Psychological well-being of mothers of youth with fragile X syndrome: syndrome specificity and within-syndrome variability

P. Lewis,[1] L. Abbeduto,[1] M. Murphy,[2] E. Richmond,[1] N. Giles,[1] L. Bruno,[1] S. Schroeder,[1] J. Anderson[1] & G. Orsmond[3]

1 *Waisman Center, University of Wisconsin-Madison, WI, USA*
2 *Kennedy Krieger Institute and the Johns Hopkins University, Baltimore, MD, USA*
3 *Boston University, Boston, MA, USA*

Abstract

Background   Research on parental well-being has focused largely on Down syndrome and autism; however, fragile X syndrome is likely to pose different challenges for parents compared with these other diagnostic conditions. Moreover, there is considerable variability among youth with fragile X syndrome; for example, 25% to 33% of affected youth meet criteria for a co-morbid diagnosis of autism. It is likely that parents of youth with fragile X syndrome will experience different degrees and patterns of stress, depending on whether their offspring do or do not have a co-morbid diagnosis of autism. In the present study, we compared mothers of three groups of young males on measures of psychological well-being and stress: those with fragile X syndrome and a co-morbid diagnosis of autism; those with fragile X syndrome alone; and those with Down syndrome.
Method   The sample consisted of mothers of adolescent and young adult males with fragile X syndrome and co-morbid autism ($n = 9$), fragile X syndrome alone ($n = 19$), and Down syndrome ($n = 19$). We screened all youth for autism using the Autism Behavior Checklist, which was completed by mothers, fathers and teachers, and the youth who scored above the suggested cut-off were evaluated by a licensed psychologist to determine autism status. The three groups of youth did not differ in chronological age (16.4, 15.8 and 16.0 years, respectively) or nonverbal mental age (3.8, 3.9 and 3.8 years, respectively). Several self-report measures were completed by mothers. These measures assessed current mental health status (e.g. the Center for Epidemiological Studies Depression Scale), perceptions of their son's and family's functioning (e.g. the Positive Affect Index, which measures closeness felt by the mother to her son and also reciprocated closeness felt by the son towards the mother, as perceived by the mother), and approach to coping with their son's disability [e.g. the Multidimensional Coping Inventory (COPE), which measures emotion-focused and problem-solving focused coping].
Results   The results suggest that fragile X syndrome creates more challenges to maternal psychological well-being than Down syndrome, and that the combination of fragile X syndrome and autism can be particularly challenging. Differences among groups, however, were manifested mainly as concerns about the affected son and about relationships within the family rather than as lower levels of mental health. Thus, mothers of sons with fragile X syndrome, regardless of the son's autism status, reported more pessimism about the son's future and more conflict within the family than mothers of sons with Down

Correspondence: Dr Pamela Lewis, 1500 Highland Avenue, Madison, Wisconsin, USA (e-mail: lewis@waisman.wisc.edu).

syndrome. Additionally, mothers of sons with fragile X syndrome and co-morbid autism reported lower levels of reciprocated closeness than the other two groups of mothers.

Conclusion   We consider possible causes of these maternal differences, the implications for clinical practice, needs for future research, and the importance of understanding child and contextual factors as well as the dynamics leading to these differences.

Keywords autism, Down syndrome, fragile X syndrome, intellectual disability, mental health, parents

Introduction

Mothers of children with developmental disabilities face challenges that vary with the nature of the child's disability (Ricci & Hodapp 2003). Most studies in this area, however, have involved comparisons of mothers of individuals with Down syndrome with mothers of individuals with autism or mothers whose children were, as a group, heterogeneous with respect to aetiology (Dunst *et al.* 1986; Marcovitch *et al.* 1986; Walker *et al.* 1992; Kasari & Sigman 1997; Ly & Hodapp 2002). This research has indicated that mothers of youth with Down syndrome generally display high levels of well-being, whereas mothers of youth with autism fare very poorly relative to mothers raising children with other forms of developmental disabilities. In the study reported here, we focused on mothers of youth who have fragile X syndrome.

