.2	20.9	42
Parent Stress	55.5	25.4	16
Child Stress	87.3	18.3	75
Subscales			
Isolation	62.0	25.2	22
Competence	54.2	26.5	21
Acceptability	93.01	14.24	92

CBCL, Child Behavior Checklist; PSI, Parenting Stress Index.
[a]Adjusted family income is total household income divided by the median household income in the ZIP code of the family's home.
[b]For the dependent variables, the normative sample scored in the 50th percentile on each domain and subscale.

Table 2. Intercorrelations Among Independent and Dependent Variables (n = 75)

	1	2	3	4	5	6	7	8	9	10
1. CBCL Total	...	−.265*	−.006	−.194	.004	.350**	−.054	.459**	.320**	.321**
		.021	.959	.095	.975	.002	.644	.000	.005	.005
2. Child FSIQ	−.265	...	.038	.134	.160	−.114	.001	−.311**	−.284*	−.160
	.021		.748	.253	.171	.331	.993	.007	.014	.171
3. Child Age	−.006	.038	...	−.207	.149	−.089	.053	.073	.170	.106
	.959	.748		.074	.203	.450	.654	.534	.144	.365
4. Cohesion	−.194	.134	−.207	...	−.008	−.168	−.271*	−.334**	−.306**	−.464**
	.095	.253	.074		.944	.151	.019	.003	.008	.000
5. Adjusted Income	.004	.160	.149	−.008	...	.002	.116	−.089	−.243*	.014
	.975	.171	.203	.944		.998	.322	.446	.035	.905
6. Global Severity	.350**	−.114	−.089	−.168	.002	...	.324**	.389**	.066	.531**
	.002	.331	.450	.151	.998		.005	.001	.576	.000
7. Isolation Raw	−.054	.001	.053	−.271*	.166	.324**	...	.180	.165	.701**
	.644	.993	.654	.019	.322	.005		.122	.157	.000
8. Competence Raw	.459**	−.311**	.073	−.334**	−.089	.389**	.180	...	.340**	.685**
	.000	.007	.534	.003	.446	.001	.122		.003	.000
9. Acceptability Raw	.320**	−.284*	.170	−.306**	−.243*	.066	.165	.340**	...	.361**
	.005	.014	.144	.008	.035	.576	.157	.003		.001
10. Parent Domain Raw	.321**	−.160	.106	−.464**	.014	.531**	.701**	.685**	.361**	...
	.005	.171	.365	.000	.905	.000	.000	.000	.001	

CBCL, Child Behavior Checklist; FSIQ, Full-Scale IQ.
*Correlation is significant at the .05 level (two-tailed); **correlation is significant at the .01 level (two-tailed).

Isolation, Parental Competence, Acceptability, and Parent Domain Stress scores as the dependent variables were all statistically significant (Table 3). Results of the analysis with the PSI Isolation score as the outcome variable indicated that Family Environment Scale (FES)-Cohesion scores and the Symptom Checklist-90-R (SCL-90-R)–Global Severity Index (GSI) scores were both significantly correlated with Isolation scores ($\beta = -.250$, $p = .031$, $\beta = .363$, $p = .003$), such that lower levels of family cohesion and higher levels of parent psychological symptoms were independently associated with increased isolation. The association between lower CBCL total scores and higher Isolation scores approached significance ($\beta = -.231$, $p = .057$).

The regression model with Competence as the dependent variable demonstrated that a high CBCL Total explained a significant amount of the variance ($\beta = .292$, $p = .009$), followed by SCL-90-R–Global Severity ($\beta = .241$, $p = .025$). These results indicate that both increased severity of child behavioral problems and higher levels of parent psychological problems were significantly associated with an increased stress response related to parenting competence. The association between family cohesion and high Competence scores approached significance ($\beta = -.212$, $p = .056$), such that decreased support from family members was correlated with higher stress levels associated with competence.

