

Predictors of stress in mothers and fathers of children with fragile X syndrome

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Abstract

This study examined parental and family stress and functioning where there is a child with fragile X syndrome. Mothers and fathers in 40 families were asked about their child with fragile X syndrome, family supports, their psychological stress, the marital relationship, and their family stress. Results indicate parents were well adjusted in terms of their levels of psychological stress and in their marital relationships, however, parents reported high levels of family stress. Mothers and fathers were found to experience similar levels of stress and to report similar levels of satisfaction with supports. Stress was predicted by different variables in mothers and fathers, suggesting that different processes underlie their experiences. The strongest predictor of maternal stress was the level of marital satisfaction while the strongest predictor of paternal stress was the level of the child's adaptive skills.

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Fragile X syndrome is the most common inherited form of intellectual disability, with an estimated incidence of 1 per 2000–4000 in the general population (Eliez & Reiss, 2000; Hagerman, 1997). It is an X-linked genetic condition that manifests in a variety of ways, with affected individuals presenting with a range of developmental, physical, behavioural and emotional characteristics (Lennox, Cohen, Slater, & Cook, 1998). The additional demands associated with the consequences of these characteristics may mean there is increased strain on parents.

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While parental functioning has been extensively examined in families with a child with a disability (e.g., Cuskelly, Jobling, Chant, Bower, & Hayes, 2002; Ricci & Hodapp, 2003; Roach, Orsmond, & Barrett, 1999), few studies have investigated parental experience specifically in families where there is a child with fragile X syndrome. When researchers have explored this area they have generally focused on the impact on parents of the diagnosis of fragile X syndrome (e.g., Bailey, Skinner, Hatton, & Roberts, 2000; Carmichael, Pembrey, Turner, & Barnicoat, 1999; Roy, Johnsen, Breese, & Hagerman, 1995). The examination of parental functioning within specific aetiological groups makes an important contribution to the literature as it is clear that parental experiences differ, depending upon the cause of their child's disability (Abbeduto et al., 2004; Fidler, Hodapp, & Dykens, 2000). It is particularly important in the case of fragile X syndrome as there is evidence that mothers of a child with fragile X may themselves have characteristics that make them vulnerable to the effects of increased child care demands (Reiss, Freund, Abrams, Boehm, & Kazazian, 1993).

While stress is not the only impact on parents of having a child with a disability, it is one of the constructs central to our understanding of their experience and of parental functioning. In essence, stress is a negative (usually, but not restricted to) psychological response to demands that are perceived to be greater than the resources available to meet them (Lazarus, 1993). The experience of stress may be manifested in a range of ways and researchers have operationalized it variously, including depression and anxiety (Spangenberg & Theron, 2001), negative impact on parents (Baker, Blacher, Crnic, & Edelbrock, 2002; Hodapp, Fidler, & Smith, 1998), and psychosomatic symptoms (Sloper, Knussen, Turner, & Cunningham, 1991). Higher levels of stress have consistently been found to occur among parents of a child with a disability when they are compared to parents whose children are all developing typically (see, for example, Cuskelly et al., 2002; Hodapp, Dykens, & Masino, 1997).

The higher levels of stress found in parents of children with a disability have been assumed to be a product of the increased demands they experience. These increased demands arise from the care needs of the child and the necessity to interact with and manage the child's environment (broadly defined and including hospitals, therapy settings and schools) in order to have the child's needs met. Both these areas of demand may be affected by the nature of the child's disability.

There is substantial evidence that the aetiology of the child's disability is an important variable in parental stress (Hodapp et al., 1998; Hodapp, Wijma, & Masino, 1997). Some disabilities are consistently associated with greater parental stress than others. For example, in a number of studies, parents of a child with Down syndrome have been found to experience lower levels of stress than do parents of children with autism (e.g., Hoppes & Harris, 1990; Kasari & Sigman, 1997). In a recent study comparing mothers of children with Down syndrome, fragile X syndrome, and autism, Abbeduto et al. (2004) found that mothers of a child with fragile X syndrome displayed lower levels of well-being than mothers of children with Down syndrome, but higher levels than mothers of children with autism.

It is likely that these differences in parental stress are due to differences in child attributes that are associated with the aetiology of the disability. Children with an intellectual disability present with more difficult behaviour than do typically developing children (Baker et al., 2002; Dekker, Koot, van der Ende, & Verhulst, 2002) and children with Down syndrome generally have fewer behaviour problems than children with an intellectual disability from other causes (Dykens & Kasari, 1997; Einfeld, Tonge, Turner, Parmenter, & Smith, 2000). This is not to suggest that behaviour is the only difference between these groups, nor that behavioural problems are the only cause of parental stress, however, behavioural problems are clearly one of the main predictors of stress in parents (Baker et al., 2002; Hastings, 2002).