Mothers of individuals with fragile X syndrome display relatively high levels of parenting stress (Sarimski 1997) and low levels of psychological well-being (e.g. more depressive symptoms) compared with mothers parenting an individual with Down syndrome (Abbeduto *et al.* 2004). In fact, on some dimensions of psychological well-being and psychopathology, mothers of youth with fragile X syndrome fare as poorly as mothers of youth with autism (Franke *et al.* 1996; Abbeduto *et al.* 2004). Mothers of youth with fragile X syndrome may be at risk simply by virtue of carrying the premutation of the gene responsible for the syndrome, which has been shown to render some women without affected children vulnerable to various forms of psychopathology relative to the general population (e.g. Sobesky *et al.* 1994, 1996; Franke *et al.* 1996; Johnston *et al.* 2001; Good-

lin-Jones *et al.* 2004; Hessl *et al.* 2005). There is also evidence, however, that various characteristics and behaviours of their children contribute to the lower well-being of these mothers (Abbeduto *et al.* 2004).

Because individuals with fragile X syndrome are not a homogeneous group, parenting challenges will differ across children. Although many individuals with fragile X syndrome display behaviours resembling those of autism, only 25% to 33% meet criteria for a co-morbid diagnosis of autistic disorder (Rogers *et al.* 2001; Demark *et al.* 2003; Bailey *et al.* 2004). These two subgroups of individuals with fragile X syndrome differ not only in the presence of a co-morbid diagnosis, but also in the severity of their impairments, with youth who have co-morbid diagnoses of fragile X syndrome and autism having more limited cognitive skills than youth with either single diagnosis (Hagerman *et al.* 1986; Cohen 1995; Bailey *et al.* 2001; Kau *et al.* 2004; Kaufmann *et al.* 2004; Philofsky *et al.* 2004; but see Reiss & Freund 1990 for an exception). Those in the co-morbid group are also characterized by receptive language and 'theory of mind' reasoning that are below mental age levels compared with individuals with only fragile X syndrome (Philofsky *et al.* 2004; Lewis *et al.* 2006).

It is reasonable to expect, given the impact of the foregoing behavioural impairments on daily functioning and the prognosis for independent living (Hodapp 1997), that youth with co-morbid fragile X syndrome and autism will pose more parenting challenges than youth with only fragile X syndrome. At the same time, however, children who are more severely affected, as in the case of those with co-morbid fragile X syndrome and autism, may come to the attention of health professionals earlier and, as a result, receive earlier and more extensive services than children with only fragile X syndrome, which might help reduce parenting challenges. Moreover, it is not known whether there are genetic differences between these two subgroups of mothers that could make them differentially susceptible to the stress associated with parenting. Thus, the comparison of mothers parenting individuals with fragile X syndrome with and without a co-morbid autism diagnosis may provide insights into the relative contributions of child, contextual, and maternal variables to maternal well-being.

The study reported here had two goals. The first was to determine whether there are differences in

psychological well-being between mothers parenting individuals with fragile X syndrome and a co-morbid autism diagnosis and mothers parenting individuals with only fragile X syndrome. We also included a comparison group of mothers of individuals with Down syndrome, as a way of facilitating interpretation of the findings for the two groups of mothers parenting youth with fragile X syndrome. Previous research suggests that mothers in both fragile X syndrome groups will fare more poorly in terms of psychological well-being than those in the Down syndrome group. The second goal was to determine what effect the youth's autism symptomatology has on maternal psychological well-being. In addressing these goals, we focused on youth at the ages of 10–23 years. This age period is one in which parenting stress may be especially acute as there are issues of puberty and its attendant impacts, transition planning for life after school (Lueckling & Fabian 1997), and declining cognitive level (IQ) in individuals with fragile X syndrome (Dykens *et al.* 2000). We should note that our focus on mothers is not meant to imply that fathers are not affected by the characteristics of their children – indeed, there is empirical evidence of a link between paternal well-being and child functioning for fragile X syndrome (Hessl *et al.* 2001); instead, our focus reflects the fact that despite changing gender roles, mothers still tend to be the primary caregivers.

