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Maternal Parenting Stress In Autism, Autism Associated With Fragile X, and Fragile X Alone: An Examination of Associated Child and Maternal Factors In Three High-Risk Groups

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MATERNAL PARENTING STRESS IN AUTISM, AUTISM ASSOCIATED WITH FRAGILE X, AND FRAGILE X ALONE: AN EXAMINATION OF ASSOCIATED CHILD AND MATERNAL FACTORS IN THREE HIGH-RISK GROUPS

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DEDICATION

To my son whose life has filled me with a passion to pursue the path on which I find myself today. And to my daughter whose humor, wonder, and strength inspire me each and every day.

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ABSTRACT

The current study examined the association between specific child and maternal factors and parenting stress in three high-risk groups of mothers - mothers of boys diagnosed with idiopathic autism (IA), mothers of boys diagnosed with autism spectrum disorder (ASD) associated with fragile X syndrome (AFXS), and mothers of boys diagnosed with fragile X syndrome (FXS) alone. These three groups of mothers are thought to share some degree of genetic vulnerability to stress, as well as exposure to varying levels of challenging child behavioral characteristics. Theories of parenting stress incorporate multiple components, including parent, child, and parent-child interaction factors. The current study examined differences in maternal parenting stress across groups of high-risk mothers, as well as the relationship between child problem behaviors and the various dimensions of parenting stress. Additionally, the current study examined the relationship between maternal characteristics of the broader autism phenotype (BAP) and parenting stress in mothers of children with IA. The differential impact of maternal BAP across dimensions of parenting stress was explored.

The primary sample of participants for the present study came from an extant dataset including 48 mothers of boys with IA, 20 mothers of boys with AFXS, and 56 mothers of boys with FXS alone. A secondary sample of 20 biological mothers of male children with IA was recruited to address secondary questions related to the maternal BAP – parenting stress relationship. Results indicated a significant difference in child-

related parenting stress among groups of mothers from the primary sample. Regression analysis indicated significant main effects for general child behavior problems and maternal IQ, but not for ASD symptomatology for the primary sample. Results also indicated a significant interaction between maternal group and general child behavior problems. Exploratory secondary analyses indicated that scores from one subscale of a BAP measure significantly predicted both child- and parent-related stress scores. Surprisingly, general child behavior problems did not make a significant contribution to the prediction of parenting stress scores for mothers from this secondary sample. Limitations of the current study and potential implications for practice are discussed.

TABLE OF CONTENTS

DEDICATION iii

ACKNOWLEDGEMENTS iv

ABSTRACT v

LIST OF TABLES viii

LIST OF FIGURES ix

LIST OF ABBREVIATIONS x

CHAPTER 1: INTRODUCTION 1

CHAPTER 2: LITERATURE REVIEW 6

CHAPTER 3: METHOD 27

CHAPTER 4: RESULTS 46

CHAPTER 5: DISCUSSION 71

REFERENCES 88

LIST OF TABLES

Table 2.1 Summary of Articles Reviewed Employing PSI 25

Table 3.1 Child and Maternal Descriptives for the Primary Sample.....43

Table 3.2 Measures Completed by Participant Group 45

Table 4.1 Pearson Correlation Matrix for Variables in the Primary Analysis.....56

Table 4.2 Hypothesis 1 Multivariate Results…….58

Table 4.3 Hypothesis 2 and 3 Multivariate Results……...…60

Table 4.4 Pearson Correlation Matrix for Variables in the Secondary Analysis.....64

Table 4.5 Child and Maternal Descriptives for the Secondary Sample.....66

Table 4.6 Hypothesis 4 Multivariate Results.....68

LIST OF FIGURES

Figure 4.1 Interaction of Group and CBCL 63

List of Abbreviations

ABC……Autism Behavior Checklist

AFXS…….Fragile X Syndrome and Autism

ABC……...Adaptive Behavior Composite

ASD……Autism Spectrum Disorder

BAP-Q…….Broad Autism Phenotype Questionnaire

CARS……..Child Autism Rating Scale

CBCL……Child Behavior Checklist

FXS……Fragile X Syndrome

GARS……Gilliam Autism Rating Scale

IA……..Idiopathic Autism

MS…….Maternal Stress

PSI-SF …….Parenting Stress Index – Short Form

PSPQ-S…….The Personality Styles and Preferences Questionnaire

SCQ……Social Communication Questionnaire

CHAPTER 1

INTRODUCTION

Studies measuring parenting stress, a construct defined in the literature as psychological distress arising from the parenting role (Abidin, 1995), have indicated significant elevations in stress related to the caregiving role for parents of children with developmental disabilities (Dumas, Wolf, Fisman, & Culligan, 1991; Sanders & Morgan, 1997). Mothers, who often serve as primary caregivers, seem particularly vulnerable to socioemotional difficulties and stress associated with adaptation to their child's disability (Eisenhower, Baker, & Blacher, 2005; Freeman, Perry, & Factor, 1991; Koegel et al., 1992; Milgram & Atzil, 1988; Sivberg 2002). Elevated parenting stress in mothers of children with developmental disabilities has been linked to a range of negative outcomes including increased maladaptive parenting behaviors, greater incidence of maternal psychopathology, failure to engage with services, and less benefit from intervention for children (Osborne et al., 2008; Robbins, Dunlap, & Plienis, 1991). These and other poor outcomes associated with high levels of parenting stress have compelled numerous efforts to explore factors contributing to mothers' experience of stress in adapting to a child's disability. As parenting stress is a complex construct thought to be impacted by a number of child, parent, and environmental characteristics; the degree to which specific factors influence stress related to the parenting role has been the focus of much research.

Comparative studies have provided consistent evidence linking type of child disability to the experience of maternal parenting stress, with some disorders emerging as having higher levels of associated stress than others (Abbeduto et al., 2004; Eisenhower et al., 2005). Mothers of children diagnosed with autism spectrum disorder (ASD) have often been found to report the highest levels of parenting stress when compared to mothers of children with other disabilities or illnesses (Abbeduto et al., 2004; Bouma & Schweitzer, 1990; Eisenhower et al., 2005; Griffith, Hastings, Nash, & Hill, 2010). Mothers of children with fragile X syndrome (FXS), a genetic condition with considerable behavioral overlap with ASD, have shown similar, though less dramatic elevations in parenting stress (Johnston et al., 2003).

Children with ASD and FXS both present with a range of social deficits and maladaptive behaviors thought to impact parental stress and well-being (Abbeduto et al., 2004). For mothers of children on the autism spectrum, there are considerable data to suggest that the severity of a child's autistic behavioral symptoms makes a significant contribution to maternal stress (e.g., Bebko, Konstantareas, & Springer, 1987; Hastings & Johnson, 2001; Tobing & Glenwick, 2003). A significant number of males with fragile X syndrome display behaviors resembling those of ASD, with approximately 25 % to 50% meeting criteria for a co-morbid diagnosis of autism spectrum disorder (Hall, Lightbody, & Reiss, 2008; Kaufman et al., 2004). The severity of autistic features in individuals with FXS has also been linked to increases in maternal parenting stress (Mankowski, 2007). In both of these populations, however, there is growing evidence that general problem behaviors (not specific to autism) may be more relevant predictors of mothers' stress (e.g., Davis & Carter, 2008; Hastings, 2003; Johnston et al., 2003; Lecavalier, Leone, &

Wiltz, 2006). As support for this relationship between general child problem behaviors and maternal parenting stress has accumulated, little is known still about how various dimensions of maternal stress (i.e., child-related, parent-related, or stress related to the parent-child interaction) may be differentially impacted by general child problem behaviors.

In addition to child factors that have been evaluated in relation to parenting stress in high risk groups of mothers, a number of maternal factors have also been considered. Among these, one factor that has recently been shown to have a strong positive association with parenting stress in mothers of children diagnosed with ASD is the presence of the broader autism phenotype (BAP) (Ingersoll & Hambrick, 2011). The BAP refers to a subclinical set of personality and cognitive characteristics found in relatives of individuals with ASD thought to serve as an index of genetic liability to autism (Piven, Palmer, Jacobi, Childress, & Arndt, 1997). In a large sample of parents of children with ASD (approximately 91% were mothers), Ingersoll and Hambrick (2011) found that parents who express a higher number of characteristics of the BAP and who have children with more severe symptoms of ASD are at increased risk of elevated parenting stress and depression. Further exploration of this relationship between maternal BAP, child behavior, and stress is needed to help inform the efforts of those working with families of children with ASD.

With the assumption that stress levels vary according to complex interactions between child behavioral characteristics and genetic vulnerabilities in mothers, the current study examined the impact of specific child and maternal factors on parenting stress in groups of high-risk mothers. Given current gaps in understanding regarding the

differential impact that child behavior problems may have on varying dimensions of parenting stress in these high-risk groups of mothers, this study also examined how challenging child behaviors may exert varying degrees of influence on the three factors typically accepted as comprising maternal parenting stress. This frequently under-utilized approach may have important implications for practice as the various types of parenting stress are likely associated not only with distinct patterns of contributing factors, but also differing outcomes. The relative impact of child behavioral characteristics on parent-related, child-related, and parent-child-interaction- related parenting stress was evaluated in three groups of mothers: 1) mothers of boys diagnosed with idiopathic autism (IA), 2) mothers of boys with autism associated with FXS (AFXS), and 3) mothers of boys with FXS alone. Few existing studies have looked at maternal stress across these groups of mothers of children with etiologically distinct but overlapping behavioral features. This methodology allowed for a unique examination of how specific child behavioral characteristics impact the excessive amounts of maternal parenting stress observed in mothers of children with ASD and FXS. In particular, inclusion of a group of mothers of children diagnosed with AFXS provided an opportunity to parse out the impact of ASD-related symptomatolgy and other child behavioral characteristics on stress levels in these three groups of mothers who are already likely predisposed to higher stress levels and greater psychopathology (by either having features of the broad autism phenotype or by being a carrier of FXS). In a subgroup of mothers of boys with IA only, the current study also examined the relationship between maternal BAP and the various dimensions of parenting stress. Potential implications of findings for developing and/or improving

screening and intervention efforts for those working with families of children with ASD and FXS are discussed.

CHAPTER 2

LITERATURE REVIEW

Defining Parenting Stress and Theoretical Framework

Abidin (1995) defined parenting stress as the parent's internal response to the relationship between the parent and the child. Deater-Deckard (2006) characterizes parenting stress simply as "the aversive psychological reaction to the demands of being a parent". Distinct from widely accepted conceptualizations of stress which focus on events as stressors, parenting stress is most often described as a reaction or outcome arising from a mismatch between parents' perceptions of parenting demands and what they perceive as available resources for dealing with these demands (Deater-Deckard & Scarr, 1996; Goldstein, 1995). While there is evidence in the literature of some overlap among various domains of stress experienced by individuals, many have set apart stress associated with the parenting role as a construct that is qualitatively distinct from stress emanating from other roles (e.g., event-related stressors such as work-related stress). Specifically, when compared to stress associated with more general life circumstances; parenting stress has been shown to exert a more direct impact on parenting behaviors and child adjustment (Creasy & Reese, 1996).