In a recent study, the strongest and most consistent predictors of maternal depression and pessimism across three disability types (Down syndrome, fragile X syndrome and autism) were the extent and severity of the behavioural symptoms of their adolescent child (Abbeduto et al., 2004). The level of a child's adaptive behaviours has also been found to affect parental experience of stress (Hodapp, Wijma, et al., 1997; Tomanik, Harris, & Hawkins, 2004). Adaptive skills may also differentiate certain aetiologies (Walz & Benson, 2002) and thereby contribute to observed differences in parental stress across disability types.

Most investigations of parental functioning have focussed on mothers. Some studies have included either mothers or fathers as respondents but typically there is an overwhelming preponderance of mothers (e.g., Dyson, 1993; Fidler et al., 2000). While it remains the case that mothers are the primary caregivers in families where a child has a disability (Roach et al., 1999; Simmerman, Blacher, & Baker, 2001), it is increasingly clear that mothers and fathers respond differently to the experience of parenting a child with a disability (Cuskelly, Chant, & Hayes, 2004).

There is some inconsistency in the results of studies that have compared the levels of stress experienced by mothers and fathers. Kazak and Marvin (1984) and Sloper et al. (1991) found that mothers experienced a higher level of stress than fathers in families where there was a child with a disability. Other studies also support this finding, suggesting that mothers experience greater psychiatric morbidity and more symptoms of distress than fathers (Romans-Clarkson et al., 1986). More recent studies, however, have reported that the level of stress reported by mothers and fathers is very similar (Cuskelly et al., 2004; Keller & Honig, 2004).

Of more importance, when the direction of interventions is considered, is the evidence that the predictors of stress differ for mothers and fathers (Cuskelly et al., 2004; Dyson, 1997). For example, family cohesion and a child's adaptive skills predicted mother's but not father's stress in families where there was a child with Down syndrome (Cuskelly et al., 2004).

These issues may have particular importance in families with a child with fragile X syndrome as this is an X chromosome linked disorder. Mothers of children with fragile X syndrome are carriers of either the full mutation or premutation of the fragile X gene (Hagerman, 1999). Mild manifestations of the physical, behavioural and emotional characteristics associated with the full mutation of the fragile X gene have been described for a subgroup of those who have the fragile X premutation (Franke et al., 1998; Hagerman, 1996; Hagerman & Cronister, 1996; Sobesky et al., 1996). Consequently, mothers and other family members may possess manifestations of the syndrome which may themselves impact on levels of family and parent stress as both full mutation or premutation have a range of sequelae, including intellectual disability, difficulties in tasks requiring executive functioning, increased social anxiety, and higher rates of affective disorders (Franke et al., 1996; Hagerman & Hagerman, 2002; Mazzocco, 2000; Sobesky, Pennington, Porter, Hull, & Hagerman, 1994). These consequences are yet to be unambiguously documented. It is possible, however, that they will add to the difficulties of parenting for mothers, and increase the likelihood of differences between mothers and fathers in both levels of stress and the processes by which it operates.

The marital relationship has been identified as an important aspect of family functioning as it contributes to the individual psychological well-being of parents (Sloper et al., 1991). Early research suggested that parents with a child with a disability were likely to experience more marital dissatisfaction and separation than comparison families (Friedrich & Friedrich, 1981; Gath, 1977). More recent studies, however, have suggested that marital satisfaction is comparable across families with and without a child with a disability (see Scorgie & Sobsey, 2000 for a review).

The quality of the marital relationship has been found to be a significant predictor of parental stress where there is a child with a disability (Friedrich, 1979; van Lieshout, Meyer, Curfs, &

Fryns, 1998). Marital distress or dissatisfaction may be indicative of parental stress or be an additional stressor for the parent. Sloper et al. (1991) found that for fathers of a child with Down syndrome, the marital relationship was associated with stress indices such as satisfaction with life and psychosomatic symptoms. The marital relationship may also indirectly influence parental and family stress by affecting the parents' perceptions of the child with the disability. For example, Cuskelly and Dadds (1992) found marital satisfaction significantly predicted mothers' reports of problem behaviours in children with Down syndrome. A good marital relationship may be construed as a resource for the parent, as the support it provides may act to moderate the impact of child problems on the parent.

The availability and type of social support has been considered to be important to parents and families where there is a child with a disability (Hanson & Hanline, 1990). Support may have different effects, however, depending upon the aetiology of the child's disability. Hodapp et al. (1998) concluded that social support alleviated family stress where there was a child with Smith–Magenis syndrome, however, Hodapp, Dykens, et al. (1997) found no consistent relationship between social support and stress in families where there was a child with Prader–Willi syndrome. Social support is of particular interest in families with a child with fragile X syndrome as there is evidence that mothers in these families may have some difficulty with social interaction (Sobesky, Porter, Pennington, & Hagerman, 1995).