Method

The present study was based on data collected in a larger ongoing study of individuals with either fragile X or Down syndrome and their parents (Abbeduto *et al.* 2003, 2004; Abbeduto & Murphy 2004; Lewis *et al.* 2006). The sample for the present study was a subset from the larger study, and consisted of mothers of youth with fragile X syndrome and co-morbid autism ($n = 9$), fragile X syndrome only ($n = 19$), or Down syndrome ($n = 19$). In cases in which a family had two children in the age range of interest, only one (randomly determined) was included in the present analyses. Two mothers of participants with co-morbid fragile X syndrome and autism were the adoptive parents of the target youth, and one mother in the fragile X syndrome only group was a stepparent; the remaining mothers were the biological parents of the target youth.

Sample

Recruitment

We recruited families locally through newspaper advertisements and mailings to families enrolled in a university research registry and, particularly for fragile X syndrome, nationally through Internet postings and newsletters of national developmental disabilities organizations. Ten of the mothers of individuals with Down syndrome and 10 of the mothers of individuals with only fragile X syndrome were also included in the sample of Abbeduto *et al.* (2004); however, the present analysis includes additional measures.

Confirmation of fragile X and Down syndrome

The parents of all participants with Down syndrome reported aetiology as trisomy 21, with medical reports confirming the karyotype available for all but four of these participants. Reports of DNA confirmation of the full mutation were available for all but three of the participants with fragile X syndrome; only cytogenetic confirmation was available for two and only parent report of DNA confirmation for the third. All participants in the two fragile X syndrome samples had the full mutation, or mosaicism (i.e. with different cells having either the premutation or full mutation). Most of the mothers in the fragile X syndrome groups had not had genetic testing completed on themselves; thus, maternal carrier status was not included as a variable in this study. In light of the fact that measures of fragile X mental retardation-1 (FMR1) variation are correlated with various cognitive and mental health measures, even among those with the premutation, this reflects a limitation of the present study.

Confirmation of autism

Participating youth with fragile X or Down syndrome were evaluated for autism using a two-step procedure (see Lewis *et al.* 2006 for more details). In Step 1, the informant-report Autism Behavior Checklist (ABC; Krug *et al.* 1980) was completed by the youth's teacher, mother and (in two-parent families) father. In Step 2, youth meeting screening criteria on the ABC were evaluated against Diagnostic and Statistical Manual of Mental Disorders – 4th edition (DSM-

IV; American Psychiatric Association 1994) criteria by a licensed psychologist (P.L.), who took a developmental history from the mother, observed the youth interacting with the mother, and interacted with the youth directly.

Approximately one-fourth of the youth with fragile X syndrome participating in the larger project were found to have autism (see Lewis *et al.* 2006). No youth with Down syndrome included in this sample met criteria for autism.

Characteristics of youth

In several previous studies, it has been found that youth with co-morbid fragile X syndrome and autism have lower IQs on average than youth with fragile X syndrome or autism alone (e.g. Demark *et al.* 2003; Philofsky *et al.* 2004). In our previous research with this sample (Lewis *et al.* 2006), we found that all of the participants with co-morbid fragile X syndrome and autism achieved the lowest standard score possible (i.e. 36) based on performance on three non-verbal subtests from the Stanford-Binet Intelligence Scale – fourth edition (Stanford-Binet; Thorndike *et al.* 1986): Copying, Pattern Analysis, and Bead Memory. To ensure that diagnostic group was not confounded with IQ in the present study, we selected only those participants with Down or fragile X syndrome from the larger study who also had a standard score of 36 on the three Stanford-Binet subtests. We also selected participants from the latter two groups to ensure that they were not significantly different in chronological age from the participants with co-

morbid fragile X syndrome and autism ($F_{2, 44} = 0.100$, $P < 0.905$). The three groups also were not significantly different in terms of non-verbal mental age, determined by the three Stanford-Binet subtests ($F_{2, 44} = 0.065$, $P < 0.937$). Finally, only males were found to have co-morbid fragile X syndrome and autism in our sample and thus, only males were included in the present study. (See Table 1 for sample characteristics.)

