

PARENTING STRESS AND SOCIAL SUPPORT IN FAMILIES WITH CHILDREN  
WITH FRAGILE X SYNDROME: A COMPARISON OF MOTHERS AND FATHERS

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## ABSTRACT

ANNA CHRISTINE JAMES LONG: Parenting Stress and Social Support in Families with Children with Fragile X Syndrome: A Comparison of Mothers and Fathers  
(Under the direction of Jane Roberts and Barbara H. Wasik)

The primary purpose of this study was to address a gap in previous research by describing and directly comparing maternal and paternal stress using an etiology-specific sample of families with children with fragile X syndrome (FXS). The study completed an across parent gender examination of the relationship of social support and parenting stress in order to: (a) initiate research regarding differences in magnitude of contribution of variables to maternal versus paternal stress, and (b) advance research regarding the development of effective parent interventions. The study sample consisted of 38 families, including 76 parents (38 mother-father dyads) and their male ( $n = 30$ ) and female ( $n = 8$ ) child with FXS. All mothers had the premutation form of FXS, while target children had the full mutation and ranged in age from 3 years- 6 months to 10 years- 6 months.

In a major finding, this study showed the importance of moving beyond typical examinations of group means when making comparisons of mothers' and fathers' parenting experiences. In the present study, no significant group mean differences were found in mothers' versus fathers' reported parenting stress or perceived social support. Yet, upon further examination of data, original findings suggesting similar levels of maternal and paternal stress and social support proved misleading. First a lack of inter-parent agreement on measures of parenting stress and social support revealed mothers and fathers often have different experiences despite living within the same households. Second, differences in

parent domain stress between mothers and fathers lent support for a vulnerability to heightened parenting stress due to mothers' premutation status.

No difference was found in the relationship of social support to parenting stress when examining across parent gender. However, consistent relationships were found between parenting stress and family support as well as parenting stress and child problem behavior when salient variables were controlled for. Post hoc exploratory analyses exposed one promising area for future research. Namely, data indicated a difference in mothers' versus fathers' stress response to similar levels of child problem behavior. As child problem behavior increased, maternal stress increased at a significantly higher rate than paternal stress.

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## ABBREVIATIONS

ABC	Adaptive Behavior Composite
ADHD	Attention Deficit Hyperactivity Disorder
CBCL	Child Behavior Checklist
CGG	Cytosine-guanine-guanine trinucleotide
DHHS	Department of Health and Human Services
DNA	Deoxyribonucleic acid
FES	Family Environment Scale
FMRP	Fragile X Mental Retardation Protein
FMR1	Fragile X Mental Retardation Gene
FSS	Family Support Scale
FXS	Fragile X Syndrome
HLM	Hierarchical Linear Model
PAIR	Personal Assessment of Intimate Relationships Inventory
PSI	Parent Stress Index
PSI PD	Parenting Stress Index: Parental Distress
PSI P-CDI	Parenting Stress Index: Parent-Child Dysfunctional Interaction
PSI DC	Parenting Stress Index: Difficult Child
SCID-I/NP	Structured Clinical Interview for the DSM-IV-I: Non-patient Edition
SES	Socio-economic status
SSRI	Selective serotonin reuptake inhibitors
SNRI	Serotonin-norepinephrine reuptake inhibitors
UNC-CH	University of North Carolina at Chapel Hill

VABS

Vineland Adaptive Behavior Scale

## CHAPTER I

### Introduction

Parents respond in many diverse ways to raising a child with a disability, ranging from gaining insight and appreciation for the gifts and personal growth that the child has brought, to feelings of grief over the loss of the “expected” child. Many different factors contribute to parents’ functioning including characteristics of the child, the parent, the family, and the social support network. As a result, parent functioning is best understood within the framework of an ecological systems context, within which individuals and systems are mutually influential on one another (Bronfenbrenner, 1979).

Compared to parenting a typically developing child, raising a child with a disability is often associated with additional demands related to behavior management, caretaking, and allocation of family time and resources. As these additional demands accumulate, parents’ abilities to cope can be overtaxed resulting in heightened levels of parenting stress (Thoits, 1995). Although it is clear that parental outcomes differ depending on multiple factors which interact to influence parent functioning, high levels of parenting stress have been consistently found among parents of children with disabilities when they are compared to parents whose children are typically developing (Britner, Morog, Pianta, & Marvin, 2003; Button, Pianta, & Marvin, 2001; Herring, Gray, Taffe, Tonge, Sweeney, & Einfeld, 2006; Noh, Dumas, Wolf, & Fisman, 1989; Woolfson & Grant, 2006; Oelofsen & Richardson, 2006; Quitter, Glueckauf, & Jackson, 1990). While parenting stress is not the only impact on parents of raising a child with a disability, it is one of primary importance due to its association with

overall parent well-being (Bailey, Sideris, Roberts, & Hatton, 2008; Bailey, Golden, Roberts, & Ford, 2007; Thoits, 1995; Wheeler, Hatton, Reichardt, & Bailey, 2007), parenting responsiveness and consistency (van Lieshout, DeMeyer, Curfs, & Fryns, 1998; Wheeler et al., 2007), abusive behavior (Aniol, Mullins, & Page, 2004; Rodriguez & Murphy, 1997) and family disruption (Bristol, Gallagher, & Schopler, 1988; Quitter et al., 1990).

Several primary factors are associated with parenting stress in families with children who have disabilities. These include child characteristics, and dimensions of the parent, family and environment. Child characteristics are important for parents' experience in their parenting role, and are amongst the most influential contributors to parenting stress. A variety of child characteristics have been investigated in regards to their relationship with parenting stress in families with children with disabilities. These include, but are not limited to, problem behavior, autistic behavior, functional abilities, chronological age, and gender. Studies investigating the impact of these variables on parenting stress have yielded mixed results. And, although it seems reasonable that the severity of the child's functional limitations (cognitive impairment and adaptive behavior) would be a strong predictor of parenting stress, this has not been consistently supported (Baker, Blacher, Crnic, & Edelbrock, 2002; Hastings, Kovshoff, Ward, Espinosa, Brown, & Remington, 2005). Instead, child problem behavior has been found to be a primary contributor to parenting stress (Baker et al., 2002; Dumas, Wolf, Fisman, & Culligan, 1991; Hastings, 2003; Herring et al., 2006). In fact, child problem behavior often accounts for the relationship between child functional abilities (Britner et al., 2003; Hastings, 2002) and parenting stress, and at times explains the association between parenting stress and other child characteristics such as gender (Hall, Burns, & Reiss, 2007).

In addition to child characteristics, several dimensions of the parent, family and environment have frequently been investigated for their relationship with parenting stress in families with children with disabilities. Some of these include family income, parent education, social support factors, and parent gender. Family income typically is used as a general indicator of socio-economic status (SES) and is almost always a control variable in analyses of parenting stress due to its frequent inverse relationship with parenting stress (Burbach, Fox, & Nicholson, 2004; Koeske & Koeske, 1990, Shin, Nhan, Crittenden, Hong, Flory, & Ladinsky, 2006; Smith, Oliver, & Innocenti, 2001; Warfield, Krauss, Hauser-Cram, Upshur, & Shonkoff, 1999). Interestingly, despite the strong association between income and educational attainment in the general population, parent education has not reliably been shown to associate with parenting stress in families with children with disabilities (Boyce & Behl, 1991; Lecavalier, Leone, & Wiltz, 2006; White & Hastings, 2004). Some research suggests that the inconsistency of the relationship between parent education and parenting stress may, in part, be due to mothers' greater tendency in these families to stay at home in order to manage the additional care needs of the child (Sen & Yurtsever, 2007; Simmerman, Blacher, & Baker, 2001). This situation leads to lower family income unrelated to educational attainment. Lower family income may exacerbate parenting stress by placing limitations on the social support resources available to parents of children with disabilities. Parents often experience stress when they perceive that the supports available are inadequate to deal with the demands posed by caring for a child with a disability.

Family disability research has documented the significance of social support for parenting stress (Johnston et al., 2003; Kersh, Hedvat, Hauser-Cram, & Warfield, 2006;



Sarimski, 1997; Smith et al., 2001; White & Hastings, 2004). These studies highlight the strong correlation between social support and successful coping as well as the potential of social support to act as a stress buffer (Koeske & Koeske, 1990). Recognition of the significant role of social support has spawned much research, especially since social support factors are readily available for professionals to target for intervention. Yet, little research to date has described differences and similarities in mothers' compared to fathers' perceptions of social support. This prerequisite knowledge is essential for developing effective modes of intervention that will positively influence parents' successful coping.

With respect to parent gender, there is very little research investigating gender differences in parenting stress associated with parenting a child with a disability. Thus, differences and similarities between maternal stress and paternal stress remain unclear. Often, what literature is present does not use samples of mother-father dyads, lumps mothers and fathers together in data analyses, or simply neglects to make direct across gender comparisons. Recent research suggests that the amount of parenting stress experienced by mothers and fathers of children with disabilities is similar (Hastings et al., 2005; Keller & Honig, 2004; Wanamaker & Glenwick, 1998). In addition, similar factors interact to influence maternal and paternal stress (Saloviita, Itälina, & Leinonen, 2003). Although, differences for mothers and fathers have been evidenced in which factors most powerfully predict their stress. For example, there is some growing support suggesting that sources of stress related to dimensions of parent functioning (e.g., health, psychopathological symptoms, parental self-esteem, role restriction) are more important for mothers' stress (Johnston et al., 2003; Koeske & Koeske, 1990; Krauss, 1993; Wanamaker & Glenwick, 1998), while parent-child attachment and child characteristics that influence parents'

interactions with their children (e.g., child acceptability, mood, adaptability, reinforcement) are more important for fathers' stress (Hastings et al., 2005; Keller & Honig, 2004; Krauss, 1993; Noh et al., 1989). Yet, to date, research outlining these findings is weak due to significant limitations in study designs and a lack of use of statistical analyses which allow for direct comparison between maternal and paternal stress. Thus, examining gender differences and similarities in parenting stress is one of the primary purposes of this study.

Numerous studies have indicated the importance of etiology-specific research in examining parenting stress (Abbeduto, Seltzer, Shattuck, Krauss, Orsmond, & Murphy, 2004; Hodapp, Dykens, & Mashino, 1997; Hodapp, Fiddler, & Smith, 1998; Johnston et al., 2003), as some etiologies are consistently associated with greater parenting stress than others. Much research in this area has compared parents of children with autism with parents of children with Down syndrome, or with parents of children with other disabilities. Frequently, where a significant finding is reported, parents of children with autism fair poorly (Abbeduto et al., 2004; Dumas et al., 1991; Noh et al., 1989; White & Hastings, 2004), while parents of children with Down syndrome fair well relative to parents of children with other disabilities (Abbeduto et al., 2004; Eisenhower, Baker, & Blacher, 2005; Lewis et al., 2006; Noh et al., 1989). It is generally agreed upon that these differences in parenting stress are driven by differences in the behavioral phenotypes associated with the etiology of each disability (McCarthy, Cuskelly, van Kraayenoord, & Cohen, 2006). Most often differences in parenting stress across etiologies is related to the level of expressed problem behaviors, with some disability types exhibiting more behavioral problems than others. For example, in a study investigating the influence of syndrome-specific behavior on maternal well-being, Eisenhower et al. (2005) found that behavior problems differed by syndrome group, with

highest levels found among children with autism or cerebral palsy and similarly low levels found among typically developing children and children with Down syndrome. As expected, specific syndromes related to children's expression of behavior problems which then related to parents' experiences of stress.

As with parents of children with disabilities of varying etiologies, the majority of parents of children with FXS exhibit heightened stress levels (Johnston et al., 2003; Lewis et al., 2006; McCarthy et al., 2006; Sarimski, 1997; von Gontard et al., 2002; Wheeler et al., 2007). In fact, some studies suggest that parents of children with FXS exhibit relatively comparable levels of negative parenting outcomes to parents of children with autism (Abbeduto et al., 2004; Franke et al., 1996). Despite this noteworthy comparison to parents of children with autism and research indicating the importance of etiology-specific research, few studies have focused their investigations on parenting stress in families with children with FXS (e.g., Johnston et al., 2003; McCarthy et al., 2006).

The lack of focus of research in families with FXS is unfortunate since an investigation of parenting stress in these families could provide a unique opportunity to examine a broad and diverse range of variables thought to explain how and why some parents' with known etiology diagnoses adapt successfully, while others do not. First, mothers of children with FXS may be especially vulnerable to the stressors of having a child with a disability due to their fragile X mental retardation gene (FMR1) premutation status, which has been associated with increased risk for psychiatric conditions (Franke et al., 1996). Second, their knowledge of having passed on a genetic disorder to their child and reports of a lack of perceived social support from professionals (Bailey, Skinner, & Sparkman, 2003; Poehlmann, Clements, Abbeduto, & Farsad, 2005) can exacerbate feelings

of guilt and isolation. Third, parents of children with FXS often must bear an additional burden of explaining to extended family members complex genetic information and informing them that they too might be carriers of FXS (Bailey et al., 2003). Fourth, the process to obtain a diagnosis of FXS for their child is often challenging and drawn out (mean age of diagnosis for boys= 35 months; Bailey, Skinner, Hatton, & Roberts, 2000). This difficulty in obtaining a diagnosis results in delayed knowledge regarding reproductive risk, leading to increased probability of having more than one child with a disability. In fact, Bailey et al. (2003) found that 57% of the children born to families after the birth of their first child with FXS but before the FXS diagnosis also were diagnosed with the full mutation of FXS. Thus, a substantial number of families of children with FXS have two children with the disability. Finally, because of the diversity in levels of functioning abilities, and high expression of problem behavior in children with FXS, these children provide an especially informative look at how child characteristics influence parenting stress.