CBCL Total ($\beta = .292$, $p = .026$) and family adjusted income ($\beta = -.245$, $p = .024$) were both significant predictors of high Acceptability, whereas low family cohesion approached significance ($\beta = -.212$, $p = .056$). These findings indicate that severity of behavioral problems and lower family income may influence whether a child meets the expectations of the parent. In addition, low family cohesion may negatively influence a mother's feelings of acceptability toward her child.

In the analyses with PSI Parent Domain scores as the outcome variable, low FES-Cohesion and high CBCL Total explained a significant amount of the variance in high Parenting Stress ($\beta = -.408$, $p < .001$, $\beta = .229$, $p = .038$), such that less family support and more child behavioral problems were associated with higher overall levels of parenting stress.

DISCUSSION

This study is among the first empirical investigations examining the potential sources of parental stress in mothers of children with fragile X syndrome (FraX). Consistent with our hypotheses, mothers of children

Table 3. Results of Multiple Regression Analyses Predicting Different Domains of Maternal Stress (n = 75)

Dependent Variable	Independent Variable	Beta	p
Isolation	CBCL total	−.231	.057
Adj. R^2 = .142	FSIQ	−.004	.972
p = .011	Age	.015	.896
	FES cohesion	−.250	.031
	Family adjusted income	.113	.311
	Global Severity	.363	.003
Competence	CBCL total	.292	.009
Adj. R^2 = .303	FSIQ	−.170	.102
p < .0001	Age	.072	.477
	FES cohesion	−.200	.056
	Family adjusted income	−.076	.447
	Global Severity	.241	.025
Acceptability	CBCL total	.260	.026
Adj. R^2 = .214	FSIQ	−.161	.144
p = .001	Age	.165	.130
	FES cohesion	−.212	.056
	Family adjusted income	−.245	.024
	Global Severity	−.064	.566
Parent domain	CBCL total	.229	.038
Adj. R^2 = .221	FSIQ	−.047	.663
p < .0001	Age	.022	.834
	FES cohesion	−.408	.000
	Family adjusted income	.014	.894

CBCL, Child Behavior Checklist; FSIQ, Full-scale IQ; FES, Family Environment Scale.

affected by FraX had higher levels of total parenting stress when compared with the mean normative scores for the Parenting Stress Index (PSI).[32] The Child Domain stress was higher than the Parent Domain stress, indicating that the characteristics of the child are the major factors contributing to the overall stress experienced by mothers. These results replicate the pattern of findings by Sarimski in which mean scores in parents of children with FraX were in the 96th percentile on the PSI Child Domain (raw score = 133) and in the 65th percentile on the PSI Parent Domain (raw score = 129).[33]

Although mean scores on the Isolation and Competence subscales were within the normal range, approximately one quarter of the sample was in the clinically high range for both subscales. These results show that a proportion of mothers experience levels of social isolation and diminished parenting confidence to an extent that may warrant professional referral. The mean score for the Acceptability scale was in the high range, suggesting that children in our sample demonstrate challenges to the mother that can influence feelings of acceptability toward the child.

The identification of parental stress domains that are significantly influenced by child, family, and parent characteristics are of potential importance to health care providers, teachers, and members of advocacy and support groups. This knowledge will assist in recognizing problems that are potentially amenable to intervention and will ultimately guide the way for future longitudinal studies in which the effectiveness of focused treatments on family functioning are evaluated. Accordingly, in subsequent sections, we discuss personal, family, and environmental factors that can influence the perception of stress and determine the relative contribution of these factors to measured domains of maternal stress.