1. Aims of this study

The purpose of this study was to examine the contributors to the stress experienced by mothers and fathers of a child with fragile X syndrome. Several indicators of parental stress were measured, including psychological well-being, pessimism, and parents' views of parent and family problems. Demand on parents was construed as the child level of functioning with respect to level of behavioural problems, lack of adaptive skills, and the Physical Limitations and the Child Characteristics scales of the QRS-F. In addition, the study aimed to examine differences in the levels of stress between mothers and fathers as well as investigating the contributors to stress in each parental group. The role of marital satisfaction as a mediator of stress for both parents was of particular interest.

2. Method

2.1. Participants

The participants were 40 Australian families who had at least one child with fragile X syndrome between the ages of 4 and 18 years. The 40 families comprised 12 families from Queensland, 2 from New South Wales and 26 from Victoria. Sixty-seven parents (39 mothers and 28 fathers) from the 40 families were interviewed. Thirty-two of the families had both mothers and fathers present; however, only in 27 of these families did both the mother and father agree to participate. In the other five families only the mother participated. Eight families (20%) were headed by a sole parent (7 mothers and 1 father). The average age of mothers was 39.8 years (S.D. = 5.30, range = 31–53 years) and fathers had a mean age of 41.5 years (S.D. = 6.91, range = 29–58 years).

Thirty-one males and nine females diagnosed with fragile X syndrome were identified as target children for the parent interviews. The average age of the children was 10.4 years (S.D. = 3.59, range = 4–17 years). Sixty percent of the target children were on medication for their behaviour, including attentional problems and anxiety. The mean number of children per

family was 2.8 children (S.D. = 1.22, range = 1–6 children). Twenty-six families (65%) had only one child with the fragile X full mutation gene, 10 families (25%) had 2 children with fragile X, 3 families (7.5%) reported 3 children with fragile X, and 1 family (2.5%) had four children with fragile X syndrome. In addition, 3 families (7.5%) had 1 other child with the premutation and 1 family (2.5%) had 2 children with the premutation. Two of the families had another child who had a developmental disorder of a different aetiology.

2.1.1. *Diagnostic information*

Evidence of the fragile X status of the target child and biological mother was requested from each family but was not required for them to be included in the sample. From the information available, the mean age of the child at diagnosis of fragile X syndrome was 3.54 years (S.D. = 2.52, range = 6 months to 10 years). The mean number of years since the target child was diagnosed was 6.93 years (S.D. = 3.77, range = 1–16 years). Evidence that the target child had the fragile X full mutation gene was sighted for 33 (82.5%) families, with evidence not provided by the remainder.

Of the 39 mothers, 33 mothers (82.5%) reported that they had the fragile X premutation gene and 3 mothers (7.5%) reported they had the fragile X full mutation gene. Thirty mothers (75%) were able to provide diagnostic evidence of their fragile X status. Two mothers reported that they did not know their actual status and it was not relevant for a third, the non-biological mother.

2.2. *Instruments*

2.2.1. *Brief Symptom Inventory (BSI)*

The BSI (Derogatis, 1993) is a 53-item self-report scale that assesses the presence and frequency of psychological distress as evidenced by somatic and psychosomatic symptoms. The General Severity Index is an indicator of psychological stress. *T* scores are calculated using separate norms for males and females. Test–retest and internal consistency reliability have shown to be very good for the primary symptom dimensions of the BSI (Derogatis). *T* scores of 63 and above are considered to be in the clinical range (Derogatis).

2.2.2. *Questionnaire on Resources and Stress—Friedrich edition (QRS-F)*

The QRS-F (Friedrich, Greenberg, & Crnic, 1983) is a measure of perceived stress and coping that explores the impact of a child's disability or illness on other family members. It has a true/false response format for 52 items that yields four factor scores including Parent and Family Problems, Pessimism, Child Characteristics and the Physical Limitations of the child. A Total scale score is also calculated and serves as an index of parental stress. Higher factor scores indicate a higher level of family stress. The reliability of the total scale is .95 (Friedrich et al., 1983). Glidden (1993) pointed out that two of the scales, Child Characteristics and Physical Limitations were more properly understood as demands rather than as stress (or strain as she labelled it). We have adopted this approach and therefore do not use the Total score although we do report them in Table 1 to allow comparison with other studies to be made. The scales titled Parent and Family Problems and Pessimism are used as indicators of parental stress and the scales called Child Characteristics and the Physical Limitations are used as indicators of demand, and therefore as possible predictors of stress.