Theories of parenting stress have typically incorporated multiple components, including parent, child, parent-child interaction, and environmental factors (see e.g., Mash & Johnston, 1990). Abidin, author of the Parenting Stress Index (PSI; Abidin,

1995), a measure of parenting stress frequently employed in the literature, posits an ecological theory of parenting stress where the experience of stress is determined by the interplay of parent, child, and situational factors. Parental factors included in the model include depression, attachment, role restriction, competence, parental health, social support/isolation, and the spousal relationship. Child factors are defined as adaptability, acceptability, demandingness, mood, hyperactivity, and being reinforcing to the parent. In Abidin's theory, parenting stress is proposed to negatively affect parenting behaviors, which in turn affects child outcomes.

Abidin's conceptualization of parenting stress is in line with other well-established theories of family functioning such as Family Systems Theory (Bowen, 1978) and Belsky's (1984) process model of parenting, both of which characterize parental functioning as impacted by a bi-directional process of socialization. These theories emphasize the reciprocal interactions within family systems that make parenting behaviors and child behaviors and outcomes inextricably linked. Given the wide-spread application of Abidin's model of parenting stress and its consistency with other established models from the larger parenting literature, it was used as the conceptual framework for the current study. Because Abidin's three-factor theory emphasizes the importance of understanding the parent-related, child-related, and parent-child transactional components contributing to the broader construct of parenting stress, scores from each of the three corresponding subscales from the abbreviated version of the Parenting Stress Index – Short Form (PSI-SF) were considered in the current study.

Effects of Maternal Parenting Stress

Given the bi-directional nature of the parent-child relationship, it is not surprising that studies have shown maternal parenting stress to be associated with a host of negative outcomes for both mother and child. One consistent finding across clinical and non-clinical populations is a positive association between maternal parenting stress and maternal depression (Gelfand, Teti, & Fox, 1992; Milgrom & McCloud, 1996). Parenting stress and maternal depression represent distinct constructs which have some degree of overlap in terms of risk factor profiles and associated behavioral outcomes (Leigh & Milgrom, 2008). Although the direction of the relationship between parenting stress and depression remains debatable, the literature clearly points to an important link between the two. High levels of parenting stress, with and without associated depression, have also been found to impact a range of parenting behaviors which are, in turn, thought to influence child behaviors (Crnic, Gaze, & Hoffman, 2005; Deater- Deckard, 1998). Specifically, studies have shown a link between elevated parenting stress and low levels of warmth and parent-child reciprocity, along with elevated risk of abusive parenting behaviors (Rogers, 1993; Holden & Banez, 1996). Results of some investigations have indicated that elevations in specific dimensions of parenting stress may differentially impact parenting outcomes. For example, Holden and Banez (1996) found that aspects of parent-related stress actually moderated the relationship between child-related stress and abuse potential in parents.

Studies which have specifically focused on the impact of stress in mothers of children with ASD have revealed significant associations between high levels of parenting stress and poor outcomes (Osborne & Reed, 2009; Robbins et al., 1991).

Osborne and Reed (2009) found that both mother-child communication and maternal limit setting behaviors decreased as parenting stress increased in mothers of children with ASD. Recent evidence also suggests that high initial levels of maternal parenting stress can, over time, lead to a worsening of behavior problems in children with ASD (Lecavalier et al., 2006), and to reduced effectiveness of early intervention efforts (Osborne, McHugh, Saunders, & Reed, 2008).

Autism Overview

Autism spectrum disorder is characterized by impaired social communication and reciprocity, as well as restricted, repetitive, and/or stereotyped patterns of behavior, interests, and activities (APA, 2013). While all children who receive a diagnosis of ASD exhibit some degree of difficulty across each of the core domains outlined in the diagnostic criteria, expression of the disorder may vary widely among affected individuals. An estimated 75% of children with ASD have some degree of intellectual disability, with cognitive profiles that are frequently remarkable for unevenly developed abilities. The current data suggest that 1 in 88 children have ASD, with boys being affected by the disorder at higher rates than girls (CDC, 2012). While the high recurrence rate of 15-20% in first degree relatives of an individual with autism suggests a significant genetic basis for the disorder (Constantino, Zhang, Frazier, Abbacchi, & Law, 2010), current research points to a multi-factorial inheritance pattern in ASD involving a complex interplay of multiple genetic and environmental factors.

Parenting Stress in Mothers of Children with ASD

Mothers of children with ASD have reported higher levels of stress stemming from their role as parents than mothers of children with Down syndrome, fragile X syndrome, cystic fibrosis, and cerebral palsy, as well as children with undifferentiated developmental delays (e.g., Abbeduto et al., 2004; Bouma & Schweitzer, 1990; Eisenhower et al., 2005; Griffith et al., 2010). Although experiencing some degree of parenting stress is largely considered normative and even adaptive (Deater-Deckard & Scarr, 1996), the levels of parenting stress in mothers of children with ASD have often been reported as falling within the range of clinical significance (Davis & Carter, 2008; Noh, Dumas, Wolf, & Fisman, 1989; Tomanik et al., 2004). Given the heightened risk of excessive parenting stress in this population of mothers, a host of child and maternal variables that may influence stress levels have been examined in the literature (Bouma & Schweitzer, 1990; Gray & Holden, 1992; Sharpley, Bitsika & Efremidis, 1997).

Child Factors

Investigators have frequently reported on the link between child factors such as age, cognitive functioning, and adaptive functioning and maternal parenting stress over the last 20 years with varying results (Bebko et al., 1987; Bouma & Schweitzer, 1990; Konstantareas & Homatidis, 1989; Wolf, Noh, Fisman, & Speechley, 1989). However, the literature has become increasingly focused on the relationship between child behavioral characteristics and maternal stress in ASD. In particular, a great deal of attention has been given to how maternal parenting stress is impacted by the severity of

child autism symptoms, as well as by more general (not autism specific) child problem behaviors.

Correlational studies relying on parent-report measures of overall autism symptom severity such as the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1988), the Gilliam Autism Rating Scale (GARS; Gilliam, 1995), and the Autism Behavior Checklist (ABC; Krug, Arick, & Almond, 1979) have indicated a positive relationship between severity of autistic symptoms and maternal ratings of parenting stress (Bebko et al., 1987; Bravo, 2006; Freeman et al., 1991; Kasari & Sigman, 1997). Studies which have focused on specific symptoms in ASD have also suggested a strong positive relationship between parent and professional reports of communication difficulties and social deficits and maternal stress (Kasari & Sigman, 1997; Phetrasuwan & Miles, 2008; Tomanik et al., 2004). Some evidence for an association between severity of stereotyped and repetitive behaviors and maternal parenting stress has also been reported in the literature (Richardson, 2010; Stoddart, 2003). It is worth noting that studies linking autistic symptom severity to maternal stress have inconsistently considered the impact of symptoms across dimensions of parenting stress. Results from a small number of studies suggest that child-related parenting stress might be influenced by severity of symptoms to a greater degree than other parenting stress dimensions (Bravo, 2006; Kasari & Sigman, 1997). However, too few studies have assumed the type of approach needed to draw clear conclusions in this area.

Individuals with ASD may present with a number of challenging symptoms not directly tied to the core features of ASD, including motor deficiencies, hyperactivity, aggression, self-injurious behavior, anxiety disorders, depression, eating problems, and

erratic sleep patterns (Ozonoff & Rogers, 2003). Studies have shown that children with ASD as a whole suffer from emotional and behavioral problems at rates much higher than those of children from other clinical populations (Brereton, Tonge, & Einfeld, 2006; Kasari & Sigman, 1997). It is not surprising that these excessive problem behaviors might contribute to mothers' experience of stress in this population. A range of externalizing and regulatory behaviors in children diagnosed with ASD have been linked to elevations in maternal parenting stress including self-abuse and hyperirritability (Konstantareas & Homatidis, 1989; Tomanik et al., 2004), high activity level (Konstantareas & Papageorgiou, 2006), noncompliance (Tomanik et al., 2004), eating difficulties (Archer & Szatmari, 1991), and sleep disturbances (Hoffman, Sweeney, Lopez-Wagner, Hodge, Nam, & Botts, 2008). The nature and direction of the association between child internalizing behaviors and maternal parenting stress in ASD is less well-understood. However, there is emerging evidence of a link between the two which appears to be reciprocal in nature (Bauminger, Solomon, & Rogers, 2010). Studies have frequently examined the association between mothers' overall ratings of child problem behaviors (incorporating both externalizing and internalizing behaviors) and parenting stress in ASD. Studies which have taken this approach have also consistently indicated a positive association of general child maladaptive behaviors and maternal parenting stress (Hastings, 2003; Herring, Gray, Taffe, Tonge, Sweeney, & Einfeld, 2006). As with studies focused on the severity of autism symptoms, studies examining the relationship between more general problem behaviors and maternal parenting stress have also seldom considered the differential impact that general problem behaviors may have on the various dimensions of parenting stress.

Given mounting evidence for the impact that child behavior problems not directly related to the autism diagnosis can have on the experience of maternal stress, some researchers have sought to explicitly compare the influence of core autism features to general behavior problems on parenting stress in mothers of children with ASD. In one such study, Hastings et al. (2005) found that ratings of child behavior problems were more strongly associated with maternal parenting stress than severity of autism symptoms, child adaptive behaviors, partner anxiety, and partner depression. Studies by Konstantareas and Papageorgiou (2006) and Davis and Carter (2008) replicated and extended these findings. Konstantareas and Papageorgiou (2006) found that, more than any other factor, including severity of autism as measured by the CARS, maternal ratings of child activity level on a measure of temperament best predicted parenting stress, with higher levels of activity predicting greater stress. Similarly, Davis and Carter (2008) found that mothers' total stress scores on an abbreviated version of the PSI (PSI-Short Form; PSI-SF) were impacted more by behaviors not directly tied to the diagnosis than by any autism-specific deficits. Specifically, while deficits in social relatedness were associated with parenting stress for mothers and fathers, mothers' ratings of self-regulatory problems (e.g., feeding issues, sleep difficulties, and poor emotional regulation) were more predictive of overall parenting stress than impaired social or communication skills.

Using methods which allowed for the examination of maternal perceptions of several different dimensions of symptomatolgy in autism, and the association between these various dimensions with maternal socioemotional functioning, Ekas and Whitman (2010) also found that ratings of behavior problems were strongly associated with reports

of overall maternal parenting stress. Consistent with findings from Davis and Carter (2008) and Hastings and colleagues (2005), results of this study indicated that behavior problems unrelated to the autism diagnosis (e.g., hyperactivity, rapid mood swings, self-injury, non-compliance, and sleep problems) were the only unique predictors of maternal parenting stress.

In a study on which the current study proposes to build and expand, Mankowski (2007) examined the association between child autistic and/or general behavior problems and maternal mood, anxiety, and stress in mothers of children with FXS, IA, and AFXS. This is the only known existing study to have looked at maternal parenting stress across these three high-risk groups of mothers. Interestingly, though mothers of children with FXS alone reported significantly less stress than mothers of children with IA and mothers of children with AFXS in this study, no differences in ratings of maternal parenting stress were found between the IA and AFXS groups. Despite the differing levels of stress reported by the groups, results indicated that general child problem behaviors predicted overall maternal parenting stress across all groups, without any interaction of group and difficult child behaviors. While the impact on maternal parenting stress of both general problem behaviors and autistic behavior were considered in this study, analyses did not specifically assess which type of behavior served as the more salient predictor of stress. Also, only total scores of parenting stress were analyzed in this study, which precluded examination of how various dimensions of stress (i.e., child-related, parent-related, and parent-child interaction-related) may be differentially impacted by these child factors. A summary of the various dimensions of parenting stress evaluated in this and previously described studies in this section is provided in Table 2.1.