Child Behavioral Problems

We observed that behavioral problems of the child were significantly associated with the overall parental stress, as well as with maternal feelings of competence and acceptability. Similar to these findings, studies on children with general developmental delay have shown that child behavioral problems are significant sources of general parental stress.[1,6,44] Because behavioral problems are associated with dimensions of maternal stress in this population, well-designed parent support programs focused on positive behavioral management could improve both the behavioral competence of children affected within FraX as well as the adaptability and mental health of their mothers. Other investigators have demonstrated that such training programs lead to significantly decreased parental reports of depression and overall family stress.[45]

Another interesting finding was the association (approaching significance) between fewer behavioral problems and higher levels of isolation. Previous literature has shown that increased social support will lead to successful adaptation in parents of children with developmental delay.[46–48] It is probable that mothers of children who exhibit difficult behaviors are more inclined to join support groups, thereby leading to decreased feelings of isolation.

Child Intelligence

In addition to child behavioral problems, we hypothesized that cognitive functioning of the child would be significantly associated with maternal stress. Contrary to our expectations, cognitive functioning was not associated with any of the outcome variables. A plausible explanation for our results is that parenting stress is more directly associated with the challenges of managing difficult behavior than coping with the child's mental impairment. This is supported by other researchers[18,49–51] who found that mothers of children with Down syndrome have much healthier adaptation and coping mechanisms, as well as less parenting stress, when compared with mothers of autistic children. This finding may be, in part, due to the reduced level of behavioral problems associated with Down syndrome when compared with autism.

Child Age

Contrary to our hypothesis, child age was not associated with any dimension of maternal stress. Previous studies have found that as a developmentally delayed child grows older, family stress increases as parents develop a more realistic view of the child's future and outcome.[4,49,50,52] A possible interpretation of our results is that whereas other

studies focused on parents of children with autism or idiopathic conditions, our study specifically investigated mothers of children with an identifiable genetic disorder. It is possible that when there is a readily identifiable biological explanation for a child's limitations, parental expectations for the child's outcome are more realistic from the time of diagnosis. In this case, stress levels might not be affected as a child passes through different stages of development. It should be noted, however, that there have been studies of general developmental delay in which the age of the child has no effect on parental stress levels.[23,53,54]

Family Cohesion

Consistent with our hypothesis, cohesiveness of the family was significantly correlated with global parental stress and isolation. In addition, the statistical influence of family cohesion approached significance with acceptability and competence. The Cohesion subscale of the Family Environment Scale (FES) includes questions about sharing responsibilities and interpersonal support within the home. Mothers in our study appeared less susceptible to stress, reported fewer feelings of isolation, and felt more confident in their parenting skills when they had assistance and support from other family members. Recognizing informal support systems as valid providers of intervention for families with children with special needs is justified by the outcomes of this study.

Family Income

A review of the literature confirms the impact of socioeconomic status and family support on maternal stress and coping resources on children with autism and developmental delay.[4,6,53,55,56] In the current study, family income was found to be negatively associated with maternal feelings of acceptability. There is more than one possible explanation for this finding. Consistent with a broadly studied phenomenon in the literature, we found a correlation ($\beta = .289$, $p = .013$) between family income and maternal educational levels in our sample. Consequently, a relative lack of exposure to the characteristics, needs, and capabilities of children with developmental delay may engender unrealistic expectations on the part of the parents of these children, thereby leading to lower acceptability of the child. Another explanation is that meeting the basic financial needs of one's family may supersede a mother's time to access and take advantage of support and informational resources, thus resulting in a decreased ability to accept her child. Efforts should be made to ensure support services are not only available but also used by families from all socioeconomic strata.

Maternal Psychological Well-Being

The psychological well-being of mothers of children with FraX was significantly associated with parental feelings of isolation and, to a significant but lesser extent, with feelings of competence. It can be presumed that if a mother is suffering from significant psychological problems, she might not have adequate personal resources available for others or her child, thereby increasing her feelings of isolation and reducing a sense of competence in her parenting skills. The relationship between parental stress and psychopathology could be bidirectional, as a mother's feelings of isolation and lack of confidence related to parenting skills could also, in turn, affect her psychological well-being.