2.2.3. *Behavior Assessment System for Children—Parent Rating Scale (BASC-PRS)*

The BASC-PRS (Reynolds & Kamphaus, 1998) is used to assess problem and adaptive behaviour in children on a number of dimensions of behaviour. These are then grouped into

Table 1
Means and standard deviations for mothers and fathers (including both married and sole parents)

Measures	Mothers			Fathers		
	<i>n</i>	<i>M</i>	S.D.	<i>n</i>	<i>M</i>	S.D.
QRS-F Total	39	19.87	9.11	28	18.18	8.55
QRS-F Parent and Family	39	6.66	4.55	28	5.04	4.29
QRS-F Pessimism	39	6.15	2.53	28	6.21	2.46
QRS-F Child Characteristics	39	5.97	2.92	28	5.89	2.44
QRS-F Physical Limitations	39	1.08	0.87	28	1.04	1.11
BSI	39	52.77	9.93	27	54.93	9.14
FSS Number of Supports	39	12.83	2.20	28	13.68	2.36
FSS Satisfaction	39	28.18	10.60	28	26.32	9.79
BASC-PRS Internalizing	39	51.16	9.57	28	50.74	9.19
BASC-PRS Externalizing	39	55.61	9.07	28	56.19	12.25
BASC-PRS Behavioral Index	39	60.36	8.64	28	60.46	9.67
BASC-PRS Adaptive Skills	39	35.36	8.19	28	35.29	6.9
DAS	32	49.31	9.74	27	50.00	6.52

composites of Internalising and Externalising Problems, a Behavioral Index and Adaptive Skills. The BASC has recording forms for three different age levels—preschool (2.5–5 years), child (6–11 years); and adolescent (12–18 years). The age-appropriate form for each child was selected for use in the interview. Reliability of the composite scores range from the mid .80s to the low .90s at all three age levels (Reynolds & Kamphaus).

On the Internalizing and Externalizing scales and the Behavioral Index, standardized scores between 41 and 59 are classified as average, between 60 and 69 are classified as at-risk, and scores of 70 and above are classified as clinically significant. On the Adaptive Skills composite, standardized scores are classified as average when between 41 and 59, as at-risk when between 31 and 40, and as clinically significant if 30 or below.

2.2.4. Dyadic Adjustment Scale (DAS)

The DAS (Spanier, 1989) is a 32-item scale, the total of which provides a measure of satisfaction with the marital relationship. Raw scores are converted to *T* scores. The total scale has an internal consistency reliability of .96 (Spanier, 1976 in Spanier, 1983). Higher scores suggest better adjustment, with *T* scores below 30 being indicative of distress and discord in the marital relationship.

2.2.5. Family Support Scale (FSS)

The FSS (Dunst, Jenkins, & Trivette, 1984) is an 18-item scale which determines how helpful (i.e., supportive) 18 sources of support have been to families with children with a disability during the preceding 6 months. A coefficient alpha computed from the average correlation among the 18 scale items was .77 and the test–retest reliability of the total score was established at .91 (Dunst et al., 1984).

2.3. Procedure

The study was approved by the University of Queensland's Ethics Committee for Research with Human Subjects prior to recruitment of participants. Families were recruited through the Hear: I Am Queensland Fragile X Association Inc. in Queensland, and the Fragile X Alliance

Clinic and Fragile X Support Group Inc. in Victoria, Australia. These organizations agreed to send invitations to participate in the study to their members/clients.

Families were invited to participate if they had at least one child with fragile X syndrome between the ages of 4 and 18 years. Families who responded to the invitation to participate in the study were telephoned by the first author to establish family contact details, marital status, and the age and fragile X status of the children. In those families where there was more than one child with fragile X syndrome in the target age range parents were asked to nominate the child with fragile X syndrome who presented with the most difficult behaviour, or if this was not appropriate, to nominate the child with the highest support needs. Families were also requested, where it was possible, to locate copies of the diagnostic information to confirm that their child had fragile X syndrome, and to confirm fragile X status of the mother if she was to be interviewed.

Face-to-face interviews with parents were conducted by the first author and were completed in the family home, the Fragile X Alliance Clinic in Melbourne, Victoria or in a location most convenient to the parent, such as a cafe or their place of employment. Mothers and fathers completed all questionnaires independent of each other during the interview. Parents were presented with the five questionnaires in the following order: the BASC-PRS, the FSS, the DAS (which was not included for sole parents), the QRS-F, and the BSI.

Demographic information was collected at the end of the interview. Parents were asked for details of their age, parental and marital status, educational level, employment, and the availability of diagnostic information. Parents were also asked about family characteristics such as family income, other children and their fragile X status, whether others resided with the family and medications used by the target child.

3. Results

Parents of one family did not complete the BASC-PRS scale for their child when interviewed and therefore mean scores for the subscales were substituted. One father completed the BSI questionnaire, providing a rating of zero for all items. This was considered an invalid administration (Derogatis, 1993) and the fathers' mean score was substituted. Data provided by mothers and by fathers were examined separately and all scales were found to have normal distributions.

Not all mothers in the study had partners who participated (some because they were sole parents and some because the fathers refused), therefore the mother/father correlations and the mother/father comparisons used data only from those families in which both parents participated. In all analyses $p < .01$ was used to indicate significance in order to reduce the possibility of Type 1 error.

After descriptive information had been determined, parents' agreement and differences on both dependent and independent variables were examined. This was followed by a series of hierarchical regression analyses that examined the predictors of the dependent variables, by parent group. Finally, an exploratory investigation of differences between married and sole mothers was undertaken.