Maternal Factors

Though a number of maternal factors, including age, education, and income have been reported as impacting the degree to which mothers of children with ASD experience parenting stress (Bebko et al., 1987; Yau & Li-Tsang, 1999), the results of efforts to understand the impact of these sociodemographic variables have often returned unclear or even contradictory results. In recent years, increasing attention has been focused on trying to understand what other maternal characteristics may at least partially account for the excessive levels of parenting stress observed in this population. One potentially important factor identified recently in the literature is the existence of what has been referred to as the broader autism phenotype (BAP). The BAP describes the observed tendency for parents of children with ASD to exhibit subtle manifestations of core autism features, such as social aloofness, stereotypic behaviors, and pragmatic language difficulties, as well as higher rates of several psychiatric disorders, including mood and anxiety disorders (Delong & Dwyer, 1988; Piven et al., 1997; Piven, 2001). There is rapidly growing evidence for the existence of this broad autism phenotype, with some studies indicating that parental onset of the observed associated psychopathology often precedes the birth of the child with ASD (Micali et al., 2004; Smalley, McCracken, & Tanguay, 1995) supporting a genetic predisposition to these features.

In the only known study that has considered the potential impact of maternal BAP on the experience of parenting stress, Ingersoll and Hambrick (2011) found that parent BAP and child symptom severity were both positively correlated with parenting stress and depression. Specifically, in their sample of 149 parents (> 91 % mothers), parents with higher BAP scores were more likely to use maladaptive coping strategies, which

were in turn, associated with increased stress and depression. Higher BAP scores were also associated with less social support, which again appeared to partially mediate the relationship between parent BAP and stress and depression. Taken together, research on this broad autism phenotype suggests that an underlying genetic susceptibility linked to shared characteristics with the child with ASD may leave mothers of children with autism more vulnerable to psychological problems, and with fewer resources for effectively coping with the difficult child characteristics often associated with an ASD diagnosis.

Summary

Results of studies examining the influence of child behavioral characteristics on parenting stress in mothers of children with ASD have consistently indicated a strong association of both autism symptoms and more general behavioral problems with the experience of parenting stress. Recent investigations, however, have begun to suggest that the presence of behavior problems not linked to core autism symptomatology may in fact be the most salient predictor of maternal stress. Because existing studies have primarily considered the impact of these child behavioral characteristics on mothers' total stress scores on various parenting stress measures, little is known about how child behavior problems may impact the various dimensions of parenting stress. In addition to child behavioral characteristics, maternal characteristics which may help explain the high levels of maternal parenting stress in this population have been the focus of recent investigations. In particular, there is emerging evidence that maternal characteristics of the BAP may be linked to an underlying genetic vulnerability to stress and psychopathology in this group of mothers that may be exacerbated by the presence of child problem behaviors.

FXS Overview

With an estimated prevalence rate of 1 in 2500 to 1 in 4000 males, FXS is the leading known genetic cause of autism, as well as the leading known cause of inherited intellectual disability. FXS is a genetic condition resulting from the mutation of a single gene – the fragile X mental retardation gene (FMR1) on the X chromosome. In individuals with FXS, the number of trinucleotide repeats (CGG) in the FMR1 gene becomes unstable and expands (Hatton et al., 2002). While in unaffected individuals, this DNA segment is usually repeated from 5 to about 40 times, in individuals with FXS, this segment is repeated more than 200 times. Males and females with 55 to 200 repeats of the CGG segment are said to have a premutation of the *FMR1* gene.

FXS is associated with a range of behavioral and physical symptoms that vary according to gender. Specifically, because of the X-linked inheritance pattern in FXS, males are generally more severely affected by the disorder. The effect of FXS on females is highly variable with approximately 50% displaying some degree of cognitive impairment, and the remaining percentage presenting with few to no cognitive or behavioral sequelae (Bailey, Raspa, Olmsted, & Holiday, 2008). A majority of males with FXS are impacted cognitively by the disorder, with most testing in the mild to moderate range of cognitive impairment (Bailey, Hatton, & Skinner, 1998; Hatton et al., 2002). Challenging behaviors, such hand flapping, tactile defensiveness, avoidant eye contact, hyperactivity, social anxiety, tantrums, and perseveration are also frequently observed in affected males (Hatton et al., 2002). A significant number of males with FXS exhibit autistic-like behaviors with approximately 25-50% of individuals with FXS meeting DSM criteria for autistic disorder using DSM-IV criteria (Hall et al., 2008;

Kaufman et al., 2004). In boys with and without a co-morbid diagnosis of ASD, elevated autistic symptoms have been found to be associated with poorer developmental outcomes in this population (Hatton et al., 2006; Rogers et al., 2001).

Parenting Stress in Mothers of Children with FXS

As with mothers of children on the autism spectrum, numerous studies have indicated elevated levels of parenting stress in mothers of children with FXS (Johnston et al, 2003; McCarthy, Cuskelly, van Kraayenoord, & Cohen, 2005). Though not as extensive as the available research on maternal parenting stress and ASD, the existing literature on parenting stress and FXS clearly suggests elevated stress levels in these parents when compared to parents of typically developing children (Johnston et al., 2003; McCarthy et al., 2006; von Gontard et al., 2002; Wheeler et al., 2007) and to groups of parents raising children with other disabilities (Lewis et al., 2006; von Gontard et al., 2002). In fact, studies have found that as many as 30% of mothers of children with FXS report levels of parenting stress in the clinically significant range on the PSI (Bailey et al., 2008; Mankowksi, 2007).

Child Factors

Consistent with the ASD literature, studies which have examined parenting stress in mothers of children with FXS have suggested that child characteristics may have a direct influence on the experience of stress in this population (Wheeler et al., 2007). When compared to other child characteristics such as age and intelligence, Johnston and colleagues (2003) found general child behavior problems to have the strongest association with parenting stress in mothers of children with FXS. Similarly, Wheeler,

Skinner, and Bailey (2008) reported a strong correlation between overall parenting stress and reported general child problem behaviors in mothers with a child affected by FXS. Because of the significant behavioral overlap between FXS and ASD, some researchers have examined the influence of autistic behaviors on parenting stress in this population. Studies using general measures of autistic symptoms (i.e., the CARS; Mankowski, 2007) and more specific measures (i.e., the Repetitive Behavior Scale- Revised and the Sensory Experiences Questionnaire; Richardson, 2010) have suggested a predictive relationship between ASD symptoms and parenting stress in mothers of boys with FXS.

As was noted in the review of existing ASD literature, the literature surrounding child behavior in FXS and maternal stress is also remarkable for inconsistent consideration of how child behaviors may differentially impact different dimensions of parenting stress. Whereas some studies have used only total scores of maternal stress in their analyses (Mankowski, 2007; Richardson, 2010), others have focused only on one parenting stress dimension (e.g., Johnston et al., 2003). No known studies have yet allowed for an examination of how child behavioral characteristics in FXS impact parenting stress across child-related, parent-related, and parent-child interaction-related domains.

Maternal Factors

Premutation status in mothers of children with FXS has been shown to be associated with an increased risk of certain psychiatric disorders and/or symptoms which may increase their vulnerability to the stress associated with raising a child with FXS (Abbeduto et al., 2004; Franke et al., 1996). Elevated depressive symptoms, as well as

increased rates of affective and anxiety disorders, have been reported in these mothers (Bailey et al., 2007; Franke et al., 1996). There is increasing evidence to suggest that this increase in reported psychopathology reflects a true genetic predisposition and not just the impact of raising a child with a disability. For example, Franke and colleagues (1998) found that women with the FXS premutation with and without children affected by the disorder presented with similar levels of anxiety. In a more recent study, Roberts and colleagues (2009) found that approximately half of their sample of mothers with the FMR1 premutation met criteria for major depressive disorder *prior* to the birth of their first child with FXS.

Though available evidence points to an underlying genetic susceptibility to psychopathology in premutation carrier mothers, understanding the nature of this genetic liability has proven to be complex. Interactions between maternal genotype and environmental experiences (e.g., managing difficult child behaviors) appear to be at work in this group of mothers (Roberts et al., 2009). In a study using CGG repeat length as an indicator of genetic vulnerability, Seltzer and colleagues (2011) explored this complex interaction by examining how repeat length impacts the association between life stressors and psychological (depressive symptoms and anxiety) and physiological outcomes (cortisol response) in mothers with the premutation. Results suggested that mothers with midsize CGG expansions evidenced a greater susceptibility to environmental influences (positive and negative) than did mothers with smaller or larger expansions. Using a different index of genetic susceptibility, Hartley and colleagues (2012) provided further evidence of the complex gene-environment interactions impacting functioning in these mothers. Using a diathesis-stress model, the authors of this study examined the degree to

which maternal activation ratio (diathesis) impacts the association between child behavior problems (stress) and maternal cortisol responses. Results indicated that mothers with greater genetic risk (i.e., those with lower activation ratios) had a lower level of cortisol on mornings following days when their child with FXS displayed more problematic behaviors.

Summary

Like mothers of children with ASD, mothers of children with FXS report significant elevations in parenting stress. Underlying mechanisms for these elevations appear to involve interactions among multiple child, maternal, and environmental factors. Autistic behaviors, as well as more general problem behaviors, are linked to increases in parenting stress in mothers of children with FXS. Genetic vulnerabilities in these mothers associated with their premutation status appear to interact in dynamic ways with environmental stressors (e.g., child behavior problems) to impact the experience of stress.

Current Aims

The current study sought to increase understanding of how of specific child and maternal factors impact maternal parenting stress in three high-risk populations of mothers raising children with developmental disabilities with overlapping behavioral profiles. Mothers of children with IA, AFXS, and FXS represent three groups of mothers thought to have similar genetic risk factors which may impact their threshold for dealing with their child's difficult behaviors. Looking across these groups of genetically at-risk mothers of children with varying etiologies, as well as, varying degrees of behavioral symptomatology, allows for a unique examination of the child behavior – maternal stress

relationship. Specifically, looking across these three groups will allow for an improved understanding of the role that certain types of child behavioral characteristics may play in the excessive maternal stress levels typically observed in mothers of children with ASD compared to mothers of children with FXS. Because the existing literature lacks adequate data concerning the impact of child behavioral characteristics on the various dimensions of parenting stress, this present study considered the impact of child and maternal variables across child, parent, and child-parent interaction domains of parenting stress. This type of approach may have important implications for practice as elevations in differing types of parenting stress have been linked to different types of parenting outcomes (Holden & Banez, 1996). In mothers of boys with IA only, this study also examined the relationship between maternal BAP and the various dimensions of parenting stress. The following specific research questions and associated hypotheses were addressed:

1) How do levels of reported parent-related, child-related, and parent-child-interaction-related parenting stress vary across these three high-risk groups of mothers?