Characteristics of mothers

Across the three groups, the mothers did not differ on level of education (college or higher vs. high school or less, $\chi^2 = 1.36$, $P < 0.51$) or family income (categorized into 11 levels, $F_{2, 43} = 0.07$, $P < 0.93$), and all but one mother was Caucasian. There was a marginally significant difference in maternal age ($F_{2, 44} = 3.05$, $P = 0.06$), reflecting the fact that, as might be expected, the mothers of sons with Down syndrome were somewhat older than the other mothers. (See Table 2 for sample characteristics.)

Measures

Mothers completed two types of measures to index their psychological well-being: (1) measures focused on parenting and family issues (i.e. the Positive Affective Index, selected subscales of the Family Environment Scale, and the Pessimism subscale of the Questionnaire on Resources and Stress) and (2) more general measures of psychological well-being (i.e. the Center for Epidemiologic Studies Depression Scale,

Table 1 Means (standard deviations) for youth characteristics

	Fragile X only	Fragile X + autism	Down syndrome
Chronological age in years	15.83 (3.42)	16.38 (2.39)	16.04 (2.93)
Non-verbal MA in years*	3.86 (0.43)	3.79 (0.46)	3.84 (0.55)
Mother ABC – total score	35.47 (18.60) ($n = 19$)	71.67 (19.00) ($n = 9$)	13.53 (14.36) ($n = 19$)
Father ABC – total score	37.11 (27.97) ($n = 18$)	56.29 (27.48) ($n = 7$)	18.74 (16.30) ($n = 19$)
Teacher ABC – total score	31.94 (20.22) ($n = 18$)	44.63 (19.92) ($n = 8$)	12.71 (12.18) ($n = 17$)

*Determined from the Copying, Pattern Analysis, and Bead Memory subtests of the Stanford-Binet, 4th edition.

ABC, Autism Behavior Checklist; MA, mental age.

Table 2 Means (standard deviations) for maternal characteristics

	Fragile X only ($n = 19$)	Fragile X + autism ($n = 9$)	Down syndrome ($n = 19$)
Age (years)	43.6 (6.1)	44.0 (3.7)	48.3 (7.2)
Maternal education (% with college degree or higher)	52.6	33.3	36.8
Family income mean category*	7.74 (3.23)	7.33 (2.87)	7.44 (2.79)

*Income categories: 1 = less than $10 000 annually, 2 = $10 000–20 000 annually, 3 = $20 000–30 000, . . . 11 = $100 000 or more annually.

the Life Satisfaction Rating Scale, and the Multidimensional Coping Inventory).

Closeness of the mother–child relationship was measured by the Positive Affect Index (PAI; Bengston & Black 1973). This 10-item scale assesses (1) the level of understanding, trust and affection the mother feels towards her son and (2) the mother's perception of reciprocated closeness (i.e. the extent to which the mother perceives that the son feels close to his mother). Each item is rated on a 6-point scale, with higher scores indicating better relationship quality. Cronbach's alpha reliability was 0.70 for maternal feelings towards the child and 0.66 for reciprocated closeness for the present sample.

Quality of family relationships was measured by the cohesion, expressiveness and conflict subscales of the Family Environment Scale (FES; Moos & Moos 1981). The respondent indicates whether each of the 27 items is presently true or false for most family members. Higher scores indicate less cohesion and expressiveness and more conflict on the respective subscales. Cronbach's alpha reliability was 0.70 for cohesion, 0.63 for expressiveness, and 0.67 for conflict for the present sample.