In summary, heightened levels of parenting stress have been consistently found in parents of children with disabilities in comparison to parents of typically developing children. Both child problem behavior and parents' perceived social support are important factors for parents' exhibited stress; however, to date, research has largely neglected fathers in their investigations of parenting stress and therefore less is known about factors related to their stress. Within families with children with FXS a delineation of paternal versus maternal stress may be particularly important due to the unique constellation of variables which interact to influence parents' stress in these families. The present study aimed to address the limitations of previous research by directly comparing maternal and paternal stress, using an

etiology-specific sample of families with children with FXS and mother-father dyads. Specifically, this study describes in detail differences and similarities in mothers' and fathers' parenting stress. As well, it describes differences and similarities in mothers' and fathers' perceptions of social support. Finally, the relationship between parenting stress and social support factors was examined comparing across mothers and fathers. Results from this study are intended to advance research regarding differences in the magnitude of contribution of variables to maternal versus paternal stress and to inform parent intervention research in families with children with FXS. Finally, findings from this study may help to guide family policy and inform treatment efforts, which aim to promote overall family well-being.

## CHAPTER II

### Literature Review

#### *Fragile X Syndrome Overview*

Of the known genetic causes of mental retardation and developmental delay, FXS is considered the most common inherited form with an estimated prevalence of 1:4000 male and 1:8000 female births (Sherman, 2002). FXS is a single gene disorder resulting from an expansion of an unstable CGG (cytosine-guanine-guanine) trinucleotide repeat sequence in the promoter region of the FMR1 gene. DNA testing provides the diagnosis for both the affected status referred to as the “full mutation” condition and the carrier status referred to as the “premutation” condition. Research has revealed that both males and females can be carriers of the FMR1 mutation, passing it down from one generation to the next with increasing chances of transmitting the full mutation to their offspring. Because of the X-linked inheritance of FXS, male carriers transmit the FMR1 mutation only to their daughters. Interestingly, they only transmit this mutation in its premutation form. However, female carriers can transmit the FMR1 mutation to both their daughters and sons in either the premutation or full mutation form. It is believed that the reduction of fragile-X-mental retardation protein (FMRP), assumed essential for normal brain function and development, and the interaction of FMRP on other genes and proteins causes the various physical, cognitive and behavioral symptoms associated with FXS.

A categorical classification of the FMR1 gene based on the molecular characteristics and associated clinical affectedness is used to categorize individuals into three groups (Reiss,

Freund, Abrahams, Boehm, & Kazazian, 1993). These categories include: (1) normal or absent for FXS, (2) carrier or premutation form, and (3) affected or full mutation form. Repeats in individuals absent for FXS range from approximately 6 to 55 CGGs. Importantly, the number of CGG repeats in this population remains stable from one generation to the next. Conversely, carrier females and males have an expansion of repeats ranging from approximately 55 to 200 and have a mutation at the FMR1 gene that often has an unstable number of repeats. Due to its instability the number of repeats often increases through subsequent generations when the mutation is transmitted through a female. Finally, individuals with the full mutation demonstrate expansions of the mutation beyond 200 repeats. The full mutation is associated with methylation of the adjacent CpG island (short stretch of DNA in which the frequency of the CG sequence is higher than other regions) and presumed inactivation of the FMR1 gene. Thus, the full mutation presents with the hypermethylation of the FMR1 gene, which impedes protein production (Pieretti et al., 1991) and is associated with moderate mental retardation in most males (Bailey & Nelson, 1995).

#### *Full Mutation Phenotype*

The phenotypic expression of the full mutation of FXS is quite variable and appears to be associated with gender, FMRP, and co-occurring psychiatric conditions. FXS occurs in both males and females. While approximately one half of females with the full mutation of FXS have intellectual disabilities and most exhibit only mild delays (Lachiewicz, 1995; Mazzocco, 2000), males typically are more severely affected due to the X-linked inheritance and random X-inactivation of FXS. Males with FXS typically exhibit developmental delays in all domains, although their intellectual and communication skills are likely to be most affected and their motor skills least affected (Bailey, Hatton, & Skinner, 1998a; Roberts,

Hatton, & Bailey, 2001). The majority of males are also characterized by mild to moderate intellectual disabilities (Bailey et al., 1998a; Hagerman et al., 1994). Children with the full mutation often have characteristic physical features including a long narrow face, protruding ears, and in males, post-pubertal macrochidism (Meyer & Batshaw, 2002). The spectrum of functioning in FXS can be most easily understood by a gradual decrement of FMRP production from normal levels to a complete absence. In mildly affected individuals, typically females, the intellectual functioning may be normal; however, learning and/or psychosocial problems are commonly evident (Mazzocco, 2000). In general, as FMRP levels decrease, the severity of intellectual impairment and presence of characteristic physical features increase. In the full mutation, little or no FMRP is present (Hagerman, 2002).

An increasingly well-defined behavioral phenotype is observed in FXS. Children with the full mutation often display low adaptive functioning, stereotypic behavior, poor eye contact, tactile defensiveness, hyperactivity/ hyperarousal, inattention, aggression, and social anxiety/avoidance (see Hagerman, 2002; McCarthy et al., 2006). Many children with the full mutation also have co-morbid psychiatric conditions with Attention Deficit Hyperactivity Disorder (ADHD) reported in approximately 35% of females and 70% of males (Baumgardner, Reiss, Freund, & Abrams, 1995; Sarimski, 1997; von Gontard et al., 2002; Freund et al., 1993). Autism is another psychiatric condition associated with FXS with up to 90% of children with the full mutation displaying one or more characteristics of autism such as avoidant eye contact, hand flapping, perseveration in speech, shyness and tactile defensiveness (Bailey, Mesibov, Hatton, Clark, Roberts, & Mayhew, 1998b; Meyer & Batshaw, 2002; Sarimski, 1997). In addition, approximately 30% of children with FXS meet diagnostic criteria for autism (Bailey et al., 1998b; Bailey, Hatton, Mesibov, Ament, &



Skinner, 2000; Bailey et al., 2004; Kaufmann et al., 2004; Rogers, Wehner, & Hagerman, 2001). Importantly, many researchers have reported that children with co-morbid diagnoses of FXS and autism fair poorly in comparison to children who have FXS without autism. In general, studies indicate that they have lower functional abilities including lower intellectual functioning and adaptive behavior (Bailey et al., 2000; Kau, et al., 2004; Lewis et al., 2006; Hatton et al., 2003) and greater problem behavior (Hatton, Hooper, Bailey, Skinner, Sullivan, & Wheeler, 2002; Kau et al., 2004).

### *Premutation Phenotype*

Females with the premutation may exhibit some clinical involvement. The most consistent body of findings pertains to the physical effects of the premutation, particularly the increased frequency of premature ovarian failure (Allingham-Hawkins et al., 1999; Sherman, 2000). Literature regarding cognitive and psychosocial effects of the premutation is often inconsistent. Generally, females with the premutation exhibit average cognitive ability with some evidence for learning, attention and executive functioning difficulties in subgroups (Mazzocco, 2000; Mazzocco, Pennington, & Hagerman, 1993). Increasingly, however, the premutation is drawing attention because of its relationship to psychiatric conditions. For example, excessive rates of affective disorders have been found in females with the premutation in comparison to females in the general population (Bailey et al., 2007; Franke et al., 1996; Roberts et al., in press). As well, a significantly higher frequency of anxiety disorders has been found in premutation mothers compared to mothers of children with autism or typically developing children (Franke et al., 1996; Franke et al., 1998). Age of onset analysis indicates that the onset of psychiatric disorders of mothers with the premutation is considerably earlier (on average approximately 9 years earlier) than their

child's diagnosis of FXS (Franke et al., 1996). Additionally, mothers with the premutation (with affected children) and their siblings with the premutation (without affected children) exhibit similar levels of anxiety (Franke et al., 1998). Taken together these findings strongly suggest that psychiatric conditions in mothers with the premutation may in part be a consequence of their premutation status (Franke et al., 1996; Franke et al., 1998), thus placing them at heightened risk for maternal distress related to the psychosocial demands of raising a child with a disability.

### *Family Adaptation to Disability*

Raising a child with a disability often brings about additional demands and challenges not faced by parents raising typically developing children such as managing significant behavioral problems, finding appropriate services, completing additional caretaking tasks, and dealing with the uncertainty of their child's developmental difficulties. Because of these additional demands, there has been considerable research investigating the impact that raising a child with a disability has on family functioning and child development. To date, a wide range of parental outcomes have been examined outlining their adaptation. Much of the research suggests that there is diversity of adaptation across various outcomes for parents and that many parents over time cope successfully with the additional demands placed upon them (Bailey et al., 2008; Gray, 2002). However, there has been focus on documenting adverse effects of raising a child with a disability. For example, research indicates that parents of children with disabilities report higher rates of depressive and anxiety symptoms (Bitsika & Sharpley, 2004; Dumas et al., 1991; Eisenhower et al., 2005; Sharpley, Bitsika, & Efremidis, 1997; Wheeler et al., 2007), more family disruption (Bristol et al., 1988; Eisenhower et al., 2005; Quitter et al., 1990) and more social isolation (Quitter et al., 1990), as well as lower

levels of parenting self-efficacy, and positive coping and cognitive appraisal (Hassall, Rose, & McDonald, 2005; Kuhn & Carter, 2006; Plant & Sanders, 2007; Wanamaker & Glenwick, 1998).

Of the adverse effects documented, parenting stress has proven to be an important construct, fundamental to our understanding of parents' experience (McCarthy et al., 2006). Parenting stress can act as a barometer for overall parent functioning (Bailey et al., 2008; Bailey et al., 2007; Wheeler et al., 2007), and is commonly used as a marker for family disequilibrium. Stress may manifest itself in many ways and researchers have operationalized it in various ways (McCarthy et al., 2006). In families of children with disability, stress is most often operationalized as parental distress specifically related to parenting roles and relationships and thus termed "parenting stress" accordingly. Parenting stress has been defined in different ways; however, generally, it can be thought of as the negative feelings and beliefs that arise about the self, family, and child as a result of the parenting role (Deater-Deckard, 2004). Parenting stress is most commonly thought to be comprised of three components: (1) a parent domain (aspects of parenting stress that are associated with problems in the parents' own functioning), (2) a child domain (aspects of parenting stress that arise from the child's behaviors), and (3) a parent-child domain (aspects of parenting stress related to the degree of conflict in the parent-child system). Most measures of parenting stress provide an overall composite or total stress score as well as domain scores representing the three components of parenting stress. Increases in the three domains of parenting stress are thought to lead to a break down in the quality and effectiveness of parenting behavior as well as the child's functioning. For example, heightened parenting stress has been shown to negatively impact the attachment between the parent and child

(Abbeduto et al., 2004; Hadadian, & Merbler, 1996; Keller & Honig, 2004; Krauss, 1993), and parents' competency in their parenting role (Hassall et al., 2005; Wanamaker & Glenwick, 1998). As well, heightened parenting stress can lead to poor parenting skills such as lack of responsiveness (van Lieshout et al., 1998; Wheeler et al., 2007) and abusive behavior (Aniol et al., 2004; Rodriguez & Murphy, 1997).

### *Parenting Stress in Parents of Children with Fragile X Syndrome*

It has been well documented that parenting stress is high across families of children with disabilities when compared with parents of typically developing children (Baker et al., 2002; Hassall & Rose, 2005; Herring et al., 2006; Hodapp, Fidler, & Smith, 1998; Noh et al., 1989). Importantly, literature reveals considerable variability among parents in their stress experiences, with positive experiences and adaptations being increasingly acknowledged (Hastings & Taunt, 2002). A lack of understanding by the general population about FXS coupled with demands to continually manage challenging behaviors associated with the expression of the full mutation cause many parents to struggle with difficult family adaptations and chronic parenting stressors (Sarimski, 1997; von Gontard et al., 2002). As noted earlier, mothers of children with FXS may be particularly vulnerable to these stressors due to their premutation status, which may biologically predispose them to psychiatric conditions (i.e., affective and anxiety disorders) that make the demands of parenting overwhelming (Franke et al., 1996; Franke et al., 1998). Additionally, mothers bear the burden of knowing that they transmitted the disorder to their child. Furthermore, research indicates that when parents first learn of their child's diagnosis they often are responsible for informing extended family members about the genetic implications, which many describe as

a stressful experience (Bailey et al., 2003). Thus, it is probable that parents of children with FXS, particularly mothers, are at risk for increased levels of parenting stress.

Although parenting stress has been extensively studied in families of children with disabilities, it has not yet been thoroughly examined in families with children with FXS (e.g., Johnston et al., 2003; McCarthy et al., 2006). In fact, a review of current literature for the current study revealed two studies that focused their investigation on parenting stress specifically (Johnston et al., 2003; McCarthy et al., 2006) and 8 studies which examined parenting stress as one of its key variables. This research strongly suggests that parents of children with FXS experience higher levels of parenting stress than parents of typically developing children (Johnston et al., 2003; McCarthy et al., 2006; von Gontard et al., 2002; Wheeler et al., 2007) and comparable or higher levels of parenting stress to parents of children with other disabilities (Lewis et al., 2006; Sarimski, 1997; von Gontard et al., 2002). In addition, the level of parenting stress experienced often reaches a level of clinical significance indicating that the parent-child system is at risk and in need of intervention (Johnston et al., 2003; Sarimski, 1997; Wheeler et al., 2007). Most recently, Bailey et al. (2008) reaffirmed these findings in a study delineating the overall adaptation of 108 mothers of children with FXS. Bailey et al. (2008) found that as many as 29.6% (n= 32/108) of the mothers in their sample had clinically significant scores on the Parenting Stress Index (Abidin, 1995). As well, a substantial number (an additional 16%) of the remaining mothers had scores in the at-risk range. Finally, out of approximately seven measures of maternal adaptation administered, the parenting stress measure had the highest proportion of mothers above a clinically meaningful threshold.