Specific hypothesis

Compared to scores from mothers of boys with FXS alone, scores from all three domains of parenting stress on the Parenting Stress Index-Short Form (PSI-SF Difficult Child, Parental Distress, and Parent-Child Dysfunctional Interaction domains) were predicted to be significantly higher for mothers of boys with IA and mothers of boys with AFXS. Parenting stress scores for mothers of boys with IA and mothers of boys with AFXS were not predicted to differ significantly.

2) Can we account for a meaningful amount of variability in maternal parenting stress in these three groups of high risk mothers using maternal (age and intellectual functioning) and child-level (age, ASD symptoms, general problem behaviors) variables?

Specific hypothesis

Looking at the total sample of mothers maternal and child-level variables were predicted to account for at least 25% of variability observed in maternal parenting stress across all three domains of parenting stress on the PSI-SF.

3) How do general child behavior problems influence the different dimensions of parenting stress across these three high-risk groups of mothers?

Specific Hypothesis

Total problem behavior scores on the Child Behavior Checklist (CBCL) were predicted to show a unique association with child-related stress scores on the Parenting Stress Inventory – Short Form (PSI-SF Difficult Child score) compared to other domains of parenting stress. The nature of this association was not predicted to vary significantly by group.

4) How do severity of symptoms of ASD, severity of general behavioral problems, and maternal features of the BAP differentially impact the experience of parenting stress in mothers of children with IA?

Specific Hypothesis

For mothers of children with IA, both total problem behavior scores on the CBCL and maternal scores on a measure assessing for features of the BAP (the Broad

Autism Phenotype Quotient; BAP-Q) were predicted to make significant contributions to the prediction of maternal parenting stress across dimensions of stress measured on the PSI-SF while controlling for child autism severity as measured by the SCQ.

Table 2.1

Summary of Articles Reviewed Employing PSI

	Sample Size N	Stress Dimension(s) assessed	Comparison group
Baker-Ericzen et al., 2005	110	PSI total score + All 3 subscales	Typically developing children
Bravo, 2006	234	PSI total score + All 3 subscales	None
Davis & Carter, 2008	108	PSI total score + All 3 subscales	None
Donenberg & Baker, 1993	64	PSI DC subscale	Typically developing children and children with externalizing behaviors
Ekas & Whitman, 2010	119	PSI-SF total score	None
Freeman et al., 1991	41	PSI total score + All 3 subscales	None
Hoffman et al., 2008	72	PSI total score	None
Holmberg, 2007	210	PSI total score	None
Kasari & Sigman, 1997	82	PSI total score + DC and PD subscales	Children diagnosed with ID and typically developing children
Lecavalier et al., 2006	243	PSI-SF total score	None

Table 2.1 cont.

Summary of Articles Reviewed Employing PSI

Mankowski, 2007	113	PSI-SF total score	Boys diagnosed with FXS and AFXS
Matthews, 2010	55	PSI total score	None
Richardson, 2010	30	PSI-total score	None
Tomanik et al., 2004	60	PSI-SF PD subscale	None

CHAPTER 3

METHOD

Participants

Participants were comprised of three groups of mothers: a) mothers of children with IA, b) mothers of children with FXS only, and c) mothers of children with AFXS. The primary sample of mothers came from an extant data base which includes 48 mothers and their male child with IA, 56 mothers and their male child with FXS, and 20 mothers and their male child with AFXS. All children from this data set were males between the ages of 1 and 14 years. Investigators originally selected this age range due to specific interests in the effects of child behavior on maternal outcomes. Because the challenges faced by parents of children approaching transition are often qualitatively different than those faced by parents of younger children, "childhood" was broadly defined as 14 and under by investigators originally collecting these data. This dataset is managed by Dr. Jane Roberts who is continually adding new data with ongoing studies. See Table 3.1 for maternal and child demographic information for this primary sample. An additional sample of 20 mothers of male children with IA was recruited to address secondary study questions. Due to the contribution of genetic factors in both ASD and FXS, only data from biological mothers were used in the current study. Additionally, because of the more variable expression of ASD and FXS in females, the current study only included data from mothers having at least one diagnosed male child.

Mothers of a Child with IA

The 48 mothers of children with IA from the extant data set were recruited through four primary sources: (1) the Autism Society of North Carolina's parent listserv; (2) the Autism Subject Registry Core of the UNC Neurodevelopmental Disorders Research Center; (3) existing studies at UNC (two ongoing studies of children with autism), and (4) ongoing research efforts of the USC Neurodevelopmental Research Lab. Autism status of each child was confirmed by an existing Autism Diagnostic and Observation Schedule (ADOS) and a current Childhood Autism Rating Scale (CARS). Children had to have received the ASD diagnosis at least one year prior to enrollment in the study. Record review and extensive family history interviews were conducted to ensure that none of the children with autism also had a diagnosis of FXS.

An additional sample of 20 mothers of children with idiopathic autism was recruited to allow for the collection of data regarding the association of maternal stress and characteristics of the broad autism phenotype in this population of mothers. Biological mothers and their male children aged 4-14 years were targeted in recruitment efforts. The target child age range for this secondary sample was chosen in an effort to maintain consistency with that of the primary sample. The minimum age requirement, however, was moved to four years for this sample to meet requirements of one study measures which differed from that completed by participants in the primary sample (the Social Communication Questionnaire which has a minimum age requirement of four years). Additional inclusion criteria included (a) available documentation to verify that diagnosis was made via ADOS administration by a qualified professional; including licensed psychologists and other qualified medical professionals, and (b) verification via

parent documentation of no known co-morbid developmental disability, including FXS. At the time of enrollment, time since diagnosis of ASD must have been ≥ 1 year.

Recruitment

Following collaborative review and study approval by the USC and Greenville Hospital System (GHS) IRBs (with GHS serving as the lead institution) in March 2012, the PI began pilot recruitment efforts for the new sample of 20 mothers of children with IA in April of 2012. This new sample was recruited through three primary sources: (1) The Children's Hospital Autism Wonders Program of GHS (the PI's place of employment); (2) two local parent support organizations – the Greenville offices of Family Connection of SC and the SC Autism Society; and (3) postings on two active on-line parent support networks for parents of children with ASD – the LUCAS Network and the on-line SC Autism Society Network.

Autism Wonders program recruitment plan. Autism Wonders is a program of the Children's Hospital of the Greenville Health System. This program offers families diagnostic services for ASD as well as assistance linking to community resources. Mothers meeting the inclusion criteria who were existing patients in the Children's Hospital Autism Wonders program within the Department of Developmental-Behavioral Pediatrics (D-B Peds) were provided information about the current study. These mothers were provided a standard flyer which included a brief overview of the present study, as well as an area where mothers could provide their written approval to be contacted about the study by providing their name and preferred contact information. Flyers were made available in patient waiting areas and select patient exam rooms. Additionally, clinicians

in the Autism Wonders program were provided study flyers and information about the current study via verbal presentation at weekly staff meetings and email notifications. Clinicians were asked (a) to provide flyers to eligible mothers coming in for follow-up visits and (b) to notify the principle PI when available on-site (this ranges from 25 to 30 hours per week) of mothers who provided written approval to be contacted on the standard flyer. When notified of this written approval, the PI met face to face with mothers to provide additional information, obtain consent, and distribute measures.

Recruitment via local support organization events. The PI attended local autism parent-support organization meetings and events sponsored by the Greenville offices of Family Connection of SC and the SC Autism Society (SCAS) to share information about the current study. Each month, Family Connection hosts a support meeting for parents and caregivers of children diagnosed with ASD. Approximately 5-10 mothers attend the Family Connection support meeting each month. The PI attended 4 meetings between June 2012 and May 2013 to provide both written and verbal information about the current study to parents in attendance. Mothers who provided written approval by supplying their contact information on the standard flyer were contacted via their preferred contact method following the meeting. In April of 2012, SCAS held its annual "Strides for Autism" walk – an event with an average attendance over the last four years of approximately 100 - 125 mothers of children on the autism spectrum. The PI obtained permission from SCAS representatives to set up a booth at this event where verbal and written information was provided about the present study. The PI distributed eight packets to mothers who expressed interest in the study by providing their contact information on the standard flyer. The consent form was reviewed and signed on site by

all interested mothers. In addition to the efforts described above, representatives from Family Connection and SCAS were provided study flyers and asked to make these available in their office waiting areas.

On-line support forum recruitment plan. The PI distributed information about the current study to mothers participating on two local on-line parent-support forums – the LUCAS Network and the SCAS on-line forum. Approximately 600 parents or caregivers of individuals diagnosed with ASD are members on these forums. With the moderators' permission, a brief text explanation of the study and the PI's contact information was posted on these forums on four occasions between November 2012 and May 2013. Interested mothers were mailed packets containing study questionnaires and a pre-paid return envelope.

Incentive Plan for New Recruits. Mothers who completed and returned study measures received $10 as a thank you for their participation in the study. Checks were mailed by a member of Dr. Roberts' lab within two weeks of receiving the completed packet of study measures.

Mothers of a Child with FXS only or AFXS

The data from the 56 mothers of children with FXS and the 20 mothers of children with AFXS were gathered via a study at the University of North Carolina at Chapel Hill study - *Family Adaptation to Fragile X Syndrome*. Recruitment of these families occurred through three main sources: (1) funded projects at University of North Carolina at Chapel Hill that had an enrolled sample of children with FXS; (2) pilot studies at the University of Kansas; and (3) the FX Subject Registry Core of the UNC

Neurodevelopmental Disorders Research Center. Record review confirmed the FXS premutation in all mothers, and FXS in all sons from this group. Though formal diagnosis of autistic disorder was not confirmed, behavioral criteria for autism was met by the 20 children comprising the FXS/autism group as evidenced by a CARS score above the autism cut-off.

Measures

Descriptions of measures used in the current study are included below. Table 3.2 provides a summary of measures completed by mothers comprising the existing dataset as well as measures administered to the 20 newly recruited mothers of sons with IA.

Demographic Information Form. Mothers from the extant dataset completed a general information form which asked for information about the mother's ethnic background, age, age at child's birth, marital status and education. The child's age, FXS or autism diagnosis dates, ethnic identity, and family income was also recorded on this form. The 20 newly recruited mothers were asked to provide basic demographic information on a form adapted from a template developed by members of the Neurodevelopmental Research lab for use in Dr. Roberts' ongoing studies. This demographic form asked for the same maternal and child demographic information as recorded on the general information form completed by mothers comprising the extant dataset. The 20 newly recruited mothers were also asked to indicate on this form if their child has been diagnosed with any other developmental disabilities which may cause them to be excluded from the present study (e.g., fragile X, Down syndrome, or other genetic conditions). Information recorded on these forms were used to determine parent-

level (e.g., maternal age and education), and child-level (e.g., age) co-variates to be included in this and other ongoing projects.