Correlations between demographic variables (child age, number of children living at home, mother and father education and occupation, and family income) showed only one significant association which was between child age and fathers' score on the QRS-F subscale of Physical Limitations ($\rho = -.50, p < .01$). In the light of this, demographic information was ignored in subsequent analyses. Finally, a comparison of all dependent and independent measures was undertaken for those children who were on medication and those who were not.

Table 2

Number (percentage) of children with fragile X syndrome in the clinically significant and at-risk mutually exclusive categories on maternal and paternal report on the BASC-PRS Subscales

Subscales	Mothers (<i>n</i> = 39)		Fathers (<i>n</i> = 28)	
	Clinically significant	At-risk	Clinically significant	At-risk
Internalizing	2 (5%)	6 (15.4%)	1 (3.6%)	4 (14.3%)
Externalizing	2 (5%)	11 (28.2%)	3 (10.7%)	7 (25%)
Behavioral Index	6 (15.4%)	15 (38.5%)	4 (14.3%)	12 (42.9%)
Adaptive Skills	10 (25.6%)	21 (53.9%)	7 (25%)	13 (46.4%)

There were no differences between the groups on any measure and therefore groups were combined for all analyses.

3.1. Descriptive information

Table 1 contains the mean scores for mothers and fathers. Data from all mothers who contributed to the study are included in these values. As there were very few differences between married and single mothers (see below) they were combined in this table. Using the suggested cut-offs described in Section 2, it is clear that the children with fragile X syndrome, as a group, were in the at-risk category on the BASC-PRS Behavioral Index, although they did not meet the criterion for this classification on either the Internalizing or Externalizing scales. Their parents' ratings also placed them in the at-risk category for their Adaptive Skills. Table 2 shows the numbers of children whose score placed them in the at-risk or clinically significant categories on the basis of mothers' and fathers' reports.

The scores of only two mothers were in the distressed range on the DAS. The mean scores for this scale indicate that both mothers and fathers were similar to other couples in their level of marital satisfaction. Six (15.4%) mothers and 5 (17.9%) fathers were in the clinical range on the BSI General Severity Index. The means of the groups, however, suggest that, on average, these parents were not distressed. There are no interpretative guidelines for the QRS-F, however, a number of previous studies of stress in parents of children with a disability have found mean total scores between 14.95 and 25.14 (Dyson, 1991; Friedrich et al., 1983; Hodapp et al., 1998; Hodapp, Dykens, et al., 1997). The parents in this study had scores at the lower end of this range.

3.2. Mother and father differences

Repeated measures ANOVA was used to test for significant differences between mothers and fathers on all dependent and independent variables. Gender of the child was not included in this analysis as there were only four girls for whom data from both parents were available. There were no significant differences between parent groups on any of the measures used in the study.

3.3. Associations between measures

Correlations between measures are presented separately for mothers and fathers in Tables 3 and 4, respectively. The subscales of the QRS-F were generally moderately to highly

Table 3
Correlations between maternal variables

	1	2	3	4	5	6	7	8	9	10	11	12	13
1 QRS-F Total	1												
2 QRS-F Parent and Family Problems	.89***	1											
3 QRS-F Pessimism	.79***	.51**	1										
4 QRS-F Child Characteristics	.85***	.62***	.63***	1									
5 QRS-F Physical Limitations	.66***	.49*	.58***	.50**	1								
6 BSI	.51**	.56***	.40*	.33	.35	1							
7 FSS Number of Supports	-.28	-.35	-.18	-.10	-.27	-.15	1						
8 FSS Satisfaction with Support	-.44*	-.35	-.41*	-.37	-.29	-.20	.54***	1					
9 BASC-PRS Internalizing	.28	.32	.19	.21	.01	.21	-.16	-.05	1				
10 BASC-PRS Externalizing	.23	.27	.27	-.16	-.16	.38	-.05	-.27	.16	1			
11 BASC-PRS Behavioral Index	.49**	.40*	.44*	.47*	.19	.33	-.06	-.30	.66***	.60***	1		
12 BASC-PRS Adaptive Skills	-.41**	-.26	-.41*	-.42*	-.26	-.26	.10	.29	-.21	.10	-.20	1	
13 DAS	-.11	-.31	-.05	-.17	-.21	-.50	.17	.19	-.03	-.08	-.09	-.05	1

* $p < .01$.

** $p < .001$.

*** $p < .0001$.

intercorrelated for both groups, with the exception of the Physical Limitations subscale for fathers which was unrelated to all other measures on the QRS-F. Maternal scores on the Parent and Family Problems and Pessimism subscales of the QRS-F were significantly correlated with the BSI. Fathers' scores on the Parent and Family Problems subscale were significantly correlated with the BSI, but the correlation with the Pessimism subscale did not reach significance.