As noted, parenting stress is typically comprised of three dimensions associated with parent functioning (parent domain stress), child characteristics (child domain stress), and parent-child interactions (parent-child domain stress). Of these dimensions, the stress response of parents of children with FXS appears most influenced by stress imposed by child characteristics as opposed to parent and parent-child interaction problems (Sarimski, 1997; Wheeler et al., 2007). Thus, although personal factors of the parent and family dynamics contribute to parenting stress, in families with children with FXS parenting stress levels are predominantly driven by characteristics of the child, such as problem behavior, that make him or her either easy or difficult to manage (Johnston et al., 2003; Sarimski, 1997; von Gontard et al., 2002). For instance, Wheeler et al. (2007) found that 61% of mothers of children with FXS reported clinically significant levels of parenting stress related to difficult characteristics of the child, while 29% reported clinically significant levels related to their own personal factors, and 38% reported clinically significant levels related to dysfunctional parent-child interactions. These results are similar to the pattern of findings by Sarimski (1997) in which mean scores of parents with children with FXS were in the clinical range on the PSI Child Domain, but not on the PSI Parent Domain.

### *Child Characteristics*

As noted, child characteristics are among the most important predictors of parenting stress in FXS. Key child characteristics that influence parenting stress include problem behavior, autistic behavior, functional abilities, chronological age, and gender. Studies investigating the impact of these variables on parental stress have yielded mixed results. However, research generally indicates that autistic behavior and especially problem

behavior are consistent and important predictors of parenting stress, while child functional abilities and other demographic variables such as chronological age and gender are not.

*Problem behavior.* Child problem behavior typically refers to two major classes of behavior: externalizing and internalizing (Achenbach, 1991). Externalizing behavior is expressed outward towards others or has an impact on the child's environment, which often makes it most disruptive (e.g., aggression, conduct problems, hyperactivity; Campbell, 2002). Internalizing behavior is a child's self-focused expressions of distress and often is ignored or goes unrecognized by adults because it is less visible and bothersome to others (e.g., depression, anxiety, somatization; Campbell, 2002). The negative impact that child problem behavior has on parenting stress has been illustrated with a broad and diverse range of samples (Hassall et al., 2005; Hastings, 2002; Hastings, 2003; Hastings et al., 2005; Herring et al., 2006; Plant & Sanders, 2007; Ricci & Hodapp, 2003; Shin et al., 2006; White & Hastings, 2004). Studies show that compared to typically developing children, children with intellectual disability engage in more problem behavior and this problem behavior is strongly associated with parenting stress above and beyond the impact of other salient child (i.e., IQ, adaptive functioning), parent (i.e., coping, social support) and family (i.e., household income) variables (Beck, Hastings, Daley, & Stevenson, 2004; Hassall et al., 2005; Hastings, 2002; Hastings, 2003; Wanamaker & Glenwick, 1998). In fact, most studies suggest that problem behavior is the primary cause of parenting stress (Baker et al., 2002; Dumas et al., 1991; Hastings, 2003; Herring et al., 2006), with trends in child problem behavior paralleling patterns in the change of parenting stress (Eisenhower et al., 2005) and parent-free measures of child problem behavior maintaining a strong ability to predict changes in parents' stress levels (Hauser-Cram, Warfield, & Shonkoff, 2001).

Corresponding with literature on parenting stress in families with children with disabilities, child problem behavior is one of the most influential contributors to parenting stress in families with children with FXS (Abbeduto et al., 2004; Johnston et al., 2003; McCarthy et al., 2006; von Gontard et al., 2002; Wheeler et al., 2007). Problem behaviors exhibited by children with FXS contribute directly to increased levels of stress in parents and appear to be cumulative in their contributions to parenting stress such that parenting stress and child problem behavior interact with each other to cause each to worsen overtime (Baker, McIntyer, Blacher, Crnic, Edelbrock, & Low, 2003; Hall et al., 2007; Wheeler et al., 2007). Additionally, problem behavior influences multiple dimensions of parenting stress not just child domain stress regarding the ease or difficulty of managing the child. Three studies that examined parenting stress in families with children with FXS revealed that child problem behavior affected parenting stress related to the parents' personal and family problems (Johnston et al., 2003; McCarthy et al., 2006; Wheeler et al. 2007). Thus, stress caused by problem behavior seemed to have a spillover effect into others aspects of the parents' experience. For example, Johnston et al. (2003) found an increased stress response on dimensions of parent functioning including their social isolation and competence as children's total problem behavior scores increased. As well, McCarthy et al. (2006) found that stress regarding parents' perception of problems for themselves and the family increased due to child problem behavior. These findings correspond with other research which indicates that a child's exhibited problem behavior can exacerbate other parent and family stressors such as parents' mental health and marital conflict (e.g., Hall et al., 2007; van Lieshout et al., 1998).



*Autistic behavior.* While autistic behavior falls within the broad category of problem behavior, it differs in that it encompasses a set of syndrome-specific behaviors. Specifically, autistic behavior describes behavioral patterns including absent or poorly developed communication skills, abnormal socialization, repetitive body movements, ritualistic behavior, lack of eye contact, and various speech abnormalities (American Psychiatric Association, 2000). Since approximately 30% of children with FXS meet criteria for a diagnosis of autism and many more exhibit autistic behavior (Bailey et al., 1998b; Bailey et al., 2000; Kaufmann et al., 2004; Meyer & Batshaw, 2002; Rogers et al., 2001), it is important to understand its relationship with parenting stress. In general, studies indicate that parents of children with autism experience higher parenting stress than both parents of typically developing children and children with intellectual and developmental disabilities not associated with autism (Abbeduto et al., 2004; Dumas et al., 1991; Dunn, Burbine, Bowers, & Tantleff-Dune, 2001; Sanders & Morgan, 1997), even when the comparison group of parents have children of similar cognitive functioning and expression of problem behavior (Eisenhower et al., 2005; White & Hastings, 2004). Therefore, the heightened parenting stress levels expressed in families with children with autism appear to be linked to the additional impact of child autistic behavior, with higher rates of expressed autistic behavior typically corresponding to higher levels of parenting stress (Abbeduto et al., 2004; Hastings et al., 2005; Tobing & Glenwick, 2002). In exploring the relationship between autistic behavior and parenting stress in families with children with pervasive developmental disorders, Tomanik, Harris, and Hawkins's (2004) study suggests that specific autistic behaviors are particularly important for parents' stress. For example, they found that lower communication skills and less social interaction were related to increased parenting stress,

while stereotypic behavior and inappropriate speech were not. Similarly, Ornstein Davis and Carter (2008) found that aspects of social relatedness were strongly negatively correlated with parenting stress for both mothers and fathers, while in comparison the relationship between atypical behavior and parenting stress was much diminished.

*Functional abilities.* Child functional abilities generally include a child's cognitive ability and/or adaptive behavior functioning (e.g., communication, self-care, social and motor life skills). Children with FXS generally display significant limitations in their functional abilities, reflected in mild to moderate intellectual disabilities and poor adaptive behavior (Hatton et al., 2003; McCarthy et al., 2006). Therefore, it seems reasonable that parents of children with FXS would experience heightened levels of parenting stress due to the additional care demands their children elicit. A recent study by McCarthy et al. (2006) examining parent and family stress where there is a child with FXS partially supports this hypothesis. In this study, child adaptive behavior and physical limitations in combination with other child behavior problems and attributes were found to predict parenting stress. Additionally, for mothers, the target child's physical limitations were an important individual predictor of parenting stress related to pessimism about their child's prospects of achieving self-sufficiency. While, for fathers, their child's adaptive behavior was particularly predictive of the presence and frequency of paternal somatic and psychosomatic symptoms.

Yet despite this apparent support for the unique contribution of child functional abilities to parenting stress, few other studies have found similar results (e.g., Most, Fidler, Laforce-Booth, & Kelly, 2006; Plant & Sanders, 2007) and an overwhelming majority of studies reveal little or no contribution of child functional abilities (e.g., Boyce & Behl, 1991; Britner et al., 2003; Hastings et al., 2005; Hassall et al., 2005; Herring et al., 2006; Kersh et

al., 2006). Further, of those studies that displayed strong direct associations between child functional abilities and parenting stress, few controlled for child problem behavior (e.g., Button et al., 2001; Smith et al., 2001; White & Hastings, 2004), which has been shown to more likely predict parenting stress and often accounts for the relationship between child functional abilities and parenting stress (Britner et al., 2003; Hastings, 2002). Yet, the relationship between parenting stress and child adaptive behavior may be best described as indistinct as other studies suggest that child functional abilities are more likely to demonstrate an impact on parenting stress when specific dimensions of adaptive behavior or more homogeneous samples are used to examine their relationship (Cameron & Orr, 1989; Hodapp et al., 1998; Plant & Sanders, 2007; Sloper, Knussen, & Cunningham, 1991; Tomanik et al., 2004).

*Chronological age and gender.* While child chronological age and gender are frequently controlled for in analyses regarding parenting stress, there is no consistent support for their affect on parenting stress (Kersh et al., 2006; Lecavalier et al., 2006; Spratt, Saylor, & Macias, 2007; Woolfson & Grant, 2006). However, to date, few studies have reported the affect of child chronological age and gender on paternal stress, as research on parenting stress has typically focused on mothers rather than fathers. Preliminary analyses investigating paternal stress have produced mixed results. For example, McBride, Schoppe, and Rane's (2002) study suggests a significant influence of child gender in that fathers report higher stress levels related to their son's difficult behavior than their daughters. And, Hauser-Cram et al. (2001) revealed both child chronological age effects as well as gender effects on paternal stress, finding sharper increases in paternal stress during their children's infant and toddler years versus their middle childhood years and an association between increases in

paternal stress and child gender. Fathers of girls reported steep increases in parenting stress across the early childhood period, while fathers of boys reported a more gradual increase across both early and middle childhood. In contrast, other studies have found no such effect of child chronological age and gender on paternal stress (e.g., Herring et al., 2006). Thus, the influence of child chronological age and gender on paternal stress in families with children with developmental disabilities is unclear.

The effect of child chronological age and gender on parenting stress specifically in families with children with FXS is also unclear. To date, few studies have reported the effects of child chronological age and gender on parents in this population (Johnston et al., 2003; Hall et al., 2007), despite most studies controlling for these variables in analyses. Importantly, recent findings of studies with FXS including Johnston et al. (2003) and Hall et al. (2007) demonstrated results corresponding with a broader range of disability research, suggesting that neither child chronological age nor gender typically have a substantial impact on parents' stress levels.

### *Parent and Family Variables*

The diagnosis of a developmental disability in a child can elicit various behavioral and emotional responses in parents and family systems, frequently placing additional demands on limited time and resources. Increasingly fathers in addition to mothers play a significant role in caretaking routines. However, mothers typically remain the primary caregiver and therefore take on much of the burden of care associated with raising a child with a disability (Pleck, 1997; Young & Roopnarine, 1994). Scholars now emphasize that parent adaptation and child development occur within the context of an ecological system (Sameroff & Fiese, 2000), where child characteristics, the immediate family, and the

community environment interact with each other. Inherently in this model, individuals and systems mutually influence each other and therefore changes in one family member will impact other members and the family system as a whole. The ability of any stressor to bring about either family crisis or adaptation is then a product of characteristics of the context within which the stressor occurs and can be moderated by the resources that the family has to deal with that stressor. Thus, knowledge of parent and family characteristics such as family income, parent education, social support, and gender are important to our understanding of predictors and moderators of developmental change and parent adaptation that are amenable to intervention (Hauser-Cram et al., 2001).

Given the heritability of FXS, many parents find themselves in a unique situation of needing to understand complicated genetic information and bear the responsibility of passing this information on to extended family members (Bailey et al., 2003). Passing on information regarding the heritability of FXS can often be a difficult and stressful process that sometimes has negative consequences for family relationships (Bailey et al., 2003). Parents, especially mothers, may not only experience guilt relating to transmitting a disorder to their child, but also anger and resentment regarding the diagnosis and subsequent change in how decisions must be made related to reproductive planning (Bailey et al., 2003). Furthermore, greater than half of families with children with FXS have additional children without the knowledge of the reproductive risk due to its late diagnosis (mean age 35 months; Bailey et al., 2000). Therefore, parents must frequently bear the additional burden and potential stress of raising more than one child with a developmental disability. This unique constellation of factors provides a rare and informative opportunity to examine how

multiple facets influence parenting stress in families with a child with a developmental disability.

*Family income.* Traditionally, household income has served as an indicator of socioeconomic status (SES) and is commonly controlled for in analyses due to a frequent inverse relationship with parenting stress (Burbach et al., 2004; Koeske & Koeske, 1990, Shin et al., 2006; Smith et al., 2001; Warfield et al., 1999). Literature suggests that families of low SES experience more ongoing strains, making them especially vulnerable or emotionally reactive to additional stressors from raising a child with a developmental disability (Thoits, 1995). In addition, limited finances potentially exacerbate parenting stress by restricting the number and quality of coping resources available for parents in times of strain. Although most studies with FXS report collecting family income in order to control for it, only one study described its influence on parenting stress (Johnston et al., 2003). Corresponding with previous research, Johnston et al. (2003) found that lower family income contributed to higher parenting stress.