Maternal Measures

Parenting Stress. The Parenting Stress Index- Short Form (PSI/SF; Abidin, 1995) is a 36-item self- report measure that is used to assess parenting stress in parents of young children. Parents are asked to rate their agreement with statements using a 5-point Likert-type scale (1 = *strongly agree* to 5 = *strongly disagree*). The measure yields a Total Stress score which incorporates responses from three subscales: Parental Distress, Parent-Child Dysfunctional Interaction, and Difficult Child Characteristics. The Parental Distress subscale of the PSI-SF assesses a parent's sense of competence in the parenting role, stress related to restrictions on his/her life, aspects of social support, as well as some symptoms of depression. The Parent-Child Dysfunctional Interaction subscale deals with how a child has met or failed to meet a parent's expectations, as well as a parent's satisfaction with interactions with his/her child. The Difficult Child subscale assesses how difficult or easy the parent perceives his/her child's challenging behaviors. The PSI-SF has strong psychometric properties including good test-retest reliability (r=.84) and internal consistency (α=.91). Evidence of validity is based on correlation with the full length version (r=.94). The PSI/SF has been used widely in studies of parents of children with autism and other developmental disabilities (Davis & Carter, 2008; Tomanik et al., 2004). The measure typically takes approximately 10 minutes to complete. In the current study, each subscale score served as a dependent variable given the PI's primary interest in the association between problem behaviors and the various dimensions of maternal

parenting stress. In the current sample, Cronbach's alpha was .77 for the PD subscale, .83 for the P-CDI subscale, and .90 for the DC subscale.

Maternal IQ. For mother's from the extant dataset, the Wechsler Abbreviated Scale of Intelligence (WASI; Psychological Corporation, 1999) was used to provide an estimate of cognitive functioning. The WASI consists of four subtests (Vocabulary, Similarities, Block Design, and Matrix Reasoning) which together produce a composite full-scale IQ score (FSIQ), as well as Verbal and Performance IQ standard scores (VIQ and PIQ). An estimate of general intellectual ability can be obtained from administering the two subtest form of the WASI, which includes only the Vocabulary and Matrix Reasoning subtests. This abbreviated version can be given in about 15-30 minutes, and provides only the FSIQ score. Reliability for the adult FSIQ-2 has been reported at .96 (Psychological Corporation, 1999).

Maternal Characteristics of the Broad Autism Phenotype. The new sample of mothers of children with idiopathic autism recruited as part of the current study were administered a measure to assess characteristics of the broad autism phenotype. The Broad Autism Phenotype Questionnaire (BAP-Q; Hurley, Losh, et al., 2006), which is titled The Personality Styles and Preferences Questionnaire (PSPQ-S) was administered. This self-report questionnaire is comprised of 36 questions that tap social behaviors and personality styles believed to constitute the Broad Autism Phenotype. Participants are asked to indicate the degree to which they endorse each statement on a scale from 1 (very rarely) to 6 (very often). Total scores on the BAP-Q were used to address the secondary research question. Internal consistency has been reported at .95 for the BAP-Q total score, with individual subscales ranging from .85 (Pragmatic Language subscale) to .94

(Aloof subscale) (Hurley et al., 2007). For the current sample, internal consistency across the three subscales fell within acceptable ranges (Aloof subscale α = .95, Pragmatic Language subscale α = .80, and Rigid subscale α = .87).

Child Measures

General Child Behavior Problems. Two versions of the *Child Behavior Checklist* (the CBCL 1½ to 5 and the CBCL 6-18; Achenbach, 1991; Achenbach & Rescorla, 2000) were used as an assessment of the child's competencies and behavioral/emotional problems. The CBCL is a standardized questionnaire that asks parents to rate statements describing various child behaviors on a three-point scale ranging from 0 (not true) to 2 (very true or often). The two versions of the CBCL are designed to evaluate similar constructs across age groups. The 99-item CBCL 1½ to 5 produces factor scores across the following areas of behavioral symptomatology: Anxious/Depressed, Somatic Complaints, Withdrawn, Emotionally Reactive, Attention Problems, Aggressive Behavior, and Sleep Problems. These factor scores contribute to three broad scales: Internalizing, Externalizing, and Total Problems. Internal consistency for the CBCL Total Problems score was reported at .76 with a range from .53 on the Withdrawn subscale to .64 on the Anxious/Depressed subscale. Test-retest reliability for the Total Problem score was .90 with a range on subscales from .68 on the Anxious/Depressed subscale to .92 on the Sleep Problems subscale. The 113-item CBCL 6-18 produces scores for the child's competencies in the following areas: Activities, Social, and School. It also produces scores corresponding to Internalizing (i.e., Anxious/Depressed, Withdrawn/Depressed, and Somatic Complaints) and Externalizing (i.e., Rule-Breaking and Aggressive Behaviors) Problems, as well as a Total Problems score. Test-retest reliability for the

Total Problem scale on the CBCL 6-18 is .94 with a range on subscales from .82 on Anxious/Depressed to .92 on Somatic Complaints, Attention Problems, and Externalizing Behavior. Internal consistency for the CBCL for ages 6-18 years was .81 for Total Problems, ranging from .64 for Somatic Complaints to .82 for Aggressive Behavior. Both questionnaires typically require approximately 20 minutes to complete. Based upon precedent established in the existing literature, Total Problem scores on the CBCL served as an independent variable in analyses conducted to address the primary and secondary research questions in the current study. Internal consistency for the Total Problem Behavior score for the current sample was .76.

Child Autistic Symptoms (Childhood Autism Rating Scale, CARS; Schopler et al., 1988). For children from the extant data set, the CARS was used to provide a rating of autistic features. The CARS is a 15-item measure on which professionals rate a child across 15 areas using a score from 1 (within normal limits for age or skill level) to 4 (severely abnormal for age or skill level). The following areas are assessed: Relating to People; Imitation; Emotional Response; Body Use; Object Use; Adaptation to Change; Visual Response; Listening Response; Taste, Smell, and Touch Responses; Fear and Nervousness; Verbal Communication; Nonverbal Communication; Activity Level; Intellectual Response; and General Impression of Autism. The CARS has good internal consistency (.94) and test-retest stability over a one-year period (.88). Inter-rater reliability, which is crucial in a behavioral observation measure, is reported at (.71) by the CARS manual. The total score on the CARS is used as an index of autistic symptom severity in the current study. CARS data were not collected for participants comprising the new sample in the present study. The 75 children from the extant dataset with FXS

syndrome were grouped depending on whether their total CARS score was above or below the autism cut-off. Out of the 76 children with FXS, 20 (25%) children were placed into the FXS/autism group, while the remaining 56 (75%) children comprise the FXS only group.

Social Communication Questionnaire. Because clinician administration of the CARS with the 20 newly recruited mothers of boys with IA was beyond the scope of the current study, the Social Communication Questionnaire (SCQ) was used to reflect severity of autistic symptoms in sons of newly recruited mothers. The SCQ is a 40-item parent questionnaire designed as an autism screening instrument for individuals aged 4 years and up. The items on the SCQ are derived from the Autism Diagnostic Interview – Revised (ADI-R) (Lord et al., 1994) and cover the areas of communication, reciprocal social interactions, and restricted and repetitive behaviors and interests (Rutter, Bailey, & Lord, 2003). Each item is checked as 'yes' or 'no', and assigned a point rating of '1' (presence of abnormal behavior) or '0' (absence of abnormal behavior). The first item is not included in the scoring, as it indicates if the child has sufficient verbal skills for language items to be scored. If the child is not scored as verbal, the six language items are skipped. The points are summed and the cut-off is established as ≥22 for autism and ≥15 for ASD. Good internal consistency has been reported for the SCQ (between .81 and .93). Using a total score of 15 or higher for differentiating ASD from other diagnoses, sensitivity of .85 and specificity of .75 has been reported for the SCQ. Using the same cut-off for differentiating autism from other diagnoses (excluding intellectual disability), sensitivity of .96 and specificity of .80 have been reported. The SCQ typically takes approximately 10 minutes to complete. Total scores on the SCQ were used to address the

secondary research question. For the current sample, internal consistency for SCQ total score was .80.

Procedures

Extant Data. The PI is included as a member of the research team on Dr. Roberts' IRB through USC, and hence was approved access to the extant dataset. A dataset was compiled according to the inclusion criteria for the current study.

New IA Recruits. All mothers fitting study criteria who expressed an interest in participating in the study by providing their information on the standard flyer were contacted by the investigator according to their indicated preferred method of contact. For mothers indicating a preference for face to face contact with the PI when available (i.e., during already scheduled medical appointments at D-B Peds, or during parent support meetings) the PI arranged for a brief on-site meeting to discuss the study. During this short face to face meeting, the PI obtained consent, distributed measures, and discussed collection options with mothers. Mothers were given the option to either complete measures before leaving the site (during the medical appointment or support meeting), or to complete measures at home and return them in a self-addressed stamped envelope provided by the PI. For mothers who opted to complete measures at home, the PI explained that measures should be returned within two weeks from the date of the initial contact, and asked mothers if they felt that completing and returning the measures within this time frame was reasonable. If the measures were not returned within the two-week time frame, the PI contacted participants either by phone or e-mail (depending on the mother's preferred method of contact) on two occasions following distribution of

measures. The PI contacted mothers once two weeks after they received the measures, and again three weeks after consent. During both contacts, the PI thanked participants for agreeing to participate in the study and requested that measures be returned within one week. During the second contact, the PI explained that this would be the last contact regarding the study, and asked that participants call if they would like additional time to complete and return the measures.

For mothers indicating their interest in the study on the standard form who did opt for a face-to-face meeting, the PI made an initial contact according to their indicated preferred method of contact. In this phone or e-mail contact, the PI answered any questions the mother had about the study, reviewed the consent form, and requested verbal permission to mail the study packet to their preferred address. The PI requested that measures be returned within two weeks from the date of the initial contact, and provided instructions for returning the forms in the enclosed self-addressed stamped envelope. Follow-up procedures again involved contact either by phone or email two and three weeks after distribution of measures.

Overall recruitment efforts resulted in 49 study packets being distributed either by mail or in person to potential study participants. Of these 49, 24 packets (49%) were returned. Of the returned packets, four were missing one or more study measures. Attempts to obtain these missing materials were not successful, which ultimately resulted in a total sample size of 20 for secondary analyses.

After they were completed and returned, the PI scored all measures. Twenty percent of scoring was verified by a consultant familiar with measures and statistical

software used in this study. Once scoring was verified, the PI created a dataset using Statistical Package for the Social Sciences version 21.0 (SPSS, Inc., Chicago IL). After the data were entered by the PI, the consultant verified the database by confirming 20% of entries. Once datasets were compiled and verified, the PI conducted all relevant analyses.

Analysis

A multivariate multiple regression approach was employed in the current study due to the associated increase in power as compared to the alternative of running separate regression analyses for each of the three outcome variables. Specifically, because one runs the risk of multiplying error rates by testing each outcome variable separately; a multivariate approach is preferable in that it allows one to maintain a constant overall Type I error rate regardless of the number of variables tested. Also, because the three outcome variables of interest in this study are highly correlated, a multivariate approach prevents us from reanalyzing the same variance through separate regressions. A test-wise significance level of $p < .05$ was established *a priori* for all analyses.

A multivariate regression analysis using PSI-SF DC, PSI-SF PD, and PSI-SF P-CDI scores as dependent variables with Group as the fixed factor was conducted to address Hypotheses 1. A Bonferonni adjusted alpha level was used for post hoc analyses to assess the exact nature of significant differences among groups. To address Hypotheses 2 and 3, a multivariate multiple regression analysis was conducted to assess the ability of two child-level variables (ASD symptom severity and severity of general problem behaviors) to predict parenting stress for the entire primary sample of mothers as

measured by the three subscales of the PSI-SF. In addition to these two primary variables of interest, relevant covariates identified via preliminary analyses were tested for inclusion in the final model (maternal age and maternal IQ). Because the investigator was interested in how the impact of CBCL scores on parenting stress may vary according to group, the interaction of group and CBCL score was also tested for inclusion in the final model. A centering approach was applied in the process of testing for and probing interaction effects. Dummy coding (for the group variable) was used in the post-hoc probing of the significant interaction.