Pessimism was measured by the pessimism subscale of the Questionnaire on Resources and Stress (QRS; Friedrich *et al.* 1983). This 11-item subscale measures the respondent's degree of pessimism about whether the child will become self-sufficient. Mothers rate each item as true or false. Higher scores indicate greater pessimism. Cronbach's alpha reliability was 0.74 for the present sample.

Maternal depressive symptoms were assessed by the Center for Epidemiologic Studies Depression Scale (CES-D; Radloff 1977). This 20-item scale assesses the frequency of depressive symptoms during the preceding week, rated on a 4-point scale (0 – rarely to 3 – most of the time). A score of 16 or higher indicates clinical depression. Cronbach's alpha reliability was 0.75 for the present sample.

Satisfaction with life was measured by the single-item Life Satisfaction Rating Scale (Campbell *et al.* 1976), with a score of 1 indicating high satisfaction and a score of 7, low satisfaction.

Coping style was measured by the Multidimensional Coping Inventory (COPE; Carver *et al.* 1989). It consists of 32 statements about the coping strategy used when the respondent faces a stressful event. Two coping style scores are derived: emotion-focused (denial, venting of emotions, behavioural disengagement and mental disengagement) and problem-focused (active seeking of a means to remove or reduce the stressor, planning, suppression of competing activities, and positive reinterpretation and growth). Cronbach's alpha reliability was 0.70 for emotion-focused and 0.85 for problem-focused for the present sample.

In addition to measures of maternal functioning, behavioural symptoms of autism manifested by the target adolescent or young adult were measured by maternal, paternal and teacher ratings on the 57-item ABC. The youth in the three groups were significantly different from each other according to mother ratings on the ABC, with those having a co-morbid diagnosis of autism and fragile X syndrome having the most symptoms, those with fragile X syndrome alone having fewer symptoms, and those with Down syndrome having the fewest symptoms ($F_{2, 44} = 35.62$, $P = 0.000$, with all three *post-hoc* pairwise comparisons significant at $P = 0.000$, one-tailed). The three groups also differed according to father ($F_{2, 41} = 7.20$, $P < 0.002$) and teacher ratings ($F_{2, 40} = 10.57$, $P = 0.000$), with all *post-hoc* pairwise comparisons significant for father and teacher ratings ($P < 0.02$ or better, one-tailed). (See Table 1.)

Results

We first determined whether there were differences among the three groups of mothers on the measures of well-being and coping style by conducting a series of ANOVAs or MANOVAs on each measure, depending on whether the measure yielded one or more than one dependent variable, respectively, and using the son's diagnosis as the independent variable.

Looking first at the measures of parenting and family issues, a MANOVA on the two scores derived from the PAI (closeness felt by the mother towards the son and the mother's perception of reciprocated closeness by the son) was significant (Wilks' lambda = 0.72, $F_{4, 86} = 3.81$, $P = 0.007$). Follow-up univariate F-tests were evaluated using the Holm sequential procedure (Holm 1979) to prevent inflation of Type I error. In this procedure, the largest F had to reach an alpha of 0.025 ($\alpha/2$) to be significant and the smallest, an alpha of 0.05. The effect of the son's diagnosis was significant for perceived reciprocated closeness by the son ($F_{2, 44} = 5.21$, $P = 0.009$), but not for maternal feelings of closeness towards the son ($F_{2, 44} = 0.95$, $P = 0.394$). *Post-hoc* pairwise comparisons [using Fisher's least significant difference (LSD) technique to maintain familywise alpha at $P \le 0.05$; Levin *et al.* 1994] revealed that mothers of sons with co-morbid fragile X syndrome and autism felt less reciprocated closeness compared with mothers of sons with Down syndrome or mothers of sons with only fragile X syndrome, with the latter two

groups not being significantly different. (See Table 3.)