*Parent education.* Due to the strong relationship that exists between educational attainment and income one would expect that educational attainment, similar to family income, acts as a stress buffer and therefore higher levels of parent education would relate to lower levels of parenting stress. However, research has indicated that families of children with disabilities are often more traditional in the way they divide family roles. Mothers tend to stay home and provide the majority of the child care, while fathers are often the sole or primary bread winners (Sen & Yurtsever, 2007; Simmerman et al., 2001). This leads to lower family incomes unrelated to educational attainment. Therefore, it is not surprising that studies examining the relationship between parent education and parenting stress have produced

mixed results (Abbeduto et al., 2004; Boyce & Behl, 1991; Hodapp et al., 1998; Koeske & Koeske, 1990; Lecavalier et al., 2006; Ricci & Hodapp, 2003; Smith et al., 2001; White & Hastings, 2004).

*Social support.* Social support is considered a coping resource from which individuals may draw when handling stressors (Thoits, 1995). It is a common assumption that social support such as positive family relationships/cohesion, spousal support, and an individual's social network act as protective factors against heightened levels of parenting stress. In fact, a number of studies have shown that actively targeting social support tends to lead to improvements in parental coping including stress (Baker-Ericzén, Brookman-Fraee, & Stahmer, 2005; Kirkham, 1993; RUPP Autism Network, 2007; Singer et al., 1999).

Additionally, social support is associated with parenting stress after other salient variables such as child functional abilities, child behavior problems, family income, and parent mental health have been accounted for (e.g., Johnston et al., 2003; Smith et al., 2001; White & Hastings, 2004). Research involving families with FXS affirms this, demonstrating that lower levels of family cohesion are associated with higher levels of parenting stress or related parent outcome variables such as symptoms of depression and anxiety (Bailey et al., 2007; Hall et al., 2007; Johnston et al., 2003; Sarimski, 1997). Similarly, von Gontard et al. (2002), with a sample of mothers and fathers of children with FXS, found that higher parenting stress was associated with less mobilization of the family to acquire and accept social support, particularly external support. As well, in a qualitative study conducted by Poehlman et al. (2005), almost all mothers interviewed spoke to the importance of social support for their individual as well as their family's adaptation to the child's diagnosis. One of the most salient themes in their interviews was that a lack of support from others, particularly from

spouses, family members and friends, made their initial and ongoing adaptation to the child's diagnosis more difficult.

In examining a broader range of literature addressing social support in families with children with developmental disabilities other than FXS, the salience of specific types of support becomes apparent. For example, some studies have revealed that informal support from spouses, family, or friends may be more important in reducing parenting stress than formal supports from professionals and services (e.g., Saloviita et al., 2003; White & Hastings, 2004). As well, several studies indicate the particular significance of spousal support for parents' experienced stress (Kersh et al., 2006; Saloviita et al., 2003; van Lieshout et al., 1998). In fact, spousal support has proven itself as an important contextual factor for overall parent wellbeing, predicting not only parenting stress but also anxiety and depressive symptoms, and observed parenting behavior (Bristol et al., 1988; Kersh et al., 2006; White & Hastings, 2004). The perception of support from ones' spouse may indirectly influence parenting stress by influencing the parent's perception of his or her child with the disability. For example, Cuskelly and Dadds (1992) found that marital satisfaction significantly predicted how mothers' perceived their children in that mothers' marital satisfaction predicted their reports of their child's problem behavior.

To date, few studies have delineated differences in perceived social support between mothers and fathers in families with children with disabilities. Recent research generally shows that mothers and fathers report similar levels of social support across various measures (Hall et al., 2007; Keller & Honig, 2004; McCarthy et al., 2006). Yet, preliminary data suggest that social support is a more important contributor to maternal stress than paternal stress (Hauser-Cram et al., 2001; Krauss, 1993; Wanamaker & Glenwick, 1998). In relation,



mothers express more need for supports than fathers of children with disabilities (Bailey, Blasco, & Simeonsson, 1992). Given research suggesting a greater risk for social anxiety in women with the premutation of FXS in comparison to the general population (Franke et al., 1996; Franke et al., 1998), the likelihood of differences between mothers and fathers in the types and contribution of social support factors may be increased. Thus, delineating differences in maternal and paternal social support factors in families with children with FXS will be essential for our understanding of how best to promote successful coping, reducing and managing parents' stress.

*Parent gender.* Studies investigating parenting stress in families with children with disabilities have predominantly focused on mothers; however, increasingly studies are including fathers, making it possible to begin examinations of similarities and differences between maternal and paternal stress. As noted, comparing mothers' and fathers' parenting stress in families with FXS is particularly important due to evidence of maternal vulnerability to stress related to their premutation status. There is some debate regarding differences between mothers and fathers with respect to parenting stress. Previous research suggested that fathers' experience lower parenting stress compared with mothers in families with children with disabilities (e.g., Kazak & Marvin, 1984). However, current literature indicates that mothers and fathers more often than not report similar levels of parenting stress (Hastings et al., 2005; Keller & Honig, 2004; Wanamaker & Glenwick, 1998). Using a sample of 27 mother-father dyads with a child with FXS, McCarthy et al. (2006) gave support for this finding in fragile X literature, reporting no significant differences in mothers' and fathers' ratings of parenting stress. Furthermore, McCarthy et al. (2006) found no differences in other parent outcomes related to parenting stress including somatic and

psychosomatic symptoms. These findings are particularly interesting given the assumption that mothers with the premutation would report higher parenting stress, as their ability to cope with the additional stressors of raising a child with a disability might be negatively impacted by a biological vulnerability. Unfortunately, to the author's knowledge no other studies with FXS appear to exist that have examined parenting stress comparing mothers and fathers.

In addition to the similarity in the amount of parenting stress reported by mothers and fathers of children with disabilities, variables that explain increases in their parenting stress are often similar. For example, Hauser-Cram et al. (2001) found that child problem behavior was a common predictor of change in parenting stress for both mothers and fathers. Differences, however, have been evidenced in the magnitude of contribution each variable makes on maternal versus paternal stress. Some preliminary research suggests that sources of stress related to dimensions of parent functioning (e.g., parental health, role restriction, psychopathological symptoms) and personal social support are more important for maternal stress. In contrast, parenting stress related to parent-child attachment and child characteristics that influence parents' interactions with their children (i.e., acceptability, gender, adaptability, mood, reinforcement), are more important for paternal stress (Hastings et al., 2005; Johnston et al., 2003; Keller & Honig, 2004; Koeske & Koeske, 1990; Krauss, 1993; McBride et al., 2002; Saloviita et al., 2003). Although an oversimplification, simply put, parent-related stress seems to be higher for mothers, while child-related stress seems to be higher for fathers. Noh et al.'s (1989) findings support this hypothesis. In their study mothers were more likely than fathers to be at risk for clinically significant parent domain stress scores. On the other hand, fathers were more likely than mothers to be at risk for clinically

significant child domain stress scores. Similarly, Krauss (1993) revealed that mothers had higher parenting stress than fathers related to their perceptions of their health, restrictions in their role, and relations with their spouse. In comparison, fathers had higher parenting stress than mothers regarding perceptions of their child's adaptability, mood, reinforcement and attachment with them. Finally, Hauser-Cram et al. (2001) revealed differences in parenting stress trajectories for mothers with high versus low levels of social support, yet no differences in parenting stress trajectories for fathers based on differences in reported social support. Yet, as noted earlier, research outlining differences in maternal and paternal stress is best stated to be uncertain, as there has been significant limitations in previous studies' designs and a general lack of use of statistical analyses which allow for direct comparison between mothers and fathers.

### *Summary*

Parenting stress has been proven to be a construct fundamental to our understanding of parents' experience. It has been well documented that parenting stress is high across families of children with disabilities, including families with children with FXS, when compared with parents of typically developing children. Several variables contribute to parents' stress in families with children with FXS including child characteristics and dimensions of the parent, family and environment. Of the child characteristics, problem behavior appears to be a major source of parenting stress for both mothers and fathers, with the degree of behavior problems associated with the child's disability making a more substantial contribution to parenting stress than the child's functional limitations. Social support factors such as spousal support, family relationships, and the wider social support network may help to reduce parents' stress; however, with few studies delineating the

differences and similarities in mothers' and fathers' parenting stress little is known about what support resources may be more important for fathers compared to mothers. Delineating these differences will be important for the development of effective parent interventions that address the needs of both mothers and fathers. As well, reducing parenting stress in mothers with the premutation of FXS may be especially important for preventing the onset and reoccurrence of affective and anxiety disorders for which women with the premutation are at heightened risk.

### *Study Aims*

The purpose of this study was to address the gap in current literature by describing and directly comparing parenting stress in mothers and fathers of children with FXS. The study completed an across parent gender examination of the relationship of social support and parenting stress in order to: (a) initiate research regarding the difference in the magnitude of contribution of variables to maternal versus paternal stress, and (b) advance research that improves the development of effective parent interventions. Given preliminary research comparing parenting stress in mothers and fathers of children with disabilities and present literature on families with children with FXS, the following research questions were posed: (1) What are the commonalities and differences in the parenting stress exhibited by mothers and fathers of children with fragile X syndrome?; (2) Are there significant differences between mothers and fathers perceptions of the social support network?; (3) Does the association of social support to parenting stress differ in mothers versus fathers of children with fragile X syndrome?

## CHAPTER III

### Methods

The data for this study were collected as part of a study at the Frank Porter Graham Child Development Institute (FPG), at the University of North Carolina at Chapel Hill (UNC-CH), which addressed a wide range of questions related to family adaptation in FXS. The *Family Adaptation to Fragile X Syndrome* study was a longitudinal study designed to collect data from a minimum of 100 families with children with full mutation FXS who are between the ages of 1 year and 12 years of age at the time of study entry. In all of the families the mother was a carrier or had the premutation of FXS. Core measures for the family adaptation study were collected from mothers and their children three times, at 18-month intervals, across a 5-year period. Although the original study proposal included only mother and child data, data from fathers were collected during the third and final round of data collection due to recognition of the importance of the father's role. Because data from fathers is an integral component of this study, only data from the third and final data collection point of the *Family Adaptation to Fragile X Syndrome* study was used in the current study.

#### *Participants*

One hundred and eight families including a target child with FXS were recruited and have participated in the larger ongoing study, *Family Adaptation to Fragile X Syndrome*. These participants were recruited through three main sources: (1) currently funded projects at UNC-CH that had an enrolled sample of children with FXS; (2) the FX Subject Registry

Core of the UNC Neurodevelopmental Disorders Research Center

([www.research\\_registry@unc.edu](mailto:www.research_registry@unc.edu)); and (3) posting of notices on the fragile X parent list serve, by contacting fragile X family support groups, and distributing brochures to other investigators and professionals in the field of FXS. A review of maternal and child DNA reports established that 95 mothers had the premutation and 13 mothers had the full mutation of FXS, while all of their sons and daughters had full mutation FXS.

The present study sample consisted of a subset of families from the larger family adaptation to FXS study who participated in the third and final data collection ( $n=96$ ). Inclusion criteria for this study were as follows: (1) mothers have the premutation of FXS; (2) target children are between the ages of 3-10 years at time of third and final assessment and have full mutation FXS; and (3) complete data from mother-father dyads consisting of biological mothers and their cohabitating spouse or partner (i.e., biological and step fathers). Thus, the present study analyzed data gathered from triads consisting of a mother, father, and target child with FXS. Several factors were considered in developing criteria to minimize the potential effects of confounding variables. First, the study restricted its sample to mother-father dyads in order to eliminate effects associated with including data from non-cohabitating parents and to ensure that contextual factors for mothers and fathers are consistent. Second, families were only included if the mother had the premutation form of FXS. Exclusion of mothers with the full mutation ensures that stress levels are not unduly influenced by a lack of coping mechanisms related to intellectual disability, learning disabilities, or psychosocial difficulties reported in some females with full mutation FXS (Mazzocco, 2000). An age range of 3 years-6 months to 10 years-6 months for the target child was selected in order to maximize the sample size, and therefore statistical power of

analyses, while keeping within the early to middle childhood age range in order to minimize error variance. The decision to have an extended age range is consistent with existing studies that included target children ranging in age from 6 to 17 years (Johnston et al., 2003) or 4 to 17 years (McCarthy et al., 2006). This precedent for extended age range is further supported by research suggesting a lack of child chronological age affect on parenting stress (Johnston et al., 2003; Hall et al., 2007).

Out of 96 families from the *Family Adaptation to Fragile X Syndrome* study, 38 families met inclusion criteria for this study, including 76 parents (38 mother-father dyads) and their male ( $n=30$ ) or female ( $n=8$ ) child with FXS. Three of the 38 parent-dyads reported to be cohabitating but are not formally married. The target children have a mean age of 68.74 months ( $SD= 20.64$ , range= 42.22-126.65). Target mothers' mean age at the time of data collection was 37.70 years ( $SD= 4.88$ , range= 24.86-46.14). The mothers have an average of 15.97 years of education, most mothers having post-secondary education ( $SD= 1.78$ , range= 12.00-19.00). Target fathers mean age at the time of data collection was 40.35 years ( $SD= 5.58$ , range= 25.99-54.60). The fathers have an average of 16.32 years of education, most fathers having post-secondary education ( $SD= 1.89$ , range= 13.00-19.00). An estimated 11% ( $n= 4$  of 38) of the families are categorized as low-income, defined as having an annual income less than 200% of the federal poverty threshold. Families are predominantly European American, with 33 parent dyads reporting their ethnicity as White, 4 as African American, and 1 as Latino. The mean number of children per family with or without FXS is 2.13 ( $SD= .62$ , range= 1.00-3.00). The mean number of children with FXS per family is 1.45 ( $SD= .56$ , range= 1.00-3.00), 22 families have one child with FXS, 15 families have two children with FXS, and one family has three children with FXS.