Study Limitations and Future Research

Several limitations restrict the interpretation of our findings. First, it should be noted that the associations in our results reflect correlational and not necessarily causal relationships. One cannot assume that the child, family, and parent characteristics in these analyses directly cause the examined dimensions of parental stress. Child problems and lack of family cohesion, for example, may cause stress, but increased parenting stress can likewise exacerbate child problems and erode family cohesion. It should be recognized that there may be a constant reciprocal interplay between the predictor and outcome variables.

Second, there are other potential demographic, psychosocial, and biological predictors of parenting stress that were not assessed in this study. These include maternal age, social networks and support, problem-solving and coping skills, religious affiliations, community resources (such as respite care), marital status and satisfaction, mother's education, mother's employment status, number of healthy children, and feelings of maternal culpability related to an X-linked disorder. In addition, it would be interesting to assess biological indicators such as *FMR1* protein, activation ratio, and methylation status. Future studies examining parenting stress should include measures of these other potentially moderating factors.

Third, it should be noted that common to most studies in the parenting stress literature, the sample in the current study was composed only of mothers. This study in no way disregards paternal stress as an outcome of raising a child with a disability, indeed it is recognized as a real and complex construct that should be studied as a unique phenomenon. Therefore, future studies should be designed to explore fathers' unique experiences.

Finally, research design and sampling techniques limit the generalizability of the findings. The lack of a comparison group of children with unspecified developmental delay or another specific neurogenetic disorder prevents a definitive conclusion that the correlations of child, family, and mother characteristics with stress are particular to mothers of children with FraX. Given that many neurogenetic syndromes are associated with specific cognitive and behavioral phenotypes,[57] it is important to examine potential sources of stress in primary caretakers of children with different disorders. A direct benefit of this research would be to provide a basis for the design of syndrome-specific interventions. In future studies, it will therefore be of interest to ascertain the stress profiles and factors related

to parental stress in mothers of children with different genetic causes of developmental disability.

Despite its limitations, the current study serves as a first step in characterizing the profile of factors associated with parental stress in mothers of children with FraX. This knowledge should permit health care providers to identify potential risk factors and warning signs of stress and will assist in determining when interventions may be necessary. Such interventions should particularly seek to address maternal feelings of stress, isolation, lack of parenting competence, and acceptability of the child.