A MANOVA on the cohesion, expressiveness and conflict scores derived from the FES was significant (Wilks' lambda = 0.74, $F_{6, 84} = 2.25$, $P < 0.046$). In evaluating the follow-up univariate tests, the Holm procedure required the largest F to reach an alpha of 0.017 to be significant (i.e. $\alpha/3$), the next largest F, an alpha of 0.025 ($\alpha/2$), and the smallest, an alpha of 0.05. This procedure revealed a significant effect of the son's diagnosis only for conflict ($F_{2, 44} = 4.77$, $P = 0.013$). Paired comparisons using Fisher's LSD ($P < 0.05$) revealed that mothers of sons with co-morbid fragile X syndrome and autism experienced more conflict in the family than mothers of sons with Down syndrome, and mothers of sons with only fragile X syndrome experienced marginally more conflict than mothers of sons with Down syndrome ($P = 0.065$). The two groups of mothers parenting youth with fragile X syndrome did not differ in conflict. (See Table 3.)

A one-way ANOVA on the pessimism subscale of the QRS yielded a significant effect of son's diagnosis ($F_{2, 44} = 6.16$, $P = 0.004$). *Post-hoc* comparisons using Fisher's LSD ($P \le 0.05$) revealed that mothers of sons with fragile X syndrome with and without a co-morbid diagnosis of autism did not differ, but each was more pessimistic about their son's future than mothers of sons with Down syndrome. (See Table 3.)

Turning to the more general measures of well-being, we examined total scores from the CES-D, and also compared the number of mothers scoring

Table 3 Mean (standard deviation) maternal well-being scores related to parenting

	Fragile X only	Fragile X + autism	Down syndrome	Statistic
PAI mother to son	27.11 (2.56)	26.00 (2.55)	26.11 (2.49)	$F = 0.95$
PAI son to mother	26.00 (2.36)	23.11 (2.47)	26.16 (2.61)	$F = 5.21$ [†§**]
FES – cohesion	7.52 (2.01)	7.54 (1.81)	7.86 (1.23)	$F = 0.23$
FES –expressive	6.53 (2.22)	6.67 (1.94)	6.29 (1.60)	$F = 0.13$
FES – conflict	2.43 (1.95)	3.44 (1.74)	1.39 (1.36)	$F = 4.77$ [†**‡*]
QRS – pessimism	6.77 (2.16)	7.67 (2.00)	4.77 (2.51)	$F = 6.16$ [†‡**]

*$P = 0.065$, **$P \le 0.01$.

[†] FXS + autism vs. DS.

[‡] FXS vs. DS.

[§] FXS + autism vs. FXS.

DS, Down syndrome; FXS, Fragile X syndrome; PAI, Positive Affect Index; FES, Family Environment Scale; QRS, Questionnaire on Resources and Stress.

above and below the cut-off for clinical depression on the CES-D (i.e. 16), and again found no group differences ($F_{2, 44} = 0.71$, $P < 0.496$, and $\chi^2 = 0.44$, $P < 0.805$, respectively). Only two mothers in the fragile X syndrome only group and one in each of the other groups met the cut-off for clinical depression. (See Table 4.)

Life satisfaction scores were analysed using a Kruskal–Wallis test because of a significant difference in the variance of scores across groups. Mothers in the three groups did not differ in life satisfaction ($\chi^2 = 3.21$, $P = 0.20$). (See Table 4.)

Finally, the three groups of mothers did not differ according to their style of coping, based on a MANOVA on the problem-solving focused and emotion-focused COPE scores (Wilks' lambda = 0.88, $F_{4, 86} = 1.49$, $P = 0.21$). (See Table 4.)