### *Protection of Human Participants*

The protocol and procedures are approved by the IRB (05-0106), and all parent participants provided written informed consent for their own and their child's participation in the study. Parents received a participant stipend and target children received an inexpensive gift (approximately \$5.00 value).

### *Measures*

Data from multiple measures was used in analyses for the study. Rating scales of parental functioning including parenting stress (Parenting Stress Index) and social support factors (Personal Assessment of Intimate Relationships Inventory, Family Environment Scale, and Family Support Scale) were used to measure data on the dependent and independent variables respectively. Rating scales of child characteristics (adaptive behavior and problem behavior) as well as a demographic survey (child age, child gender, family income, number of children in the household with full mutation FXS) were used to collect data on the control variables.

### *Parent Measures*

*Parenting Stress Index Short Form (PSI)*. In order to assess the magnitude of stress in the parent-child system, the short form of the PSI (Abidin, 1995) was completed by both mothers and fathers in each family. Parents rate each item using a Likert scale ranging from 1 (Strongly agree) to 5 (Strongly Disagree). The PSI Short Form is a self-report rating scale made up of 36-items, which yields 3 factor scores (Parental Distress, Difficult Child, and Parent-Child Dysfunctional Interaction) and a Total Stress score. The higher scores indicate a greater magnitude of stress. Total scores greater than 91 represent high stress scores in a



clinically significant range. For this study the Total Stress score as well as all three factor scores were used in analyses.

*Personal Assessment of Intimate Relationships Inventory (PAIR)*. Perceived emotional intimacy with ones partner was assessed using the PAIR (Schaefer & Olson, 1981). The PAIR has a long-standing record for its usefulness in both clinical and research contexts. The entire inventory consists of 36 items including Emotional, Social, Sexual, Intellectual, and Recreational Intimacy subscales. For this study only the Emotional Intimacy subscale was administered. This subscale documents closeness and feelings of support from ones partner and served as a measure of spousal support.

*Family Environment Scale—Family Relations Index (FES)*. The FES (Moos, 1974) was used to assess family relationships. When used in its entirety the FES is a 90-item scale assessing perceived family climate. Each item describes a potential family characteristic (e.g., “There is a feeling of togetherness in our family”) which the respondent rates as true or false. The entire inventory of the FES is composed of 10 subscales; however, only three subscales (i.e., Cohesion, Expressiveness, and Conflict) which comprise the Family Relations Index were administered as the Family Relations Index best represents the relationship dimensions important for the support construct. Adequate internal consistency reliability and stability has been reported when applied to a diverse range of samples and the items have good content and face validity (Moos, 1990). Additionally, the construct, concurrent, and predictive validity of the FES are supported by research (Moos & Moos, 1986). In Koranek’s (1989) review of the measure citing more than 200 studies, he states, “the FES is the most widely used and validated self-report measure of family functioning” (p.71). The cohesion and expressiveness subscale scores are summed and the conflict subscale score is subtracted,

resulting in a Family Relations Index score. Consistent with previous research (Hauser-Cram et al., 2001) the Family Relations dimension was used as a measure of family support.

*Family Support Scale (FSS).* The FSS (Dunst, Trivette, & Jenkins, 1988) was used to assess perceived support from family, friends, and the community or ones perceived social network support. The FSS is an 18-item self-report rating scale. Each item lists a source of support and is rated on a 5-point scale from “not at all helpful” (1) to “extremely helpful” (5). The scale is organized into four empirically derived factors: Support from Family, Support from Friends, Informal Support, and Formal Support. The FSS has high internal consistency and adequate test-retest reliability as indicated in studies (Dunst, Trivette, & Hamby, 1994). Hauser-Cram et al. (2001) demonstrated its usefulness in a longitudinal study of child development and parent well-being, finding that social support as measured by the FSS predicted change in parenting stress in families with children with disabilities. Several studies have examined the factor structure of the Family Support Scale, but each reported a different structure. Therefore, for the present study only one score representing Total Social Support was used in regression analyses.

*Structured Clinical Interview for the DSM-IV-I: Non-patient Edition (SCID-I/NP).* The SCID-I/NP (First, Spitzer, Gibbon, & Williams, 2002; First, Spitzer, Gibbon, & Williams, 1997) is a semi-structured diagnostic interview that was used to evaluate all mothers for mood and anxiety disorders. The SCID-I/NP systematically evaluates current and past psychiatric symptomatology for DSM-IV psychotic, mood, substance use, anxiety, somatoform, post traumatic stress disorder, eating, and personality disorders. For this study, only the mood and anxiety disorder modules were administered. This investigator was trained in the administration of the SCID and administered the SCID-I/NP to each mother. A

typical SCID-I/NP administration required 45 minutes of the mother's time. Fathers were not administered this measure. The following variables were derived from SCID-I/NP data to include in exploratory analyses: (1) mother met current diagnosis for a mood and/or anxiety disorder at time of data collection versus no current diagnosis, and (2) mother met for lifetime history of mood and/or anxiety disorder at time of data collection versus no history of diagnosis.

### *Control Variables*

*Demographic Questionnaire.* Demographic information was gathered on each family. The demographic questionnaire reflects information about the parents' ethnic background, age, marital status, family income, education, and employment. This form also reflects information about the child's age, FXS diagnosis date, ethnic identity, and number of individuals in the home. Several variables were derived from this questionnaire to include in analyses for the present study including child gender, child chronological age, family annual gross household income, low income versus middle-upper income, number of children in household, number of children in household with full mutation FXS, father residing in the home, and time in months since target child's FXS diagnosis.

*Medication History Questionnaire.* Information was gathered regarding mothers' and target children's medication usage. The medication history questionnaire reflects information about the medication's name, category (i.e., stimulant, sympatholytic, SSRI, tricyclic, anticonvulsant, antipsychotic, benzodiazepine, anxiolytic, beta-blocker, antihistamine, other antidepressants, SNRI, hypnotic, and mood stabilizer), prescribing doctor, dose, place of administration, target symptoms, and start and stop dates. Two variables were derived for the

present study: (1) mother's psychotropic medication use on or off, (2) target child's medication use on or off.

*Child Behavior Checklist (CBCL).* CBCL for ages 1 ½ to 5 years or 6 to 18 years (CBCL/6-18; Achenbach, 2001; CBCL/1½-5; Achenbach & Rescorla, 2000) were used to assess the child's problem behavior based on child's activities, social relations, and school performance. The CBCL/1½-5 and CBCL/ 6-18 are widely used standardized rating scales describing specific emotional and behavioral problems and are 99-items and 118-items respectively. Parents rate their child for how true each item is now or within the past 6 months using the following scale: 0 = not true (as far as you know); 1 = somewhat or sometimes true; 2 = very true or often true. Across both rating scales parent ratings produce scores indicating the presence and severity of problems on the following factors: Aggressive Behavior, Anxious/Depressed, Attention Problems, Somatic Complaints, Withdrawn. In addition to these factors, the CBCL/1½-5 produces scores for Emotionally Reactive and Sleep Problems, while the CBCL/6-18 produces scores for Rule-Breaking Behavior, Social Problems, and Thought Problems. These factors contribute to three broad scales: Internalizing, Externalizing, and Total Problems. Test-retest reliability for the CBCL/1 ½-5 Total Problems score is at .90 and at .93 for the CBCL/6-18 Total Problems score.

For this study, the mother of each child participant completed the version of the CBCL which corresponded to her child's age. Studies have shown consistency between mother and father ratings of problem behavior using the CBCL, especially in regards to externalizing behavior. In Achenbach, McConaughy, and Howell (1987) review of the measure citing more than 250 samples in 119 studies, on average they found statistically significant ( $p < .001$ ) correlations of .60 between similar informants (e.g., pairs of parents),

with many studies reporting correlations as high as .70 to .80. As well, they reported that correlations as significantly higher for children up to 11-year-olds than for adolescents (12-19 years of age) and found no significant differences between the sexes and between mothers and fathers ratings. For the present study, the CBCL Total Problems score was used in analyses.

*Vineland Adaptive Behavior Scales, Interview Edition (VABS).* The VABS (Sparrow, Balla, & Cicchetti, 1984) was used as a global measure of overall child functional status and abilities, including estimating the child's level of personal and social sufficiency. The VABS contains 265 items in a semi-structured interview format: 67 items measuring communication, 92 items measuring daily living skills, 66 items measuring socialization, and 36 items measuring motor skills, each of which has a rating of never, sometimes/partially, or yes. The VABS produces an Adaptive Behavior Composite (ABC) standard score (Mean of 100, standard deviation of 15). Additionally, it has test-retest reliability ( $r=.88$ ) as well as acceptable content and criterion-related validity. For this study, the mother of each child participant was interviewed using the VABS. The Adaptive Behavior Composite was used in analyses representing child's overall functional abilities.

#### *Procedures*

As noted earlier, all subjects were tested as part of an ongoing study examining family adaptation to FXS. To determine family eligibility, existing genetic reports were reviewed for mothers and their targeted children. Data were gathered in two stages. First, parents were mailed a set of rating scales to complete prior to a scheduled assessment visit. Mother and father rating scales were separated into individual envelopes, with each envelope providing independent instructions directed to either the mother or the father. Second, an

assessment visit was completed with mothers and the target child at which time rating scales were collected from each parent and reviewed for completion. For the present study four rating scales (PSI, PAIR, FES, & FSS) were completed by both mothers and fathers. In addition to these rating scales, measures including a child problem behavior rating scale (CBCL), an interview regarding the target child's functional abilities (VABS), an interview regarding mothers' mental health status (Structured Clinical Interview for the DSM-IV-I: Non-patient Edition), a demographic questionnaire, and a medication history questionnaire were completed by mothers only.

## CHAPTER IV

### Results

#### *Preliminary Analyses*

Preliminary analyses were conducted to assess the normality of the distributions of variables and test the assumptions of the models employed. No out of range or suspicious values were found in the data. In addition, distributions of all scaled measures, including the PSI, PAIR, FES, FSS, CBCL, and VABS met assumptions of normality with the highest recorded kurtosis less than 3.96 ( $SE = .75$ ) and the highest recorded skewness less than -1.71 ( $SE = .38$ ). Following examination of distributions, inter-correlations of independent and control variables were completed separately for mothers and fathers to investigate collinearity between variables in order to avoid problems in estimating multi-level model regression coefficients. These correlation matrices also were utilized as a data reduction strategy. The correlation matrices for mothers and fathers are presented in Tables 1 and 2 respectively. Due to the high correlation found between child age and adaptive behavior ( $r = -.51, p < .01$ ) and literature that suggests child adaptive behavior and problem behavior share a significant amount of variance with parenting stress (Britner et al., 2003; Hastings, 2002), the VABS (child adaptive behavior) was dropped from control variables included in the hierarchical linear models. As well, the spousal support measure (PAIR) was dropped from the independent variables included in hierarchical linear models due to its high correlation with the family support measure (FES) for both mothers ( $r = .60, p < .001$ ) and fathers ( $r = .60, p < .001$ ). A significance level of  $p < .05$  was established *a priori*.

## *Descriptive Statistics*

### *Parent Descriptives*

Descriptive statistics of mothers' and fathers' ratings on independent and dependent variables are provided in Table 3. Data for all mother-father dyads, who represent a sub-sample of parents from the larger longitudinal study on family adaptation to fragile X syndrome, are included in these values. In addition to descriptive statistics on primary measures, information on mothers' mental health status and medication use was collected and summarized. At the time of data collection, out of 38 mothers, six mothers met for a current diagnosis of a mood and/or anxiety disorder including three mothers who met criteria for a current anxiety disorder, one mother who met for major depressive disorder, and two mothers who met for major depressive disorder and one or more anxiety disorder. Twenty-two of the 38 mothers had a lifetime history of a mood and/or anxiety disorder (including the six mothers who met for current diagnoses). Four mothers met criteria for a lifetime history of an anxiety disorder, 10 mothers met for a major depressive disorder, and eight mothers met for a major depressive disorder and one or more anxiety disorder. Fifteen mothers reported current use of a psychotropic medication. Finally, an investigation of mean differences between mothers on versus off psychotropic medication was completed. This investigation revealed no significant group differences in Total Stress or any of its three factor scores. Furthermore, there was a lack of a correlation between mothers' medication status and their level of parenting stress.

### *Child Descriptives*

Using suggested cut-offs, target children in the sample, as a group, display overall problem behavior within normal limits. However, 24% of children obtained total problem



behavior scores in the borderline range of functioning and 13% obtained total problem behavior scores in the clinically significant range of functioning. Target children exhibited universally low adaptive behavior functioning, with 89% of children in the sample obtaining an Adaptive Behavior Composite score two or more standard deviations below the mean. Table 4 provides a breakdown of the major indices for the CBCL and VABS. The mean time which had elapsed between the target child's FXS diagnosis and the point of data collection was 57.48 months ( $SD = 21.34$ , range= 25.71-126.02). Per mother report, 22 of the 38 target children were currently on medication at the time of data collection.

#### *Primary Analyses*

*Question 1: What are the commonalities and differences in the parenting stress exhibited by mothers and fathers of children with fragile X syndrome?*

To compare maternal and paternal stress, mean difference tests between mothers' and fathers' on the PSI Total Stress score and its three factor scores, Difficult Child (DC), Parental Distress (PD) and Parent-Child Dysfunctional Interaction (P-CDI), were computed using paired-samples t-tests to correct for within family clustering. Results from these analyses revealed no significant mean group differences between mothers' and fathers' reported parenting stress on the Total Stress score,  $t(37) = .61, p = .55$ , or any of its three factor scores, DC  $t(37) = .27, p = .79$ ; PD  $t(37) = 1.78, p = .08$ ; P-CDI  $t(37) = -.96, p = .34$ . Please refer to Table 3 for mother and father parenting stress mean values.