REFERENCES

1. Cameron SJ, Orr RR. Stress in families of school-aged children with delayed mental development. *Can J Rehabil*. 1989;2:137–144.
2. Dyson LL. Families of young children with handicaps: parental stress and family functioning. *Am J Ment Retard*. 1991;95:623–629.
3. Friedrich WN, Friedrich WL. Psychosocial assets of parents of handicapped and nonhandicapped children. *Am J Ment Defic*. 1981;85:551–553.
4. Gallagher JJ, Beckman P, Cross AH. Families of handicapped children: sources of stress and its amelioration. *Except Child*. 1983; 50:10–19.
5. Miller AC, Gordon RM, Daniele RJ, et al. Stress, appraisal, and coping in mothers of disabled and nondisabled children. *J Pediatr Psychol*. 1992;17:587–605.
6. Quine L, Pahl J. Examining the causes of stress in families with severely mentally handicapped children. *Br J Soc Work*. 1985;15:501–517.
7. Sanders JL, Morgan SB. Family stress and adjustment as perceived by parents of children with autism or down syndrome: implications for intervention. *Child Fam Behav Ther*. 1997;19:15–32.
8. Wilton K, Renaut J. Stress levels in families with intellectually handicapped preschool children and families with non-handicapped preschool children. *J Ment Defic Res*. 1986;30:163–169.
9. Chira S. Census data show rise in child care by fathers. *New York Times*. September 22, 1993;A20.
10. Beckman PJ. Comparison of mothers' and fathers' perceptions of the effect of young children with and without disabilities. *Am J Ment Retard*. 1991;95:585–595.
11. Bristol MM, Gallagher JJ, Schloper E. Mothers and fathers of young developmentally disabled and nondisabled boys: adaptation and spousal support. *Dev Psychol*. 1988;24:441–451.
12. Freeman NL, Perry A, Factor DC. Child behaviours as stressors: replicating and extending the use of the CARS as a measure of stress. A research note. *J Child Psychol Psychiatry*. 1991;32:1025–1030.
13. Krauss MW. Child-related and parenting stress: similarities and differences between mothers and fathers of children with disabilities. *Am J Ment Retard*. 1993;97:393–404.
14. Lieberman MA. *The Effect of Social Supports on Responses to Stress*. New York, NY: Free Press (Macmillan); 1982.
15. Milgram NA, Atzil M. Parenting stress in raising autistic children. *J Autism Dev Disord*. 1988;18:415–424.
16. Moes D, Koegel RL, Schreibman L, et al. Stress profiles for mothers and fathers of children with autism. *Psychol Rep*. 1992;71:1272–1274.
17. Willoughby JC, Glidden LM. Fathers helping out: shared child care and marital satisfaction of parents of children with disabilities. *Am J Ment Retard*. 1995;99:399–406.
18. Wolf LC, Noh S, Fisman SN, et al. Psychological effects of parenting stress on parents of autistic children. *J Autism Dev Disord*. 1989;19:157–166.
19. Cummings ST, Bayley HC, Rie HE. Effects of the child's deficiency on the mother: a study of mothers of mentally retarded, chronically ill and neurotic children. *Am J Orthopsychiatry*. 1966;36:595–608.
20. Cummings ST. The impact of the child's deficiency on the father: a study of fathers of mentally retarded and of chronically ill children. *Am J Orthopsychiatry*. 1976;46:246–255.
21. Frey K: Stress and coping among parents of handicapped children: a multidimensional approach. *Am J Ment Retard*. 1989;94:240–249.
22. Wikler L. Chronic stress in families of mentally retarded children. *Fam Relat*. 1981;30:281–288.
23. Bailey DB, Nelson D. The nature and consequences of fragile X syndrome. *Ment Retard Dev Disabil Res Rev*. 1995;1:238–244.
24. Freund LS, Reiss AL. Cognitive profiles associated with the fra(X) syndrome in males and females. *Am J Med Genet*. 1991;38:542–547.
25. Hodapp RM, Leckman JF, Dykens EM, et al. K-ABC profiles in children with fragile X syndrome, Down syndrome, and nonspecific mental retardation. *Am J Ment Retard*. 1992;97:39–46.
26. Kemper MB, Hagerman RJ, Altshul-Stark D. Cognitive profiles of boys with the fragile X syndrome. *Am J Med Genet*. 1988;30:191–200.
27. Baumgardner TL, Reiss AL, Freund LS, et al. Specification of the neurobehavioral phenotype in males with fragile X syndrome. *Pediatrics*. 1995;95:744–752.
28. Bregman JD, Leckman JF, Ort SI. Fragile X syndrome: genetic predisposition to psychopathology. *J Autism Dev Disord*. 1988;18:343–354.
29. Cohen IL, Fisch GS, Sudhalter V, et al. Social gaze, social avoidance, and repetitive behavior in fragile X males: a controlled study. *Am J Ment Retard*. 1988;92:436–446.
30. Lachiewicz AM, Spiridigliozzi GA, Gullion CM, et al. Aberrant behaviors of young boys with fragile X syndrome. *Am J Ment Retard*. 1994;98:567–579.
31. Reiss AL, Freund L. Behavioral phenotype of fragile X syndrome: DSM-III-R autistic behavior in male children. *Am J Med Genet*. 1992;43:35–46.
32. Abidin RR. *Parenting Stress Index: Manual*. Odessa, FL: Psychological Assessment Resources, Inc; 1986.
33. Sarimski K. Behavioural phenotypes and family stress in three mental retardation syndromes. *Eur Child Adolesc Psychiatry*. 1997;6:26–31.
34. Freund LS, Reiss AL, Abrams MT. Psychiatric disorders associated with fragile X in the young female. *Pediatrics*. 1993;91:321–329.
35. Hagerman RJ, Jackson C, Amiri K, et al. Girls with fragile X syndrome: physical and neurocognitive status and outcome. *Pediatrics*. 1992;89:395–400.
36. Taylor AK, Safanda JF, Fall MZ, et al. Molecular predictors of cognitive involvement in female carriers of fragile X syndrome. *JAMA*. 1994;271:507–514.
37. Mouton PY, Tuma JM. Stress, locus of control, and role satisfaction in clinic and control mothers. *J Clin Child Psychol*. 1988;17:217–224.
38. Achenbach TM. *Manual for the Child Behavior Checklist/4-18*. Burlington, VT: University of Vermont, Department of Psychiatry; 1991.
39. Wechsler D. *Wechsler Intelligence Scale for Children-Manual*. 3rd ed. San Antonio, TX: The Psychological Corporation; 1991.