Next, we used multiple regression to examine the relationship between each measure of maternal well-being and the autistic symptoms of the son, as measured by ABC scores. The aim was to determine whether the behaviours leading to the diagnosis of autism explained between- and within-group differences in maternal well-being. The regression analysis for each dependent variable was conducted in two steps. In Step 1, two dummy variables indexing whether the son had fragile X syndrome only or Down syndrome, respectively, were entered into the regression. A significant coefficient for a dummy variable indicated that the group indexed by that variable differed significantly from the mothers with sons in the co-morbid group on the dependent variable of interest.

In Step 2 of the regression, we entered the ABC scores for the sons. In doing so, we used the mean of the father- and teacher-reported ABC total scores. We did not use mother-reported ABC scores, to avoid the interpretive difficulties of determining cause and effect from a correlation between maternal well-being and maternal perception of child behaviour. In seven cases, we were missing either the father-reported or the teacher-reported ABC scores, and so we used the ABC that was available. We reasoned that if a group coefficient that was significant at Step 1 became non-significant at Step 2, we could conclude that the group differences were 'explained' by the son's autism symptoms.

We found that although there were significant group effects at Step 1, the addition of the ABC scores at Step 2 did not improve prediction compared with Step 1 for any dependent variable. Moreover, the ABC score was not a significant predictor of any dependent measure at Step 2.

Discussion

Our findings suggest that measures of well-being closely tied to family and parenting issues and concerns – pessimism about the child's future, feelings of reciprocated closeness by the child as perceived by the mother, and family relations, specifically conflict – are affected by the child's diagnostic category, at least when the comparison is between fragile X syndrome with co-morbid autism, fragile X syndrome only, and Down syndrome. The more general dimensions of psychological well-being we assessed – maternal depression, life satisfaction and coping style – were unaffected by the child's diagnosis. In contrast, several other studies (e.g. Dunst *et al.* 1986;

Table 4 Mean (standard deviation) maternal general well-being and coping style scores

	Fragile X only	Fragile X + autism	Down syndrome	Statistic
Depression	8.32 (7.07)	7.56 (7.23)	5.84 (5.41)	$F = 0.71$ ($P = 0.496$); Pearson chi-square = 0.44 ($P = 0.805$)
Life satisfaction	2.68 (1.20)	2.33 (1.12)	2.0 (0.82)	Kruskal–Wallis chi-square = 3.21 ($P = 0.20$)
Emotion-focused coping*	12.84 (5.27)	8.78 (4.89)	10.05 (4.90)	$F = 0.752$ ($P = 0.477$)
Problem-solving focused coping*	31.86 (8.81)	36.00 (5.34)	32.37 (9.60)	$F = 2.46$ ($P = 0.097$)

*Wilks' lambda = 0.875, $F = 1.485$, $P = 0.214$.

Marcovitch *et al.* 1986; Walker *et al.* 1992; Kasari & Sigman 1997; Ly & Hodapp 2002; Ricci & Hodapp 2003; Abbeduto *et al.* 2004) have observed child effects on general measures of well-being. It may be that the small sample size and resulting limits on statistical power in the present study made it difficult to detect the more subtle effects of child disability on general well-being compared with parenting and family measures.

In the current study, mothers of sons with fragile X syndrome with co-morbid autism reported that they felt less reciprocated closeness from their child than mothers of sons with fragile X syndrome only. In the Abbeduto *et al.* (2004) study, mothers of youth with autism perceived less reciprocated closeness compared with mothers of youth with only fragile X syndrome. Thus, it appears that autism, whether the sole diagnosis or co-morbid with another disorder, leads mothers to feel that their child lacks an emotional connection with them. Unfortunately, the lack of an idiopathic autism group in the present study makes it impossible to determine whether the co-morbidity of fragile X syndrome and autism creates lower levels of reciprocated closeness than would be created by either condition independently.

In contrast to the findings of Abbeduto *et al.* (2004), we found that mothers of sons with Down syndrome did not report feeling any more reciprocated closeness from their sons than mothers of sons with only fragile X syndrome. This likely reflects differences in the characteristics of the samples between the two studies. In the present study, unlike Abbeduto *et al.*, we included only males and then only those with the lowest IQs from the larger study.