Comparisons between proportions of mothers and fathers with scores in the clinically significant range on the PSI were made. Counts were generated separately for mothers and fathers for the number of parents that obtained Total Stress, DC, PD, P-CDI scores above the suggested cut-offs. Next, four separate chi-square tests of independence compared mothers'

and fathers' frequencies of clinically significant scores on total parenting stress and each of its three factors. No parent gender differences in the number of mothers versus fathers with parenting stress scores above clinical cut-offs were found for any of the PSI scores, all  $\chi^2 \leq .23$  (1,  $N = 76$ ) and all  $p \geq .63$ . Overall, there were 13 mothers and 12 fathers out of the total 76 parents (for a combined 32.89%) that had Total Stress scores in the clinically significant range (i.e.,  $\geq 90^{\text{th}}$  percentile). Fourteen mothers and 12 fathers obtained clinically significant DC scores, six mothers and five fathers obtained clinically significant PD scores, and 13 mothers and 13 fathers obtained clinically significant P-CDI scores.

Finally, inter-parent agreement was assessed with intra-class correlation coefficients. Correlations revealed that mothers' and fathers' DC scores had a near zero correlation, DC  $r(37) = .08$ ,  $p = .31$ ; while, mothers' and fathers' PD, P-CDI and Total Stress scores were moderately correlated, PD  $r(37) = .33$ ,  $p = .02$ ; P-CDI  $r(37) = .31$ ,  $p = .03$ ; Total Stress  $r(37) = .25$ ,  $p = .06$ .

*Question 2: Are there significant differences between mothers and fathers perceptions of the social support network?*

Overall, mothers and fathers reported having the most satisfaction with social support from their spouse or partner, the target child's school or daycare and professional helpers. Mothers and fathers in the sample reported accessing an average of 13.66 ( $SD = 2.84$ , range= 5-17) and 13.47 ( $SD = 2.84$ , range= 9-18) different types of social support respectively. In addition, mothers and fathers provided similar ratings of satisfaction for the various supports. Table 5 shows the proportion of mothers and fathers accessing each type of social support from the FSS.

Paired-samples t-tests were used to make comparisons of parental mean values on each of the three social support measures (i.e., PAIR, FES, FSS) to examine differences between mothers' and fathers' perceptions of social support. Results from these analyses revealed no significant mean differences by parent gender on any of the social support measures, PAIR  $t(37) = -1.93, p = .06$ ; FES  $t(37) = 1.61, p = .12$ ; FSS  $t(37) = -.47, p = .64$ . Please refer to Table 3 for mother and father social support mean values.

Again, inter-parent agreement on measures was examined. Inter-parent agreement on the PAIR, FES, and FSS support scores were assessed using intra-class correlation coefficients. Correlations revealed that mothers' and fathers' spousal support score (PAIR Emotional Intimacy Scale score) was highly correlated,  $r(37) = .61, p < .001$ . However, mothers' and fathers' ratings of family support (FES Family Relations Index score) and social network support (FSS Total Support score) covaried to a low degree, FES  $r(37) = .18, p = .14$ ; FSS  $r(37) = .30, p = .03$ .

*Question 3: Does the association of social support to parenting stress differ in mothers versus fathers of children with fragile X syndrome?*

To investigate the relationship between social support and parenting stress across parent gender, a series of hierarchical linear models (HLM) were conducted. A hierarchical linear model is a type of regression model appropriate for data with a nested structure. Analyses were therefore conducted using HLMs in order to correct for within family clustering (parents nested within family). Other benefits of HLM include its robustness and fewer restrictive assumptions compared to other statistical analyses (e.g., less sensitive to violations of normality and missing data). In the present study, all HLMs allowed for random intercepts. Categorical data were dummy-coded for analysis. Mothers were the reference

group (coded “0”) for the parent gender variable. Income was categorized to two levels, with low-income, coded “0” (based on DHHS guidelines of equal to or below 200% of poverty and factoring in family size), as the reference group. Finally, the number of full mutation FXS children in the family was also dichotomized such that families with multiple children with FXS (2 or 3) were considered a single group and compared to families with just one child with FXS, the target child. Families with just one child with FXS were the reference group and therefore coded “0”. All continuous variables were mean centered prior to analysis.

Due to the low number of female target children (nine) and research suggesting a negligible affect of child gender on parenting stress when other salient child characteristics are accounted for (e.g., problem behavior), child gender was dropped from the models. Control variables included for all HLMs were family household income, number of children in the home with full mutation FXS, child chronological age, and child problem behavior. Independent variables for all HLMs included maternal and paternal ratings of family support (FES Family Relations Index score) and social network support (FSS Total Support score), with parent gender (mother versus father) being added as an additional independent variable. Interaction terms for parent gender by family support and parent gender by social network support were also included. These interaction terms allowed for direct comparison of the relationship of the two social support factors with parenting stress for mothers versus fathers. Outcome variables of parenting stress (i.e., DC, PD, P-CDI, Total Stress scores) were run in four separate HLMs. As a result of the small sample size, extreme outliers would need to be present in order to reject the null hypothesis that the random effects of the models were

normally distributed. As noted earlier, all data were screened prior to analyses and no such outliers were found to be present.

*Child domain stress.* Results from analysis with PSI Difficult Child score as the outcome variable are presented in Table 6. No significant interactions were found in the analysis, suggesting that the relationship of social support to child domain stress is similar for mothers and fathers, FES\*parent gender  $F(1, 56.84) = .56, p = .46$ ; FSS\* parent gender  $F(1, 54.35) = .00, p = .98$ . Main effects on parenting stress were found for family support,  $F(1, 60.71) = 6.62, p = .01$  and for child problem behavior,  $F(1, 36.92) = 4.65, p = .04$ . As family support decreased child-related stress increased for both mothers and fathers. As well, as child problem behavior increased, child-related stress increased. No other independent or control variables demonstrated significant effects on child domain stress.

*Parent domain stress.* Results from analysis with PSI Parental Distress score as the outcome variable are presented in Table 6. No significant interactions were found in the analysis, suggesting that the relationship of social support to parent domain stress is similar for mothers and fathers, FES\*parent gender  $F(1, 50.95) = .01, p = .93$ ; FSS\* parent gender  $F(1, 48.92) = .22, p = .64$ . Main effects on parenting stress were found for family support,  $F(1, 54.44) = 7.83, p < .01$ , and for parent gender,  $F(1, 36.18) = 10.23, p < .01$ . Mothers reported significantly greater parental distress than fathers and as family support decreased parental distress increased for both mothers and fathers. No other independent or control variables demonstrated significant effects on parent domain stress.

*Parent-child domain stress.* Results from analysis with PSI Parent-Child Dysfunctional Interaction score as the outcome variable are presented in Table 6. Again, no significant interactions were found in the analysis, suggesting that the relationship of social

support to parent-child domain stress is similar for mothers and fathers, FES\*parent gender  $F(1, 53.61) = .14, p = .71$ ; FSS\* parent gender  $F(1, 51.25) = .17, p = .68$ . A main effect on parenting stress was found for child problem behavior  $F(1, 38.93) = 10.39, p < .01$ . Also a near significant main effect was found on parenting stress for family support,  $F(1, 57.50) = 3.89, p = .05$ . As child problem behavior increased parent-child interaction stress also increased. As family support decreased parent-child interaction stress increased for both mothers and fathers. No other independent or control variables demonstrated significant effects on parent-child domain stress.

*Overall parenting stress.* Results from analysis with PSI Total Stress score as the outcome variable are presented in Table 6. No significant interactions were found in the analysis, suggesting that the relationship of social support to overall parenting stress is similar for mothers and fathers, FES\*parent gender  $F(1, 50.20) = .57, p = .45$ ; FSS\* parent gender  $F(1, 47.98) = .03, p = .86$ . However, main effects on parenting stress were found for family support,  $F(1, 54.02) = 12.69, p < .01$ , and for child problem behavior,  $F(1, 39.05) = 6.38, p = .02$ . Overall parenting stress increased as family support decreased and child problem behavior increased. A near significant main effect was found on parenting stress for parent gender,  $F(1, 34.02) = 3.94, p = .06$ , such that mothers reported significantly greater total parenting stress than fathers. No other independent or control variables demonstrated significant effects on parenting stress.

#### *Exploratory Analyses*

Due to the very limited research comparing maternal and paternal stress in families with children with fragile X syndrome as well as the lack of clarity from primary analyses regarding differences found in maternal versus paternal stress, brief exploratory analyses

were undertaken. It was the hope of the author that these analyses could help better inform the direction of future research. Exploratory analyses were comprised of two separate correlation matrices, one for mothers and one for fathers. These matrices included a greater range of family and child characteristics that may have an important influence on parenting stress. Table 7 contains the results for mothers and Table 8 contains the results for fathers.

In general, correlation matrices for mothers and fathers were similar, with the majority of family and child variables included in the matrices demonstrating weak associations with maternal and paternal stress. Two variables, household income and child adaptive behavior, demonstrated unexpectedly low associations to maternal and paternal stress, with  $r(37) \leq .08, p \geq .62$  for household income and  $r(37) \leq .16, p \geq .34$  for adaptive behavior to Total Stress scores. These low associations contradict findings from previous research. When other variables are not accounted for, both of these variables typically demonstrate moderate negative associations with parenting stress such that lower child adaptive functioning and household income relate to greater parenting stress. Importantly there were only 4 families who met the DHHS classification of low-income; therefore, it is likely that many of the parents in this study's sample were not unduly influenced by financial stressors or greatly inhibited in obtaining supportive resources by their finances.

Paternal stress, specifically Parental Distress, was moderately associated to child chronological age  $r(37) = -.32, p = .05$ , and the number of children in the home with full mutation FXS,  $r(37) = .30, p = .07$ . Higher father Parental Distress scores were associated with lower child chronological age and greater number of children in the home with the full mutation. In contrast, maternal stress demonstrated a low-moderate association with the number of children in the home (including both those with and without FXS), associations

with Total stress, DC, PD and P-CDI scores  $r(37) \leq -.37 \geq -.21, p \leq .21 \geq .02$ . A relatively strong association between maternal stress and current mental health status (i.e. meeting criteria for any current mood or anxiety disorder or not) was found, Parental Distress  $r(37) = .42, p < .01$ . Interestingly, mothers with fewer children in the home reported more maternal stress, particularly parent-child domain stress. This finding is likely due, in part, to the target child frequently being the youngest child in the home. In many instances the target child was the first child in the household to be diagnosed with FXS. Not surprisingly, mothers with a current mental health diagnosis reported higher parenting stress, particularly higher parent domain stress. Mothers' lifetime mental health diagnosis was not found to be associated to her parenting stress.

Correlation matrices revealed one promising area for future research. There was a striking difference in the relationship between child problem behavior and maternal stress compared to paternal stress. For mothers, child problem behavior was significantly and strongly related to overall parenting stress [ $r(37) = .65, p < .001$ ] as well as child domain stress [ $r(37) = .62, p < .001$ ], parent domain stress [ $r(37) = .46, p < .01$ ], and parent-child domain stress [ $r(37) = .65, p < .001$ ]. For fathers, however, child problem behavior was only significantly associated to parent-child domain stress and that association was to a moderate degree [ $r(37) = .35, p = .03$ ]. In order to test whether differences in the association of parenting stress to child problem behavior across parent gender are relevant, a small HLM including PSI total stress as the outcome variable was computed post hoc. In this small HLM, parent gender, child problem behavior, and parent gender by child problem behavior interaction were included as independent variables. Based on this small HLM, model slopes for mothers and fathers were estimated and then the difference between these slopes was



tested for significance. Results from these analyses revealed a significant interaction of parent gender by child problem behavior on overall parenting stress,  $F(1, 36.00) = 4.22, p < .05$ . With lower levels of child problem behavior mothers reported lower levels of parenting stress than fathers. However, as child problem behavior increased mothers' parenting stress increased at a higher rate than fathers' such that at higher levels of child problem behavior mothers reported higher parenting stress than fathers (see Figure 1). Figure 1 illustrates that fathers' overall parenting stress increases by approximately half a point for every point of increase in child problem behavior. In contrast, mothers' overall parenting stress increases at a rate of approximately one and a half points for every one point increase in child problem behavior. Slope difference was significant at  $t(36) = -2.05, p < .05$ . As mothers appeared to demonstrate a heightened sensitivity to child problem behavior in comparison to fathers, the association of overall maternal stress to child problem behavior was examined to inform parent intervention research. Results of this correlation revealed that both child internalizing and externalizing behavior significantly contributed to mothers' reported Total Stress scores,  $r(37) = .56, p < .001$  and  $r(37) = .46, p < .01$  respectively.

## CHAPTER V

### Discussion

The present study addressed the gap in current literature by making a direct comparison of maternal and paternal stress in families with children with fragile X syndrome. The study improved upon previous research by including a larger sample of mother-father dyads, restricting the age range for the target child, and examining maternal and paternal stress outcomes within the same statistical models. The aim of this study was threefold: (1) describe similarities and differences in mothers' compared to fathers' parenting stress, (2) describe mothers' and fathers' perceptions of social support, and (3) investigate the relationship of parenting stress to social support examining across parent gender.

#### *Maternal Versus Paternal Stress*

Mothers and fathers from this study reported parenting stress levels commensurate with those found in the fragile X literature (e.g., Bailey et al., 2008; Johnston et al., 2003), with approximately 40% of mothers and fathers obtaining Total Stress scores in the high range (i.e.,  $\geq 85^{\text{th}}$  percentile). This large proportion of parents endorsing heightened parenting stress indicates that many families with children with fragile X syndrome would benefit from external resources or intervention (Abidin, 1995). When subscales indicating the types of stress experienced were examined, substantially more parents endorsed clinically significant stress related to the child or parent-child interactions (approximately 34%) than stress related

to their own functioning (approximately 14%). This finding is not surprising given consistent documentation of the strong direct effect of child problem behavior on parenting stress.