To address secondary aims (Hypothesis 4), a multivariate multiple regression analysis was conducted to assess the ability of two child-level (ASD symptom severity and severity of general problem behaviors) and one parent-level variable (characteristics of the BAP) to predict parenting stress as measured by the three subscales of the PSI-SF. In addition to these three primary variables of interest, maternal age was tested for inclusion in the final model as it was identified as a relevant covariate during preliminary analyses. Interaction effects between predictor variables, and between predictor variables and covariates were tested for inclusion in the final model. A centering approach was applied in the process of testing for interaction effects. Results of probing procedures revealed no significant interactions among predictor variables.

Initially, to address Hypothesis 4, CBCL Total Problem Behavior score, SCQ score, and PSPQ Total score (the BAP measure) were entered as independent variables, with maternal age as a covariate, and the three subscales of the PSI-SF as dependent variables. None of the entered predictor variables or the covariate entered in this first stage contributed significantly to the prediction of the combined dependent variables.

Because of the exploratory nature of this secondary analysis, the PI then removed the PSPQ Total score from the model and tested for effects of the individual subscales of the PSPQ (Pragmatic Language, Rigid, and Aloof subscales). Upon entering these three subscales, one (the Rigid subscale) subscale was found to exert a significant effect on two of the PSI-SF subscales, and hence was retained in the final model which included SQC Total score, CBCL Total Problem Behavior score, PSPQ Rigid score as predictor variables, maternal age as a covariate, and the three subscales of the PSI-SF as dependent variables.

Table 3.1

Child and Maternal Descriptives for the Primary Sample

	Total sample	Idiopathic autism group	Autism with fragile X group	Fragile X alone group
	N = 124	n = 48	n = 20	n = 56
Child age (in months)				
M	75.54	83.72	67.79	68.87
SD	46.44	35.53	44.12	54.28
Maternal age (in years)				
M	36.28	38.15	34.37	35.15
SD	5.92	6.25	7.05	4.83
Maternal IQ (WAIS SS)				
M	111.09	117.16	109.72	106.88
SD	13.02	10.10	13.66	12.70
Total problem behavior score (CBCL T-score)				
M	59.17	63.86	62.68	54.52
SD	10.21	8.51	8.08	10.25
Total score of autism severity (CARS total score)				
M	30.18	35.37	34.92	24.56
SD	6.75	4.53	5.08	3.26

Table 3.1 continued

Child and Maternal Descriptives for the Primary Sample

	Total sample	Idiopathic autism group	Autism with fragile X group	Fragile X alone group
	N = 124	n = 48	n = 20	n = 56
Total Parenting stress score (PSI-SF total score)				
M	86.86	94.78	91.74	80.31
SD	20.86	17.99	21.48	20.84
Range	46-137	58-137	56-130	46-130
Parent-related stress score (PSI-SF PD score)				
M	28.47	30.92	30.21	26.71
SD	9.37	8.59	9.61	9.45
Range	12-53	13-48	13-53	12-47
Child-related stress score (PSI-SF DC score)				
M	31.06	35.64	32.74	27.63
SD	9.40	8.10	9.97	8.79
Range	13-53	19-53	15-51	13-45
Parent-child Interaction stress Score (PSI-SF P-CDI score)				
M	27.34	28.22	28.79	25.98
SD	6.21	6.59	5.37	6.12
Range	17-48	19-48	20-38	17-48

Table 3.2

Measures Completed by Participant Group

	Extant dataset IA *n* = 48	Extant dataset AFXS *n* = 20	Extant dataset FXS alone *n* = 56	New IA recruits *n* = 20
Demographic Form	√	√	√	√
PSI-SF	√	√	√	√
CBCL	√	√	√	√
CARS	√	√	√	
SCQ				√
BAP-Q				√

CHAPTER 4

RESULTS

Preliminary Analyses

Analyses in the current study were conducted using the Statistical Package for the Social Sciences version 21.0 (SPSS, Inc., Chicago IL).

Initial examination and testing assumptions. The first stage of data analysis involved screening for missing data, normality, multicollinearity, outliers, and errors. Missing data were identified by inspecting frequency tables. For all three subscales of the PSI-SF, data were complete for 84% of cases. For CBCL Total Score, data were complete for 84% of cases. For CARS Total Score, data were complete for 95% of cases. For WASI IQ Standard Score, data were complete for 89% of cases. Data for child age and maternal age were complete for 96% and 100% of cases, respectively. The "Exclude Cases Pairwise" option was chosen for all analyses such that cases were excluded only if they were missing data required for the specific analysis at hand. This method for handling missing data was chosen as original raw data files were not available for the extant data set such that missing data could be imputed. The data were then examined for normal distribution of variables. Histograms and Normal Q-Q plots were visually inspected for all variables of interest. The Shapiro-Wilk Test of Normality was used to statistically assess for normal distribution. Distributions for all continuous variables met normal distribution criteria with the exception of CARS total score, child age, and the P-

CDI subscale of the PSI. Log 10 transformation procedures were applied these variables appearing to violate the normality assumption. Next, the investigator examined correlations among all variables by generating a correlation matrix. Pearson Product Moment correlation coefficients were computed and can be found in Table 4.1. Independent variables that were found to significantly correlate with the dependent variables included Cars Total score (r = .213, p <.05 for the PD subscale of the PSI-SF; r = .248, p <.05 for the P-CDI subscale of the PSI-SF; and r = .332, p < .01 for the DC subscale of the PSI-SF), CBCL Total score (r = .539, p < .01 for the PD subscale of the PSI-SF; r = .544, p <.01 for the P-CDI subscale of the PSI-SF; and r = .720, p < .01 for the DC subscale of the PSI-SF), and WASI IQ scores (r = .423, p <.01 for the DC subscale of the PSI-SF). Data were then screened for multicollinearity. Variance Inflation Factor (VIF) estimates were examined for each variable for overly high correlations among the independent variables. All VIFs were less than five, indicating that multicollinearity was not problematic with respect to stability of the regression coefficients. Bivariate scatter plots were constructed to allow for inspection of linearity between independent and dependent variables. Visual inspection of plots indicated linear relationships between variables of interest and outcome variables. Both independent and dependent variables were examined for univariate and multivariate outliers using histograms and normality plots. The assumption of homoscedasticity was examined to minimize biased significance levels through scatter plots of the residuals.

Descriptive statistics. The average age of mothers was 36.3 years (SD = 5.9; range = 20 – 51) for the total sample. Mothers from the IA group were significantly older than mothers from the other two groups. The average age of male children for the total

sample was 6.3 years (SD = 3.9; range = 11 months – 14.6 years), with no differences among groups reaching statistical significance. The mean WASI IQ score for mothers from the total sample was 111, with mothers from the IA group having significantly higher scores than mothers from the other two groups. Mothers were predominantly Caucasian (74%) and more than half reported having an educational background of a four-year college or beyond (55%). Twenty-one percent of the total sample reported income in the low-income range (<200% poverty level). Table 3.1 provides a summary of descriptive statistics for child and maternal sociodemographic and study variables for the total sample, and for each of the three maternal groups (ASD, FXS, and AFXS).

PSI Difficult Child (PSI-SF DC), Parent Distress (PSI-SF PD), Parent-Child Dysfunctional Interaction (PSI-SF P-CDI), and Total Stress scores were compared to published norms, as cited in the PSI manual (clinical cutoff = 90th percentile, Abidin, 1995). Forty-three percent of mothers from the total sample reported total stress levels exceeding the clinical cutoff (n = 46). The percentage of mothers exceeding the total score cutoff by group were as follows: ASD = 67%, FXS = 25%, AFXS = 47%).

Primary Analyses

Hypothesis 1: Compared to scores from mothers of boys with FXS alone, scores from all three domains of the Parenting Stress Index-Short Form (PSI-SF Difficult Child, Parental Distress, and Parent-Child Dysfunctional Interaction domains) will be significantly higher for mothers of boys with IA and mothers of boys with AFXS. Parenting stress scores for mothers of boys with IA and mothers of boys with AFXS are not predicted to differ significantly.

Results indicated a significant difference among groups on the combined dependent variables, F (6, 202) = 3.25, p = .005; Wilks' Lambda = .83; partial eta squared = .088. When the results for the dependent variables were considered separately, the only difference to reach statistical significance was for the DC subscale of the PSI-SF, F (2, 103) = 9.01, p = .000, partial eta squared = .15. Multivariate results and between-subjects effects for this analysis are summarized in Table 4.2. Follow-up analyses were conducted to identify the exact nature of significant differences. Using a Bonferroni adjusted alpha level, a significant difference in DC scores was found only between the IA and FXS groups (p = .000), with mothers in the IA group reporting higher DC scores than mothers in the FXS group. No significant differences were found between the IA and AFXS groups, or between the AFXS and FXS groups. Hence, the prediction that scores from all three domains of the PSI-SF would be significantly higher for mothers of boys with IA and mothers of boys with AFXS was not supported. Instead, significant differences among groups were only observed for the Difficult Child subscale of the PSI-SF, and the only significant difference found in scores on this domain was between mothers of boys with idiopathic autism and mothers of boys with fragile X alone (with the IA group having higher scores than the FXS group).

Hypothesis 2: Looking at the total sample of mothers, maternal and child-level variables will account for at least 25% variability observed in maternal parenting stress across all three domains of parenting stress on the PSI-SF.

Hypothesis 3: Total problem behavior scores on the Child Behavior Checklist (CBCL) will show a unique association with child-related stress scores on the Parenting Stress Inventory – Short Form (PSI-SF Difficult Child score) compared to

other domains of parenting stress. The nature of this association is not predicted to vary significantly by group.

In the final model, CBCL Total Problem Behavior score, CARS Total score, maternal IQ, and the interaction between group and the CBCL Total Problem Behavior score were entered into a multivariate multiple regression as covariates, with group as a fixed factor, and PSI-SF P-CDI, PSI-SF DC, and PSI-SF PD as dependent variables. Multivariate results and between-subjects effects for this analysis are provided in Table 4.3. The total variance explained by the model was 30.8% for the PSI-SF P-CDI (F (7, 79) = 5.035, p=.000), 57.8 % for the PSI-SF DC (F (7, 79) = 15.466, p=.000), and 19.8% for the PSI-SF PD (F (7, 79) = 2.780, p=.012). Hence, the hypothesis that maternal and child-level variables would account for at least 25% variability observed in maternal parenting stress across all three domains of parenting stress on the PSI-SF was partially supported as the total variance explained by the model for two of the three parenting stress subscales exceeded the hypothesized level.