The Child Characteristics subscale of the QRS-F was significantly correlated with the BASC-PRS Behavioral Index and Adaptive Skills composite for mothers but not for fathers. Satisfaction with Supports was positively correlated with Number of Supports for mothers, but was unrelated for fathers. For fathers, children's Internalizing Problems and the Behavioral Index on the BASC-PRS were positively correlated with the General Severity Index of the BSI, indicating that fathers experienced more stress when their children were exhibiting behavioural difficulty. Surprisingly, this association was not evident for mothers. The QRS-F Pessimism subscale was negatively correlated with marital satisfaction as measured by the DAS for fathers, but there were no significant correlations for mothers between marital satisfaction and other measures.

There were few significant associations between parental reports. All those that reached significance are reported below. Parents' reports of children's Externalizing Problems ($r = .54$, $p < .01$), and Adaptive Skills were significantly positively correlated ($r = .64$, $p < .001$). Mothers' and father's Dyadic Adjustment Scale scores were positively correlated ($r = .77$, $p < .001$) and their reports of the stress with respect to their child's Physical Limitations were also positively correlated ($r = .61$, $p < .001$).

Table 4
Correlations between paternal variables

	1	2	3	4	5	6	7	8	9	10	11	12	13
1 QRS-F Total	1												
2 QRS-F Parent and Family Problems	.92***	1											
3 QRS-F Pessimism	.85***	.69***	1										
4 QRS-F Child Characteristics	.86***	.72***	.67***	1									
5 QRS Physical Limitations	.34	.13	.20	.29	1								
6 BSI General Severity Index	.45	.56*	.45	.10	.01	1							
7 FSS Number of Supports	-.12	-.09	-.09	-.21	.09	.17	1						
8 FSS Satisfaction with Support	-.44	-.42	-.37	-.45	.03	-.16	.15	1					
9 BASC-PRS Internalizing	.41	.50*	.38	.21	-.06	.63**	-.60	-.04	1				
10 BASC-PRS Externalizing	.29	.38	.16	.21	-.06	.37	.07	.16	.39	1			
11 BASC-PRS Behavioral Index	.45	.50*	.38	.28	.07	.65***	.11	-.00	.82***	.66***	1		
12 BASC-PRS Adaptive Skills	-.26	-.13	-.24	-.43	-.03	.35	.23	.12	-.15	-.03	-.12	1	
13 DAS	-.33	-.31	-.57*	-.10	.11	-.37	.10	-.02	-.06	-.17	-.10	-.10	1

* $p < .01$.

** $p < .001$.

*** $p < .0001$.

Prior to other regression analyses being undertaken, a series of regression analyses was used to examine the associations between marital satisfaction and the other independent variables for both parents in order to establish if the necessary conditions for a mediating role for marital satisfaction were present. There were no significant associations between these measures for either parent. The basic requirement for marital satisfaction to be acting as a mediator between child behaviours and characteristics and parental stress was therefore absent (Baron & Kenny, 1986).

3.4. Regression analyses for mothers

A series of hierarchical multiple regressions was carried out for each parent group separately, examining the contributions of child behaviour (BASC-PRS subscales of Internalizing, Externalizing and Adaptive Skills), child attributes (Child Characteristics and Physical Limitations subscales of the QRS-F), family support (FSS Number of Supports and Satisfaction) and marital satisfaction (DAS) to stress. The independent variables were the General Severity Index of the BSI and the two subscales of Parent and Family Problems and Pessimism from the QRS-F. Data were entered into blocks in the order described above. Child behaviour was entered first as it was expected to be the most influential of the predictors, followed by the child attributes. Family support was entered next as these data were available for all parents, and marital satisfaction was entered as the final block, as these scores were available only for married parents.

For mothers, only the final model was significant $F(8,22) = 3.47, p < .01$, when BSI was the dependent variable [R^2 change $F(1,22) = 10.95, p < .01$]. The complete model accounted for 40% of the variance. Mothers' marital satisfaction was the only significant individual predictor ($\beta = -.50, t = -3.32, p < .01$).

When QRS-F Parent and Family Problems was the dependent variable, the model became significant at the second step $F(5,25) = 6.07, p < .001$ [R^2 change $F(2,25) = 6.07, p < .01$] with no improvement in the adjusted R^2 with the addition of other predictors. Forty-six percent of the variance was accounted for by the first two steps in the regression, with 33.5% attributable to the addition of the child attributes. No individual predictor contributed at the $p < .01$ level.

The same pattern applied for the Pessimism subscale, with the model becoming significant with the addition of the second step $F(5,25) = 9.74, p < .001$ [R^2 change $F(3,25) = 12.67, p < .001$] with no improvement in the adjusted R^2 with the addition of other predictors. Fifty-nine percent of the variance was accounted for by the first two steps in the regression, with 34% attributable to the addition of child attributes. The Physical Limitations scale was the only significant individual predictor ($\beta = .53, t = 3.81, p < .001$) with Adaptive Skills almost reaching significance ($\beta = -.31, t = 2.63, p = .015$).