40. Moos R, Moos B: *Family Environment Scale Manual*. 3rd ed. Palo Alto, CA: Consulting Psychologists Press Inc; 1994.

41. Derogatis LR. *Symptom Checklist-90-R. Administration, Scoring, and Procedures-Manual*. 3rd ed. Minneapolis, MN: National Computer Systems, Inc; 1997.

42. Derogatis LR, Rickels K, Rock AF. The SCL-90 and the MMPI: a step in the validation of a new self-report scale. *Br J Psychiatry.* 1976;128:280–289.

43. Horowitz LM, Rosenberg SE, Baer BA, et al. Inventory of interpersonal problems: psychometric properties and clinical applications. *J Consult Clin Psychol*. 1988;56:885–892.

44. Donenberg G, Baker B. The impact of young children with externalizing behaviors on their families. *J Abnorm Child Psychol*. 1993;21:179–198.

45. Baker B, Landen S, Kashima K. Effects of parent training on families of children with mental retardation: increased burden or generalized benefit? *Am J Ment Retard*. 1991;96:127–136.

46. Dunst CJ, Trivette CM, Cross AH. Mediating influences of social support: personal, family, and child outcomes. *Am J Ment Defic*. 1986;90:403–417.

47. McCubbin HI, Patterson JM. *Family Adaptation to Crises*. Springfield, IL: Charles C. Thomas; 1983.

48. Rimmerman A. Mothers of children with severe mental retardation: maternal pessimism, locus of control and perceived social support. *Int J Rehabil Res*. 1991;14:65–68.

49. Bristol MM, Schopler E. Stress and coping in families of autistic adolescents. In: Schopler ME, Mesibov GB, eds. *Autism in Adolescents and Adults*. New York, NY: Plenum Press; 1983: 251–278.

50. Donovan AM. Family stress and ways of coping with adolescents who have handicaps: maternal perceptions. *Am J Ment Retard*. 1988; 92:503–509.

51. Holroyd J, McArthur D. Mental retardation and stress on the parents: a contrast between Down's syndrome and childhood autism. *Am J Ment Defic*. 1976;80:431–436.

52. Dyson LL. Response to the presence of a child with disabilities: parental stress and family functioning over time. *Am J Ment Retard*. 1993;98:207–218.

53. Flynt SW, Wood TA. Stress and coping of mothers of children with moderate mental retardation. *Am J Ment Retard*. 1989;94: 278–283.

54. Trivette CM, Dunst CJ. Characteristics and influences of role division and social support among mothers of preschool children with disabilities. *Top Early Child Spec Educ*. 1992;12:367–385.

55. Rabkin JG, Struening EL. Live events, stress, and illness. *Science*. 1976;194:1013–1020.

56. Warfield M, Wyngaarden M. Adaptation during early childhood among mothers of children with disabilities. *J Dev Behav Pediatr.* 1999;20:9–16.

57. Finegan JA. Study of behavioral phenotypes: goals and methodological considerations. *Am J Med Genet*. 1998;81:148–155.

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