In the final model, child problem behavior (CBCL Total Score) was found to significantly predict all three subscales of the PSI-SF (partial eta squared = .182 for the P-CDI subscale, p = .000; partial eta squared = .45, p =.000 for the DC subscale; and partial eta squared = .128, p = .001 for the PD subscale). Maternal IQ (WASI Standard Score) was found to significantly predict the DC subscale of the PSI-SF (partial eta squared = .152, p = .000). The interaction between group and CBCL Total score was also found to significantly predict the DC subscale of the PSI-SF (partial eta squared = .092, p = .022). Post-hoc probing of this significant interaction effect indicated that each of the three simple slopes (corresponding to each maternal group) was significantly different from

zero ($t(85) = 4.94$, $p = .000$ for the FXS group, $t(85) = 4.45$, $p = .000$ for the AFXS group, and $t(85) = 4.62$, $p = .000$ for the IA group). Inspection of this interaction indicated that CBCL scores showed a stronger relationship with child-related parenting stress as measured by the DC subscale of the PSI-SF for mothers in the IA and AFXS groups than with mothers in the FXS alone group (See Figure 4.1).

To address Hypothesis 3, partial eta squared values for CBCL Total Problem Behavior score were compared across dependent variables (PSI-SF P-CDI, PSI-SF DC, and PSI-SF PD) for each group of mothers. Partial eta squared provides a measure of the proportion of the variance in the dependent variable attributable to the factor in question. For mothers from the IA group, while obtained partial eta squared values indicated that 50% of the variance in PSI-SF DC scores was attributable to CBCL Total Problem Behavior scores, only 30% of the variance in PSI-SF P-CDI scores, and 17% of the variance in PSI-SF PD scores, was attributable to CBCL scores. Similarly, for mothers from the AFXS group, results indicated that 68% of the variance in PSI-SF DC scores was attributable to CBCL scores, while only 24% of the variance in PSI-SF P-CDI scores, and 27% of the variance in PSI-SF PD scores was attributable to CBCL ratings. In contrast, for mothers from the FXS alone group, obtained partial eta squared values indicated that 26% of the variance in PSI-SF DC scores, 21% of the variance in PSI-SF P-CDI scores, and 27% of the variance in PSI-SF PD scores, was attributable to CBCL Total Problem Behavior scores. These results provided partial support for the prediction that Total Problem Behavior scores on the CBCL would show a unique association with child-related stress scores on the PSI-SF Difficult Child subscale compared to other domains of parenting stress. While it was predicted that no group differences would be

observed in the nature of this association, the unique association between CBCL Total Problem Behavior scores and child-related stress scores did not hold for mothers from the FXS alone group.

Secondary Analyses

Hypothesis 4: For mothers of children with IA, both total problem behavior scores on the CBCL and maternal scores on a measure assessing for features of the broad autism phenotype (the BAP-Q) will make significant contributions to the prediction of maternal parenting stress across dimensions of stress measured on the PSI-SF while controlling for child autism severity as measured by the SCQ.

Initial examination and testing assumptions. The first stage of data analysis involved assessing for internal consistency on measures for this sample, and screening for missing data, normality, multicollinearity, outliers, and errors. Missing data were identified by inspecting frequency tables. For all three subscales of the PSI-SF, data were complete for 90.9% of cases. For CBCL Total Score, data were complete for 86.4% of cases. For SCQ Total Score, data were complete for 95.5% of cases. For all three subscales of the BAP-Q, data were complete for 90.9% of cases. Data for child age and maternal age were complete for 95.5%. The "Exclude Cases Pairwise" option was chosen for all analyses such that cases were excluded only if they were missing data required for the specific analysis at hand. This method was chosen due to the nature of missing data for measures collected to address secondary aims. Specifically, individual data points were not found to be missing in the raw data files. Instead, missing data were for entire measures not returned with study packets. Hence, imputation techniques were not

appropriate for this analysis. The data were then examined for normal distribution of variables. Histograms and Normal Q-Q plots were visually inspected for all variables of interest. The Shapiro-Wilk Test of Normality was used to statistically assess for normal distribution. Distributions for all continuous variables met normal distribution criteria. Next, the investigator examined correlations among all variables by generating a correlation matrix. Pearson Product Moment correlation coefficients were computed and can be found in Table 4.4. Independent variables that were found to significantly correlate with the dependent variables included SCQ Total score (r = .541, p <.05 for the PD subscale of the PSI-SF; r = .678, p <.01 for the P-CDI subscale of the PSI-SF; and r = .462, p < .05 for the DC subscale of the PSI-SF), CBCL Total score (r = .526, p < .05 for the P-CDI subscale of the PSI-SF and r = .614, p < .01 for the DC subscale of the PSI-SF), PSPQ Total score (r = .574, p < .01 for the DC subscale of the PSI-SF), the Aloof subscale of the BAP-Q (r = .522, p < .05 for the DC subscale of the PSI-SF), the Pragmatic Language subscale of the BAP-Q (r = .459, p < .05 for the P-CDI subscale of the PSI-SF), the Rigid subscale of the BAP-Q (r = .591, p < .01 for the DC subscale of the PSI-SF), and maternal age (r = .452, p <.05 for the PD subscale of the PSI-SF). Data were then screened for multicollinearity. Variance Inflation Factor (VIF) estimates were examined for each variable for overly high correlations among the independent variables. All VIFs were less than five, indicating that multicollinearity was not problematic with respect to stability of the regression coefficients. Bivariate scatter plots were constructed to allow for inspection of linearity between independent and dependent variables. Visual inspection of plots indicated linear relationships between variables of interest and outcome variables. Both independent and dependent variables were examined for

univariate and multivariate outliers using histograms and normality plots. The assumption of homoscedasticity was examined to minimize biased significance levels through scatter plots of the residuals.

Descriptive statistics. The average age for mothers from the secondary sample was 37.7 years (SD = 7.0; range = 25.7 – 48.2). The average child age for this sample was 9.0 years (SD = 3.4; range = 4.0 – 14.6). Ninety-one percent of mothers from this secondary sample was Caucasian, and 35% reported having at least a four-year college degree. Thirty-one percent of these mothers reported income at the low-income level (<200% poverty level). Table 4.5 provides a summary of descriptive statistics for child and maternal sociodemographic and study variables for the secondary sample of mothers.

PSI Difficult Child (PSI-SF DC), Parent Distress (PSI-SF PD), Parent-Child Dysfunctional Interaction (PSI-SF P-CDI), and Total Stress scores were compared to published norms, as cited in the PSI manual (clinical cutoff = 90th percentile, Abidin, 1995). Seventy-six percent of mothers from the secondary sample reported total stress levels exceeding the clinical cutoff (n = 16).

Analysis. In the final model, CBCL Total Problem Behavior score, SCQ Total score, BAP-Q Rigid subscale score, and maternal age were entered into a multivariate multiple regression as covariates, and PSI-SF P-CDI, PSI-SF DC, and PSI-SF PD as dependent variables. Multivariate results and between-subjects effects for this analysis are provided in Table 4.6. The total variance explained by the model was 57.1% for the PSI-SF P-CDI (F (4, 15) = 4.984, p=.009), 67.5 % for the PSI-SF DC (F (4, 15) = 7.798, p=.001), and 56.7% for the PSI-SF PD (F (4, 15) = 4.905, p=.01). The only variable to

make a significant contribution to the prediction of the combined dependent variables was the Rigid score from the BAP-Q, F (3, 15) = 3.556, p = .045; Wilks' Lambda = .549; partial eta squared = .451. When the results for the dependent variables were considered separately, only the PD and DC subscales of the PSI-SF were significantly impacted by BAP-Q Rigid scores (F (1, 15) = 6.565, p = .022, partial eta squared = .304 and F (1, 15) = 11.566, p = .004, partial eta squared = .435). Additionally, SCQ was found to have a significant impact on the P-CDI subscale only, F (1, 15) = 4.851, p = .044, partial eta squared = .244. Hence, the prediction that child problem behaviors and maternal features of the BAP would make the most significant contributions to the prediction of maternal parenting stress was only partially supported. While one subscale of the BAP-Q was found to make a significant contribution to parent-related and parent-child-interaction-related stress domains, general child behavior problems were not found to have a significant effect on parenting stress.

Table 4.1

Pearson Correlation Matrix for Variables in the Primary Analysis

	Mom age	Child age	Cars total	CBCL	Income	WASI IQ	PD	P-CDI	DC	Total stress
Mom age	1	.495**	.168	.028	.091	.213*	-.069	-.127	.016	-.062
n	130	125	124	109	122	116	109	109	109	109
Child age	.495**	1	.127	.196*	-.271**	-.066	.009	.057	.076	.055
n	125	125	122	109	119	115	109	109	109	109
Cars total	.168	.127	1	.424**	-.087	.275**	.213*	.248*	.332**	.317**
n	124	122	124	107	117	114	106	106	106	106
CBCL	.028	.196*	.424**	1	-.172	.291*	.539**	.544**	.720**	.726**
n	109	109	107	109	108	104	106	106	106	106
WASI IQ	.213	-.066	.275**	.291**	.281**	1	.168	.052	.423**	.281**
n	116	115	114	104	111	116	104	104	104	104
PD	-.069	.009	.213*	.539**	-.189	.168	1	.442**	.567**	.836**
n	109	109	106	106	107	104	109	109	109	109

Table 4.1 cont.

Pearson Correlation Matrix for Variables in the Primary Analysis

Mom age	Child age	Cars total	CBCL	Income	WASI IQ	PD	P-CDI	DC	Total stress	
P-CDI	-.127	.057	.248*	.544**	-.112	.052	.442**	1	.593**	.763*
n	109	109	106	106	107	104	109	109	109	109
DC	.016	.076	.332**	.720**	-.09	.423**	.567**	.593**	1	.882*
n	109	109	106	106	107	104	109	109	109	109
Total stress	-.062	.055	.317**	.726**	-.159	.281**	.836**	.763**	.882**	1
n	109	109	106	106	107	104	109	109	109	109

Table 4.2

Hypothesis 1 Multivariate Results

Effect	Wilks' Lambda	*F*	*df*	Error *df*	sig.
Intercept	.004	8637.203	3	101	.000
GROUP	.832	3.250	6	202	.005

Tests of Between-Subjects Effects

Source	Dependent Variable	*SS*	*df*	*MS*	*F*	sig.
Corrected Model	Log_P-CDI	.04	2	.02	2.53	.085
	DC	1404.69	2	702.35	9.10	.000
	PD	422.61	2	211.30	2.50	.087
Intercept	Log_P-CDI	184.52	1	184.52	21285.44	.000
	DC	92150.26	2	92150.26	1194.21	.000
	PD	77133.22	2	77133.22	911.46	.000
GROUP	Log_P-CDI	.04	2	.02	2.53	.085
	DC	1404.69	2	702.35	9.10	.000
	PD	422.61	2	211.30	2.50	.087
Error	Log_P-CDI	.89	103	.01		
	DC	7947.91	103	77.16		
	PD	8716.50	103	84.63		

Table 4.2 cont.