In the present study, we found that mothers of sons with fragile X syndrome, both with and without co-morbid autism, were more pessimistic about their child's future than mothers of sons with Down syndrome. This finding is consistent with those of Abbeduto *et al.* (2004). The present findings further suggest that autism confers no additional burden on mothers in this domain beyond that associated with having a son with fragile X syndrome. Presumably, the relatively high level of behaviour problems associated with fragile X syndrome and, perhaps, the declining rates of cognitive and adaptive behaviour development characteristic of affected males (Hager-

man 1999), cause the high levels of maternal concerns about the child's ability to function independently in the future.

We also found that conflict within the family was reported to be greater by mothers of youth with fragile X syndrome, regardless of autism status, compared with mothers of youth with Down syndrome (although the difference was marginal for the group of mothers of sons with fragile X syndrome only). It may be, again, that the behaviour problems so characteristic of fragile X syndrome (e.g. social anxiety, hyperactivity, hypersensitivity to sensory stimuli) lead directly to disputes among family members about how to control the behaviour or create such stress that disputes among family members concerning other issues are exacerbated.

In trying to explain between- and within-group differences in our maternal measures, we examined the contribution of the son's autism symptoms, as measured by the ABC. We found that ABC scores bore little relationship to the maternal well-being measures. In contrast, Abbeduto *et al.* (2004) found that the total ABC score was an important predictor of maternal pessimism, reported closeness of the mother–child relationship, and depression. The difference in results may reflect the different informants used in the two studies. In the Abbeduto *et al.* (2004) analyses, the ABC scores were provided by the mother, whereas in the present analyses, the ABC scores were provided by fathers and teachers. It may be that maternal psychological well-being influences maternal reports of child problem behaviours, thereby creating an artefactual relationship between the two. A strength of the current study was that by using father and teacher ABC scores we avoided that possibility. It is also possible, however, that fathers and teachers may simply be less attuned to the maladaptive behaviours that mothers find most stressful.

There are several limitations of the present study that should be addressed in future research. First, the small sample size limited statistical power. Future research should include larger sample sizes, especially of individuals with co-morbid autism and fragile X syndrome diagnoses, as well as an idiopathic autism group, in order to obtain a fuller understanding of similarities and differences in the impact of the diagnosis on maternal well-being. Second, we did not have access to information on the

genetic status of the participating mothers. Recent evidence (Hessl *et al.* 2005) clearly demonstrates that variation in the premutation range in expansion size, fragile X mental retardation protein (FMRP) level, and FMR1 messenger RNA level are correlated with measures of mental health status. It is thus likely that maternal genetic status and child behaviours and characteristics interact to determine maternal psychological well-being, and this should be more fully explored in future studies. It will also be important to determine whether there are different genetic profiles between mothers of individuals with and without a co-morbid autism diagnosis. Third, we did not measure maternal neurocognitive status, which may affect maternal perceptions of the child and thus, indirectly, maternal well-being. In fact, Sobesky *et al.* (1996) have shown maternal executive function problems to be particularly important in this regard. Although there is no evidence to suggest that either the genetic or neurocognitive status of mothers of children with fragile X is associated with the child's autism status, future research should nonetheless examine executive function and other neurocognitive impairments among mothers and evaluate their contribution to maternal mental health. Fourth, fathers were not considered in the present study; however, paternal well-being and contributions to family functioning, as well as how they are affected by the child's diagnosis, are important questions. Indeed, there is recent empirical evidence that father characteristics and psychological functioning have an impact on the development of challenging behaviours in youth with fragile X syndrome (Hessl *et al.* 2001).

In conclusion, it is important that future research go beyond the establishment of differences in parental well-being based on child diagnostic categories, to establish the child and contextual factors as well as the dynamics that lead to those differences. It will also be important to determine whether there are 'protective factors', such as coping style, that help parents to deal with the stresses of having a child with special needs.

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