A primary finding of this study is the importance of moving beyond typical examinations of group means when making comparisons of mothers' and fathers' parenting stress. In the present study, similar to previous research, no significant group mean differences were found in mothers' versus fathers' reported parenting stress. This finding was true not only for overall parenting stress, but also its three domains (i.e., child, parent, and parent-child). In addition, no differences were found in the proportion of mothers versus fathers with parenting stress scores above clinical cut-offs. Yet upon further examination of data (i.e., investigating mothers' versus fathers' parenting stress through their inclusion in the same statistical models and at the dyadic level), original findings suggesting similar levels of maternal and paternal stress proved misleading. Not only were differences in mothers' and fathers' parenting stress found, but these differences lend support for a vulnerability to heightened parenting stress related to mothers' prenatation status.

First, a trend towards higher maternal parent domain stress versus paternal parent domain stress was found when comparing group mean values. Second, this trend was further substantiated when mothers and fathers were included in the same statistical models. Specifically, when salient variables were held constant, a significant main effect for parent gender on parent domain stress was found such that mothers reported significantly higher parent domain stress than did fathers. Therefore, mothers reported greater difficulty than fathers with aspects of parenting stress that are associated with problems in their own functioning. Finally, exploratory analyses revealed a significant association between mothers' current mental health status and her reported parent domain stress. Mothers with a

current diagnosis of an anxiety or affective disorder reported greater stress. Taken together, it is plausible that mothers' premutation status had a differential impact on their parenting stress response compared to fathers. In particular, the expressed difference in mothers' versus fathers' parent domain stress was likely influenced by mothers' increased risk for psychological distress and disorder.

In addition to revealed differences in parent domain stress, mothers and fathers displayed low to moderate inter-parent agreement with respect to parent domain stress and parent-child domain stress, and a lack of inter-parent agreement with respect to child domain stress and overall parenting stress. Contrary to the similarities displayed by investigations of parent gender group means, intraclass correlations revealed distinct differences in mothers' and fathers' parenting stress within individual households. In order to assist in the explanation of this difference, exploratory analyses were completed. Thirteen variables were examined for their association to parenting stress (i.e., child gender, child chronological age, child medication use on or off, child problem behavior, child adaptive functioning, family gross household income, low income versus middle-upper income, number of children in household, number of children in household with the full mutation of FXS, time in months since target child's FXS diagnosis, mother current diagnosis of anxiety or affective disorder versus no diagnosis, mother lifetime diagnosis of anxiety or affective disorder versus no diagnoses, and mother medication use on or off). Out of the thirteen variables only one variable, child problem behavior, demonstrated a striking difference in its relationship to maternal versus paternal stress. For mothers, child problem behavior had a strong positive association to overall parenting stress as well as its three domains. For fathers, child problem behavior demonstrated a positive, moderate association only to parent-child domain stress.

Thus, the magnitude and extent of mothers' and fathers' stress response to child problem behavior appeared to be different, with mothers demonstrating a pervasive response across parenting stress domains and fathers demonstrating a specific response related to stress from father-child interactions.

In order to better inform future research, post hoc analyses were completed to help confirm the relevance of apparent differences in mothers' and fathers' stress reactions to similar levels of child problem behavior. Results from these post hoc analyses supported differences revealing that as child problem behavior increases mothers' reported parenting stress increased at a higher rate than fathers' (see Figure 1). There are two plausible explanations for this difference. One, mothers were the sole reporters of child problem behavior. Therefore, mothers' perceptions of problem behavior may have been influenced by other factors which then drive the parent gender difference found in stress reactions. Two, there is a true difference in the stress response to problem behavior in mothers compared to fathers of children with fragile X syndrome. Currently, there is some support for both hypotheses. For example, one study implicated marital satisfaction as an important factor in mothers' reports of problem behavior in their child with a disability (Cuskelly & Dadds, 1992). Mothers tended to report greater child problem behavior as their marital satisfaction decreased. Similarly, in the present study moderate to strong correlations between mothers' ratings of problem behavior and social support were found such that higher ratings of problem behavior were associated with lower perceived spousal and family support. Yet, it appears unlikely that mothers being the sole reporters fully accounts for the extent of discrepancies in maternal versus paternal stress reactions found in the present study. In fact, there is reasonable evidence to support a true difference in stress response to child problem

behavior based on parent gender. First, as noted earlier, much research demonstrates strong inter-parent agreement when measuring child problem behavior using the CBCL, suggesting mothers and fathers perceptions should be similar. Walker and Bracken (1996) offered an explanation for the CBCL's ability to elicit consistently high and significant parental agreement. They suggested items on the CBCL represented easily agreed upon extreme behaviors making it a reliable instrument for discrimination between severe and average behavioral-emotional functioning. Second, recent family disability literature suggests mothers' and fathers' react differently to their child's problem behavior (Baker et al., 2005; Gadeyne, Ghesquière, & Onghena, 2004; Hastings et al., 2005; Kersh et al., 2006; Ornstein Davis & Carter, 2008). Gadeyne et al. (2004) found that children's externalizing behaviors and attention problems was predictive of high levels of control in mothers, while in fathers it was predictive of low levels of support. In relation, Kersh et al. (2006) found that when other salient variables were held constant, greater child problem behavior was significantly strongly related to maternal stress ( $r = .56$ ), with child problem behavior reported as the highest contributor to mothers' stress. Yet for fathers, greater child problem behavior had only a low-moderate correlation to paternal stress ( $r = .25$ ), with marital quality being the highest contributing variable to fathers' stress. Furthermore, the magnitude of influence of child problem behavior on maternal versus paternal stress differed even when a third party, teachers, provided the problem behavior ratings. Third, in the present study, a lack of association was found between mothers' and fathers' child-related stress which indicates, despite living in the same household, mothers' experienced different levels of stress related to child challenging behaviors than did fathers.

To the author's knowledge no other studies in fragile X literature have delineated findings with respect to inter-parent agreement on parenting stress measures or made comparisons by including mothers and fathers in the same statistical models. Thus, the present study makes particularly important contributions to furthering our understanding of parenting stress in these families. As well, it underscores the importance of gathering data on both parents' functioning when intervening to support optimal family functioning and child development.

#### *Maternal Versus Paternal Perceptions of Social Support*

Consistent with previous research mothers and fathers reported similar levels of perceived social support across various social support factors including spousal support, family support, and social network support (Hall et al., 2007; Keller & Honig, 2004; Oelofsen & Richardson, 2006). In addition, mothers and fathers reported a roughly equivalent number of social supports available to them and rated the usefulness of those supports similarly. Their perceptions of support proved to be analogous with those endorsed by parents of children with differing developmental disabilities (e.g., Keller & Honig, 2004; Oelofsen & Richardson, 2006). Yet in comparing parents from this study to their peers on reported levels of social network support, parents from the present sample endorsed markedly greater satisfaction with their supports than other mothers and fathers of children with FXS (McCarthy et al., 2006). This discrepancy in overall satisfaction with supports was found despite similar numbers of supports reported. For example, according to McCarthy et al.'s study, mothers mean Total Support score was 28.18 ( $SD = 10.60$ ) and fathers mean Total Support score was 26.32 ( $SD = 9.79$ ), while this study reported mean Total Support scores of 42.32 ( $SD = 11.35$ ) and 43.45 ( $SD = 13.77$ ) respectively for mothers and fathers.

Although it is unclear why there is such a substantial discrepancy between the present sample and that of McCarthy et al.'s, it is reasonable to believe that these differences are a function of a substantially smaller sample size ( $n= 27$  mother-father dyads), broader target child age range (range= 4-18 years), and greater proportion of target children in the at-risk or clinically significant range for behavioral problems compared to the present study.

Similar to findings for parenting stress, parent group mean values on measures of social support were found to provide a misleading representation of mothers' versus fathers' experiences. With the exception of spousal support, mothers' and fathers' ratings on support measures (i.e., family support and social network support) demonstrated low to moderate inter-parent agreement. The lack of parental agreement regarding perceived social support again emphasizes the importance of moving beyond common analyses of parent gender group means when aiming to describe similarities and differences in mother and father outcomes. Given research delineating the greater propensity of mothers with the premutation to have significant difficulty with social anxiety in comparison to non-FXS controls (Franke et al., 1996; Franke et al., 1998), one might have expected differences in their perceptions of social supports. However, correlations between mothers' mental health status and perceptions of family and social network support do not support this understanding. Instead, a review of preliminary research may indicate important differences in variables which contribute to maternal perceptions versus paternal perceptions of social support. For example, in the present study fathers endorsed a substantially greater range and variation in perceived family support, particularly in regard to family cohesion. Keller and Honig (2004) found that fathers' acceptance of their children's physical, cognitive, and emotional characteristics had a direct effect on their perception of family cohesion, such that the greater acceptance of the



child was related to higher perceptions of family cohesion. In contrast, for mothers increased care demands of the child and lower SES of the family were directly related to a lower perception of family cohesion.

### *Parenting Stress and Social Support across Parent Gender*

Hierarchical linear modeling assisted the author in investigating whether revealed household differences in perceived social support would lead to differences in the relationship of social support to parenting stress in mothers versus fathers. Essentially, the relationships of two social support factors (i.e., family support and social network support) to parenting stress were examined across parent gender. It was the author's intent to initiate research delineating differences in contributions of variables to maternal versus paternal stress. No difference in the relationship of social support to parenting stress was found across parent gender. Therefore, neither contributions of family support or social network support further assisted in the explanation of discrepancies found in mothers' and fathers' reported parenting stress.

Consistent with previous research, increases in family support were found to be related to decreases in maternal and paternal stress (Kersh et al., 2006; Hassall et al., 2005; Plant & Sanders, 2007; White & Hastings, 2004). This proved true for overall parenting stress and its three domains, with the relationship of family support to parent-child related stress nearing significance. However, contrary to findings regarding family support, no significant relationship between social network support and parenting stress was found. Wheeler et al. (2007) as well as McCarthy et al. (2006) obtained similar findings in parents of children with FXS, with social network support as measured by the FSS making no significant contribution to the prediction of maternal or paternal stress. Thus, support from

one's family appears to be a more salient variable in reducing parents' stress than support from the broader social network. Results from Plant and Sanders (2007) study further substantiate this hypothesis using a sample of families with children with various developmental disabilities. Their measure of partner/family support demonstrated a significant moderate relationship with parenting stress, while their measures of friend support and external/professional support demonstrated near zero correlations.

### *Limitations*

The present study had several sample limitations. First, as with many research studies of families of children with FXS, this sample was non-randomly selected and relied on families to volunteer their participation. Therefore it is plausible that there are differences between those families that volunteered and those who did not, thus impacting the generalizability of the study's findings. Second, the sample size was small which limited the power to detect effects, the number of variables that could be included in analyses and the type analyses that could be conducted. It is important to note, however, that this study's sample size included a larger number of mother-father dyads than previous research in fragile X literature and had a more tightly controlled sample (e.g., age range of target child was more restrictive and only cohabiting parents were included). The tightly controlled nature of the sample likely helped to offset the reduction in power by reducing error variance. Despite the good faith effort of the author, a larger sample size would have provided the opportunity to complete more extensive and thorough investigation of variables' interaction with parent gender on parenting stress. Third, no reliable data was collected in regards to fathers' status. Therefore it is unclear if the effect of biological versus step-father status had an effect on paternal stress. It seems reasonable that biological fathers would have been exposed to the

stressors of raising a child with a disability for a longer period of time and could have different interactions and qualities of attachments than step-fathers. However it is important to note that in a study conducted by Ricci and Hodapp (2003) no group differences were found regarding paternal stress when comparing in-home biological fathers to divorced biological fathers and in-home step-fathers of children with intellectual disabilities. Given the aforementioned study results and qualitative data from the current study (based on verbal report) suggesting that the vast majority of fathers were the biological father, it seems unlikely that there was an important influence of father status on this study's findings.

While this study compared maternal and paternal stress in families with children with fragile X syndrome, it lacks the valuable context that inclusion of comparison groups consisting of mothers and fathers of typically developing children and mothers and fathers of children with other developmental disabilities could have provided. It is possible with the addition of comparison groups the author would have interpreted the present study's findings differently. For example, if mothers of children with other developmental disabilities are shown to consistently report greater parent domain stress than fathers and demonstrate the same magnitude of relationship between their mental health status and experienced stress, than results from the present study may not suggest an influence of mothers' premutation status on their parenting stress. Thus, it will be important to follow up this study with one that includes relevant comparison groups.

Another limitation of this study was that mothers were the sole reporters of child problem behavior. Whereas past studies have been criticized for using only mothers' report of child problem behavior, results from several studies suggest strong similarity of across-parent ratings of child problem behavior (e.g., McCarthy et al., 2006), particularly in regards

to externalizing behavior which is the predominant reason why measures of child problem behavior demonstrate a relationship with parenting stress. For example, a review of the CBCL citing more than 250 samples in 119 studies conducted by McConaughy and Howell (1987) found on average statistically significant ( $p < .001$ ) correlations of .60 between similar informants (e.g., pairs of parents), with many studies reporting correlations as high as .70 to .80. As well, inter-informant agreement was significantly higher for children under age 11-year-olds compared to those for adolescents (12-19 years of age). As well, Hastings et al. (2005) found no differences in the predictions of contributions of variables on parenting stress when parents' own ratings of their children's problem behavior were replaced with that of their spouse/partner's ratings of their children's problem behavior. Taken together, it seems reasonable that a mother's report of child problem behavior captures the father's predominant perception of the child's functioning. Therefore it is feasible that only having mothers' ratings of child problem behavior had a minimal impact on the results of the present study.