3.5. Regression analyses for fathers

For fathers, the best predictor of BSI General Severity Index was child behaviour $F(3,21) = 8.95, p < .001$. These scores accounted for 50% of the variance and there was no significant improvement to prediction by adding in the next three blocks. Two individual measures reached significance. These were Externalizing Behavior ($\beta = .59, t = 4.00, p < .001$) and the measure of Adaptive Skills ($\beta = .48, t = 3.30, p < .01$).

The model became significant at the addition of the second step for QRS-F Parent and Family Problems $F(5,20) = 5.96, p < .01$ with an adjusted R^2 of .50 [R^2 change $F(2,20) = 12.01, p < .001$]. The addition of other elements did not improve the prediction.

The entire model significantly predicted QRS-F Pessimism for fathers $F(8,17) = 10.43, p < .001$, accounting for 75% of the variance. The model became significant at the entry of the

Table 5
Means and standard deviations for married and sole mother groups

Measures	Married mothers ($n = 32$)		Sole mothers ($n = 7$)	
	<i>M</i>	S.D.	<i>M</i>	S.D.
QRS-F Total	18.31	9.02	27.00	5.69*
QRS-F Parent and Family Problems	5.78	4.32	10.71	3.35**
QRS-F Pessimism	5.84	2.61	7.57	1.51
QRS-F Child Characteristics	5.63	2.96	7.57	2.30
QRS-F Physical Limitations	1.06	0.91	1.14	0.69
BSI General Severity Index	51.19	9.77	60.00	7.52*
FSS Number of Supports	13.03	2.24	12.00	1.91
FSS Satisfaction	29.63	10.60	21.57	8.28
BASC-PRS Internalizing	50.03	8.11	56.14	14.11
BASC-PRS Externalizing	55.35	8.91	56.71	10.39
BASC-PRS Behavioral Index	59.34	8.32	65.00	9.20
BASC-PRS Adaptive Skills	35.94	7.73	32.71	10.31

* $p < .05$.

** $p < .01$.

second block $F(5,20) = 5.09$, $p < .01$ [R^2 change $F(2,20) = 8.61$, $p < .01$] and prediction was again significantly improved with the addition of marital satisfaction [R^2 change $F(2,18) = 25.04$, $p < .001$]. In the final model, individual predictors were child attributes ($\beta = .49$, $t = 3.51$, $p < .01$) and marital satisfaction ($\beta = -.53$, $t = -5.00$, $p < .001$).

3.6. Married versus sole mothers

In order to explore differences between married and sole mothers independent t -tests were undertaken with both dependent and independent variables. Table 5 contains the means and standard deviations for both groups. Only the QRS-F Parent and Family subscale reached significance ($t(37) = -2.83$, $p < .01$), with the BSI General Severity Index approaching significance ($t(37) = -2.24$, $p = .03$). In these two instances, single mothers were more stressed than married mothers.

4. Discussion

The results of this study reveal that children with fragile X syndrome are clearly at risk of exhibiting problem behaviour and are also likely to have deficits in their adaptive skills. Children's mean score for the Behavioral Index of the BASC-PRS was in the at-risk category and more than 50% of the group (53% for mothers, 67% for fathers) had behaviour that was in either the at-risk or clinically significant categories. The data on adaptive skills tells an even more alarming story with 89% on mothers' report and 71% on father's report either being in the at-risk or clinically significant categories. These results need to be considered in the context of 60% of the children being on medication intended to influence their behaviour. They reflect other studies that have concluded that children with fragile X syndrome have increased behaviour problems and lower levels of adaptive behaviour than typically developing children (Hatton et al., 2002; Hatton et al., 2003).

There were no significant differences between mothers and fathers in their reports of any of the measures of child problems or adaptive behaviour. In many past studies, parental differences have been reported, however, a lack of difference between mothers and fathers has been found in several recent studies (Cuskelly et al., 2004; Dyson, 1997). This change may reflect changes in parenting practices, with fathers now taking more responsibility in child care matters and thus having a similar view of their children's behaviour to that of mothers.

If this were the case, it might be expected that reports of stress would not differ between groups, and that similar associations between child behaviour and parental stress would be found for both groups. The latter issue will be discussed below. No differences in levels of stress were found when mothers' and fathers' reports were compared. This was true for all three measures used in this study to assess stress. This is a particularly interesting finding as we had speculated that mothers would report higher stress, as this is a common finding and also as the premutation or mutation might be expected to have a negative impact on mothers' ability to deal with the increased demands associated with their child's care.

When parents are considered as a group, they appear to be managing relatively well. The mean score on the BSI was not in the clinically significant range for either mothers or fathers. Fifteen percent of mothers and 18% of fathers were in the clinically significant range on the instrument, however, clearly higher than would be expected in a general population sample (Derogatis, 1993). This finding for mothers is consistent with recent research by Abbeduto et al. (2004) who found that 18.2% of mothers of a young person with fragile X syndrome had levels of depressive symptoms above the cut-off for clinical depression.