Finally although precaution was taken in an attempt to ensure that fathers and mothers did not consult one another when filling out parent measures, there is no assurance that forms were filled out separately as they were mailed out prior to the assessment visit. Despite the lack of assurance, assessor observation and data suggests that mothers and fathers filled the forms out separately. First, mothers' and fathers' forms were provided in separate envelopes to encourage independent completion. Second, mothers and fathers were not routinely informed that any of the forms they had to complete were shared in common. Third, the majority of dates of completion on the forms for mother-father dyads were discrepant by a few days. Finally, the lack of inter-parent agreement on parenting stress and

social support measures strongly supports the independent completion of parent measures by mothers and fathers.

### *Summary and Implications for Practice and Research*

The present study found some indication of mothers' biological susceptibility to psychological distress and disorder on their parenting stress. Namely, mothers' stress related to their own parent functioning was shown to be higher than fathers'. In addition, maternal parent domain stress was strongly related to their current mental health status (meeting criteria for a current mood and/or anxiety disorder or not). As reported elsewhere, mothers and fathers in the present study as groups reported similar levels of parenting stress and perceived social support. However despite similarities in parent gender group means on measures of parenting stress and social support, mothers and fathers often reported having different experiences when examining at the dyadic level. These findings demonstrated the benefit of incorporating analyses beyond typical group level examinations. As well, they require professionals to avoid extrapolating mothers' experiences to fathers' by underscoring the need to assess and address both parents' needs when working to promote positive parent adaptation.

Although reasons for parent gender differences in parenting stress found cannot be decisively given, the contribution of mothers' mental health on their stress response and the potential differential impact of child problem behavior based on parent gender seem to be important areas for future research. If mothers' and fathers' stress reactions to similar levels of child problem behavior truly differ, this difference might help to explain their lack of inter-parent agreement on parenting stress and will provide important information for the development of effective parent interventions. For example if mothers have a heightened

sensitivity to child problem behavior compared to fathers, targeting child problem behavior may be a primary avenue for reducing mothers' stress but not fathers. Results from this study indicate a need for interventions to address both child internalizing behavior as well as externalizing behavior in order to most effectively reduce maternal stress. In particular, interventions which increase positive emotionality and social skill development, while decreasing aggressive and acting out behaviors would be beneficial. One evidence-based intervention that could address all these areas is parent-child interaction therapy (Herschell, Calzada, Eyberg, & McNeil, 2002).

Overall, results demonstrated that parents who perceived more family support experienced lower parenting stress, while support from the broader social support network was not found to be a salient contributor to parents' stress. The importance of informal supports over formal supports has been consistently shown across studies (e.g., Saloviita et al., 2003; White & Hastings, 2004) and suggests the need for an emphasis on enhancing family relations and cohesion when intervention is needed. Parent education programs may help to reduce parents' stress. First, parents' stress can be reduced through provision of effective parenting strategies to manage challenging behavior and the additional caretaking needs. Second, stress reduction can be promoted as parent education encourages improved communication and continuity of parenting behavior between parents.

Finally, given research indicating that fathers' are at greater risk for problems related to acceptance of and attachment with their child with a disability (e.g., Bristol et al., 1988; Keller & Honig, 2004), investigations of the influence of father involvement on maternal and paternal stress should also be added to future investigations. It is likely that in homes where fathers experience more disruption in these areas their parental involvement is diminished.

Lessened paternal involvement would leave a greater burden of care and behavior management responsibility to mothers. Thus, fathers who experience more difficulty in the areas of acceptance and attachment are likely to report lower parenting stress than their maternal counterparts because of a disengagement from interactions with their disabled child.

### *Conclusion*

Collectively these findings demonstrate the variation in parenting stress and perceived social support across households as well as across parent gender. It is the author's hope that future research will see the benefit of including fathers in studies and expand upon present findings by furthering the delineation of similarities and differences in maternal versus paternal stress. Exploring these areas will not only contribute to the development of effective parent interventions but may also help to reduce negative parenting behaviors that only further exacerbate undesirable child outcomes.

Table 1

*Intercorrelations between Maternal Independent and Control Variables*

Variables	1.	2.	3.	4.	5.	6.	7.	8.	9.
Mothers ( <i>n</i> = 38)									
1. Child gender	1	-.13	-.03	.03	.21	.51**	.00	-.04	-.22
2. Child age		1	-.03	-.18	-.41*	-.51**	-.04	-.06	.45**
3. Problem behavior			1	.11	-.12	-.25	-.37*	-.54**	-.15
4. Low income or not				1	.40*	.22	-.09	.06	-.06
5. # of FXS kids					1	.41*	-.04	-.07	-.02
6. Adaptive behavior						1	.03	.10	-.35*
7. Spousal support							1	.60**	.11
8. Family support								1	.18
9. Social network support									1

\**p* < .05. \*\**p* < .01.



Table 2

*Intercorrelations between Paternal Independent and Control Variables*

Variables	1.	2.	3.	4.	5.	6.	7.	8.	9.
Fathers ( $n = 38$ )									
1. Child gender	1	-.13	-.03	.03	.21	.51**	-.02	.06	.06
2. Child age		1	-.03	-.18	-.41*	-.51**	.11	.11	.27
3. Problem behavior			1	.11	-.12	-.25	-.39*	-.03	-.43**
4. Low income or not				1	.40*	.22	-.24	.05	-.07
5. # of FXS children					1	.41*	-.18	-.13	.07
6. Adaptive behavior						1	-.08	.05	.20
7. Spousal support							1	.60**	.33*
8. Family support								1	.41*
9. Social network support									1

\* $p < .05$ . \*\* $p < .01$ .

Table 3

*Descriptive Statistics of Mother and Father Dependent and Independent Variables*

	Mothers ( <i>n</i> =38)	Fathers ( <i>n</i> =38)
	<i>M</i> (SD)	<i>M</i> (SD)
PSI: Total Stress	83.97 (18.83)	81.76 (17.93)
Difficult Child	32.50 (8.16)	32.03 (8.06)
Parental Distress	27.37 (8.41)	24.68 (7.61)
Parent-Child Dysfunctional Interaction	24.11 (5.24)	25.05 (5.09)
PAIR: Emotional Intimacy Scale	21.61 (5.45)	23.12 (5.40)
FES: Family Relations Index	68.87 (17.38)	60.92 (28.75)
Cohesion	59.50 (6.36)	55.37 (12.82)
Expressiveness	56.84 (10.77)	52.76 (12.98)
Conflict	47.47 (8.90)	47.21 (11.62)
FSS: Total Support	42.32 (11.35)	43.45 (13.77)

Table 4

*Descriptive Statistics of Target Child Adaptive and Problem behavior*

	<i>M</i>	<i>SD</i>
	Target Child ( <i>n</i> = 38)	
CBCL: Total Problems	57.47	8.02
Internalizing	56.03	8.61
Externalizing	54.29	9.40
Vineland: Adaptive Behavior Composite	56.34	16.01
Communication domain	62.45	18.27
Daily Living domain	55.05	18.20
Socialization domain	67.71	13.45
Motor Skills domain	60.84	17.42

Table 5

*Percentage of Parents Accessing Types of Social Support*

Types of Social Support	% Mothers ( <i>n</i> = 38)	% Fathers ( <i>n</i> = 38)
Spouse/ partner	100	100
Family/ child physician	97.37	94.74
Spouse/ partner's relatives	94.74	81.58
Relatives	92.11	68.42
School/ daycare center	92.11	92.11
Professional helpers	92.11	97.37
Friends	89.47	76.32
Parents	86.84	78.95
Spouse/ partner's parents	86.84	92.11
Spouse/ partner's friends	81.58	86.84
Own children	76.32	73.68
Other parents	65.79	60.53
Early childhood intervention program	63.16	89.47
Church members/ minister	60.53	57.89
Professional agencies	57.89	57.89
Co-workers	47.37	47.37
Parent groups	36.84	39.47
Social groups/ clubs	36.84	47.37

Table 6

*Results of Hierarchical Linear Models for Primary Analyses*

Parameter	PSI: PD	PSI: P-CDI	PSI: DC	PSI: Total
	Estimate (SE)			
Intercept	27.49 (1.42)	24.49 (.91)	34.13 (1.39)	86.32 (3.01)
Parent gender	-4.06 (1.27)**	.38 (.90)	-1.56 (1.48)	-5.44 (2.74) <sup>∞</sup>
Family support	-.18 (.06)**	-.09 (.04) <sup>∞</sup>	-.18 (.07)*	-.49 (.14)**
Social network support	-.06 (.10)	-.02 (.07)	-.10 (.10)	-.20 (.21)
Low income/ not	-.49 (3.23)	-1.84 (2.00)	1.69 (2.95)	-.32 (6.79)
# of FXS children in home	1.44 (2.22)	.35 (1.38)	-2.72 (2.04)	-1.13 (4.67)
Child age	-.05 (.05)	.01 (.03)	-.07 (.05)	-.11 (.11)
Problem behavior	.19 (.13)	.26 (.08)**	.26 (.12)*	.67 (.27)*
Family support by parent gender	.01 (.07)	.02 (.05)	.06 (.08)	.12 (.16)
Social network support by parent gender	.06 (.12)	-.03 (.08)	.00 (.13)	.04 (.26)

PSI: PD, Parental Distress; P-CDI, Parent-Child Dysfunctional Interaction; DC, Difficult Child

<sup>∞</sup>p < .056. \*p < .05. \*\*p < .01.

Table 7

*Mother Exploratory Intercorrelations between Predictor and Parenting Stress Variables*

	PSI: PD	PSI: P-CDI	PSI: DC	PSI: Total
Child gender	.21	-.06	.17	.15
Child age	-.13	.01	-.21	-.14
Child meds (on/off)	.09	.21	.26	.21
Problem behavior	.46**	.65**	.62**	.65**
Adaptive behavior	-.02	-.23	.22	.02
Family gross household income	-.02	.02	.24	.10
Low-income/ not	-.02	-.19	.02	-.05
# of children in home	-.24	-.37*	-.21	-.30
# of FXS children	.22	-.11	.06	.09
Time since FXS diagnosis	-.03	-.03	-.21	-.11
Mother current mental health diagnosis/ not	.42**	.23	.05	.27
Mother lifetime mental health diagnosis/ not	.29	.28	.17	.28
Mother meds (on/off)	.15	.02	.04	.09

PSI: PD, Parental Distress; P-CDI, Parent-Child Dysfunctional Interaction; DC, Difficult Child

\* $p < .05$ . \*\* $p < .01$ .

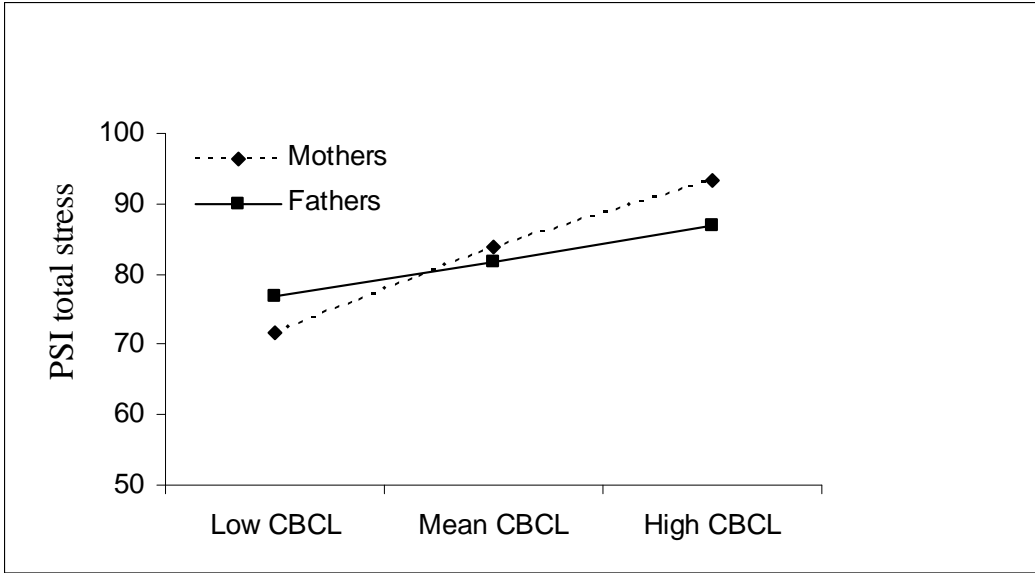
Table 8

*Father Exploratory Intercorrelations between Predictor and Parenting Stress Variables*

	PSI: PD	PSI: P-CDI	PSI: DC	PSI: Total
Child gender	-.19	-.26	-.16	-.23
Child age	-.32	-.04	-.17	-.22
Child meds (on/off)	-.02	.08	.08	.05
Problem behavior	.15	.35*	.26	.28
Adaptive behavior	-.01	-.27	-.17	-.16
Family gross household income	-.10	-.14	-.07	-.12
Low-income/ not	.09	.05	.06	.08
# of children in home	.01	-.05	-.07	-.04
# of FXS children	.30	.08	-.14	.08
Time since FXS diagnosis	-.16	.01	-.23	-.17
Mother current mental health diagnosis/ not	.16	.04	.04	.10
Mother lifetime mental health diagnosis/ not	.18	.06	.04	.11
Mother meds (on/off)	.24	.08	.04	.14

PSI: PD, Parental Distress; P-CDI, Parent-Child Dysfunctional Interaction; DC, Difficult Child

\*p< .05.



*Figure 1.* Model slopes for mothers and fathers parenting stress by child problem behavior revealing an interaction of parent gender by child problem behavior on overall parenting stress.



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