Parents had similar levels of marital satisfaction to the general population, with only two mothers being significantly distressed in their marriages. There was no difference between mothers and fathers in their satisfaction levels. This lack of difference also applied to the number and satisfaction with social supports for the family. In other studies, mothers have generally reported more social supports than fathers (e.g., [Gavidia-Payne & Stoneman, 1997](#)).

All three measures of parental stress were significantly correlated for mothers. The same was true for fathers with the exception that the correlation between the Pessimism subscale and the BSI, although moderate, did not reach significance. This convergence of data provides some validity for the use of the Parent and Family Problems and Pessimism subscales of the QRS-F as measures of stress in family research ([Glidden, 1993](#)).

There were some notable differences between parent groups when predictors of stress were identified. Forty percent of the variance of mothers' psychological well being, as measured by the BSI, was accounted for by the full model. The expectation that behavioural problems would be a significant predictor of psychological stress ([Hodapp, Dykens, et al., 1997](#); [Hodapp et al., 1998](#); [Hodapp, Wijma, et al., 1997](#)) was not met in relation to the mother's experience. Interestingly, the child's level of externalizing problems and adaptive skills were important individual contributors to fathers' well-being. This suggests that in these families there may be some reversal of typical family roles, with fathers taking a major role in child related responsibilities. Marital satisfaction was the only significant individual predictor of mothers' general psychological well being.

The combination of child behavioural problems and child attributes was significant for both mothers and fathers when stress related to parent and family problems was examined. There were no individual predictors for either parent group in these regressions. This same combination was significant for Pessimism for both parent groups, although marital satisfaction accounted for additional variance for fathers. Social support played no role in the regression analyses for either mothers or fathers. Research generally finds that social support is an important variable in reducing stress in mothers ([Honig & Winter, 1997](#)). The failure to find such an effect here may reflect certain maternal characteristics such as shyness associated with the full mutation or permutation in the mothers of children with fragile X.

For fathers, the most significant predictor of psychological stress was the child's behaviour and, in particular, the child's adaptive skills. This finding is only partially reflective of previous research where levels of adaptive behaviours have been found to affect parent stress ([Hodapp, Wijma, et al., 1997](#)). This relationship between adaptive behaviour and stress was found to apply to mothers by [Abbeduto et al. \(2004\)](#), who found that mothers of a young adult with fragile X syndrome reported higher scores on the problem behaviour measure which significantly predicted maternal pessimism on the QRS-F subscale. In contrast to our findings, however, [Abbeduto et al. \(2004\)](#) found that these behaviours also significantly predicted mothers' depressive symptoms.

While variance explained by the independent variables in this study is very high, there remains a substantial proportion of variance still to be explained. The data gathered in this study focussed solely on family matters, yet all parents have other responsibilities and relationships that may cause stress. In interview, a number of fathers spontaneously remarked that the demands associated with work caused them more stress than those associated with their child.

Fathers were found to report greater psychological stress with increasing levels of problem behaviours in their children, however, this did not apply to mothers. Given that over 75% of children in the sample were boys, this may suggest that fathers find the demands associated with problem behaviours of their sons more stressful than do mothers. The differences between mothers and fathers and their perceptions of their same gender children warrants investigation in future research studies.

As found in other studies (e.g., Olsson & Hwang, 2001), mothers who were sole parents reported a higher level of psychological stress than married mothers, although their mean score was within the average range. Although the sample was small, this trend suggests the need for a more thorough examination of outcomes for sole parent families. In addition, processes associated with stress need to be understood for this group if appropriate supports are to be offered.

4.1. *Limitations of the study*

In this study, parents were asked to respond with respect to the child with most problem behaviour or highest support needs. Therefore, the data about levels of problem behaviour and adaptive skills will likely reflect the extremes of the group, and may not be representative. Care should thus be taken when generalizing to other children with fragile X. The volunteer nature of the sample must also be considered, along with the possibility that families who agree to participate in research are those whose children are functioning relatively well (Cuskelly, 2005).

The number of children with fragile X in the family is an important issue to consider for future research. Parents who have more than one child with fragile X are likely to be subjected to increased strain. The nature of transmission of this disability, however, makes this a probable circumstance for families. Alternatively, having more than one child in the family with fragile X syndrome may result in a normalizing of the associated difficulties, thus reducing the stressful aspects for parents. While data analytic techniques that can accommodate this complexity have been developed, the data collection can be daunting for families as information has to be supplied for each child with fragile X syndrome.

4.2. *Conclusion*

Parents of a child with fragile X were found to have reasonably satisfactory marriages and most were functioning adequately with respect to mental health. Their levels of stress were at the lower end of the range reported for other families with a child with a disability. There were no significant differences between mothers and fathers when reports of their children were compared or in the levels of stress they reported. There were some differences between mothers and fathers in the processes found to contribute to stress, and these processes were somewhat different from those reported elsewhere. It is possible that the dynamics differ in families with a child with fragile X, possibly partly as a result of mothers' genetic status. This latter point is purely speculative and needs further research.

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