Hypothesis 1 Multivariate Results

Total	Log_P-CDI	216.13	106
	DC	112962.00	106
	PD	96841.00	106
Corrected Total	Log_P-CDI	.94	105
	DC	9352.60	105
	PD	9139.10	105

Log_P-CDI R^2 = .047 (adj. R^2 = .028), PD R^2 = .046 (adj. R^2 = .028),

DC R^2 = .150 (adj. R^2 = .134)

Table 4.3

Hypotheses 2 and 3 Multivariate Results

Effect	Wilks' Lambda	*F*	*df*	Error *df*	sig.
Intercept	.004	6548.49	3	77	.000
GROUP	.941	.80	6	154	.030
CBCL cent.	.546	21.34	3	77	.000
WASI IQ cent.	.764	7.94	3	77	.000
CARS cent.	.938	1.70	3	77	.173
Group*CBCL	.854	2.10	6	154	.076

Tests of Between-Subjects Effects

Source	Dependent Variable	*SS*	*df*	*MS*	*F*	sig.
Corrected Model	Log_P-CDI	.230	7	.03	5.04	.000
	DC	4250.13	7	607.16	15.47	.000
	PD	1172.26	7	167.47	2.78	.012
Intercept	Log_P-CDI	125.16	1	125.16	19138.43	.000
	DC	61526.58	1	61526.58	1567.23	.000
	PD	57532.44	1	57532.44	954.96	.000

Table 4.3 cont.

Hypotheses 2 and 3 Multivariate Results

GROUP	Log_P-CDI	.02	2	.01	1.72	.042
	DC	1.08	2	.54	.01	.000
	PD	20.77	2	10.39	.17	.004
CBCL cent.	Log_P-CDI	.12	1	.12	17.57	.000
	DC	2535.89	1	2535.89	64.60	.000
	PD	700.32	1	700.32	11.62	.001
WASI IQ cent.	Log_P-CDI	.004	1	.004	.59	.445
	DC	557.13	1	557.13	14.19	.000
	PD	7.65	1	7.65	.13	.723
CARS cent.	Log_P-CDI	.03	1	.03	3.77	.056
	DC	.48	1	.48	.01	.912
	PD	1.73	1	1.73	.03	.866
GROUP*CBCL cent.	Log_P-CDI	.01	2	.01	.74	.480
	DC	314.18	2	157.09	4.00	.022
	PD	8.04	2	4.02	.07	.935

Table 4.3 cont.

Hypotheses 2 and 3 Multivariate Results

Error	Log_P-CDI	.52	79	.01
	DC	3101.39	79	39.26
	PD	4759.42	79	60.25
Total	Log_P-CDI	180.64	87	
	DC	99538.00	87	
	PD	88488.00	87	
Corrected Total	Log_P-CDI	.75	86	
	DC	7351.52	86	
	PD	5931.68	86	

Log_P-CDI R^2 = .308 (adj. R^2 = .247), DC R^2 = .578 (adj. R^2 = .541)

PD R^2 = .198 (adj. R^2 = .127)

Figure 4.1

Interaction of GROUP and CBCL

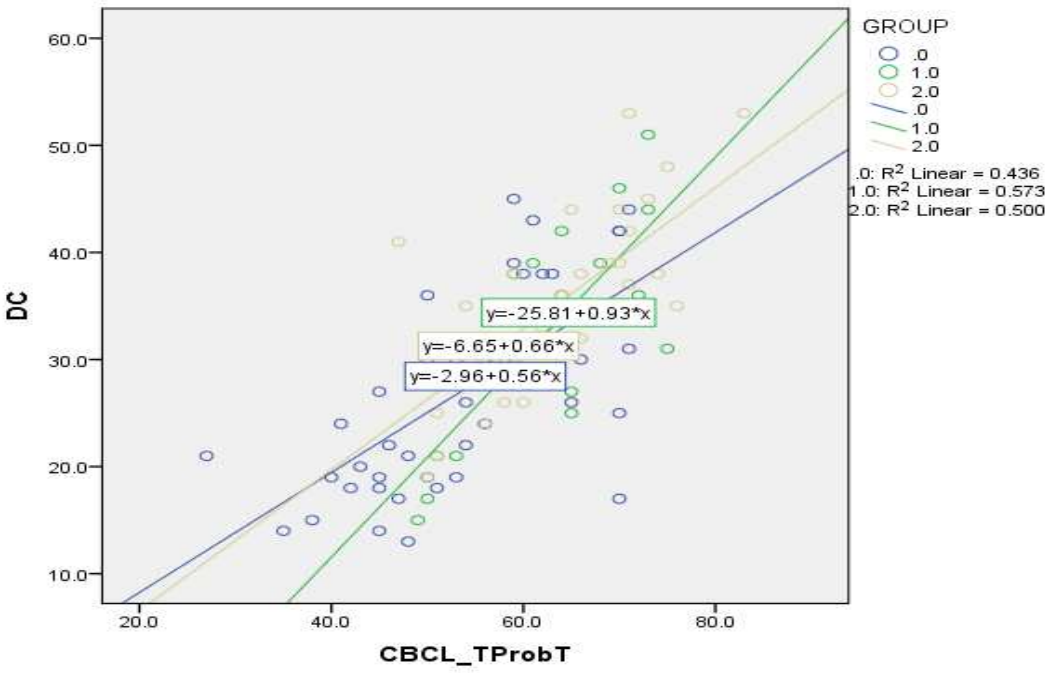

Table 4.4
Pearson Correlation Matrix for Variables in the Secondary Analysis

	Mom age	Child age	Income	SCQ total	CBCL	PD	P-CDI	DC	Total stress	PSPQ Aloof	PSPQ PL	PSPQ Rigid
Mom age	1	.537**	.171	.526*	-.028	.452*	.289	-.022	.247	-.059	.132	-.384
n	22	22	19	22	20	21	21	21	21	21	21	21
Child age	.537**	1	.284	.164	-.201	.060	.088	-.162	-.017	.021	.148	-.261
n	22	22	19	22	20	21	21	21	21	21	21	21
SCQ total	.526*	.164	-.033	1	.539*	.541*	.678**	.462*	.625**	-.559*	-.096	-.356
n	22	22	19	22	20	21	21	21	21	21	21	21
CBCL	-.028	-.201	-.300	.539*	1	.391	.526*	.614**	.585**	.174	.373	-.114
n	20	20	17	20	20	20	20	20	20	20	20	20
PD	.452*	.060	-.041	.541*	.391	1	.735**	.693**	.899**	.423	.644**	.240
n	21	21	18	21	20	21	21	21	21	21	21	21
P-CDI	.289	.088	.145	.678**	.526**	.735**	1	.624**	.876**	.311	.364	.292
n	21	21	18	21	20	21	21	21	21	21	21	21
DC	-.022	-.162	-.343	.462*	.614**	.693**	.624**	.1	.888**	.341	.459*	.225
n	21	21	18	21	20	21	21	21	21	21	21	21
PSI total	.247	-.017	-.108	.625**	.585**	.899**	.876**	.888**	1	.522*	.288	.591**
n	21	21	18	21	20	21	21	21	21	21	21	21

Table 4.4 continued

Pearson Correlation Matrix for Independent and Dependent Variables for the Secondary Analysis

	Mom age	Child age	Income	SCQ total	CBCL	PD	P-CDI	DC	Total stress	PSPQ Aloof	PSPQ PL	PSPQ Rigid
PSPQ	-.215	-.124	-.478*	.155	.603**	.349	.386	.574**	.503	.888**	.740**	.745**
n	21	21	18	21	20	21	21	21	21	21	21	21
PSPQ Aloof	-.059	.021	-.559*	.174	.423	.311	.341	.522*	.452*	1	.557**	.619**
n	21	21	18	21	20	21	21	21	21	21	21	21
PSPQ PL	.132	.148	-.096	.373	.644**	.364	.459*	.288	.412	-.557*	1	.223
n	21	21	18	21	20	21	21	21	21	21	21	21
PSPQ Rigid	-.384	-.261	-.356	-.114	.240	.292	.225	.591**	.433	.619**	.223	1
n	21	21	18	21	20	21	21	21	21	21	21	21

Table 4.5

Child and Maternal Descriptives for the Secondary Sample

N = 20	*Frequency*	*Mean*	*Standard Deviation*	*Range*
Child age (in months)		108.05	40.37	48 - 175
Maternal age (in years)		37.67	25.7	25.7 – 48.2
Maternal Race				
White	*90.9%*			
African American	*9.1%*			
Maternal Education				
Percent with college or above	*35%*			
Maternal Employment				
Percent working outside the home	*32%*			
Total problem behavior score (CBCL T-score)		67.65	9.62	50 - 90

Table 4.5 cont.

Child and Maternal Descriptives for the Secondary Sample

Total score of autism severity (SCQ total score)	17.86	6.33	5 - 31
Total Parenting stress score (PSI-SF total score)	100.10	22.86	42 - 141
Parent-related stress score (PSI-SF PD score)	33.71	7.70	12 - 49
Child-related stress score (PSI-SF DC score)	39.29	9.80	16 - 54
Parent-child interaction stress score (PSI-SF P-CDI score)	27.10	8.26	14 - 44
BAP-Q total score	2.93	.71	1.61 – 4.5
BAP-Q Aloof score	2.93	1.02	1.1 – 4.8
BAP-Q Pragmatic Language score	2.69	.71	1.6 – 4.3
BAP-Q Rigid score	3.15	.84	1.8 – 4.5

Table 4.6

Hypotheses 4 Multivariate Results

Effect	Wilks' Lambda	*F*	*df*	Error *df*	sig.
Intercept	.936	.30	3	13	.826
MomAge	.636	2.49	3	13	.107
CBCL cent.	.877	.61	3	13	.622
SCQ	.675	2.09	3	13	.151
PSPQrig	.549	3.56	3	13	.045

Tests of Between-Subjects Effects

Source	Dependent Variable	*SS*	*df*	*MS*	*F*	sig.
Corrected Model	Log_P-CDI	777.75	4	194.44	4.98	.009
	DC	1280.94	4	320.24	7.80	.001
	PD	664.75	4	166.19	4.91	.010
Intercept	Log_P-CDI	13.68	1	13.68	.35	.563
	DC	16.98	1	16.98	.41	.530
	PD	34.61	1	34.61	1.02	.328
MomAge	Log_P-CDI	12.24	1	12.24	.31	.584

Table 4.6 cont.

Hypotheses 4 Multivariate Results

	DC	.001	1	.001	00	.996
	PD	177.75	1	177.75	5.25	.037
CBCL	Log_P-CDI	16.55	1	16.55	.42	.525
	DC	78.33	1	78.33	1.91	.187
	PD	15.75	1	15.75	.47	.506
SCQ	Log_P-CDI	189.26	1	189.26	4.85	.044
	DC	116.66	1	116.66	2.84	.113
	PD	29.79	1	29.79	.88	.363
PSPQrig	Log_P-CDI	101.15	1	101.15	2.59	.128
	DC	474.97	1	474.97	11.57	.004
	PD	222.41	1	222.41	6.57	.022
Error	Log_P-CDI	585.21	15	39.01		
	DC	616.01	15	41.07		
	PD	508.21	15	33.88		
Total	Log_P-CDI	15997.00	20			

Table 4.6 cont.

Hypotheses 4 Multivariate Results

	DC	32395.00	20
	PD	23685.00	20
Corrected Total	Log_P-CDI	1362.95	19
	DC	1896.95	19
	PD	1172.95	19

Log_P-CDI R^2 = .571 (adj. R^2 = .456), DC R^2 = .675 (adj. R^2 = .589)

PD R^2 = .567 (adj. R^2 = .451)

CHAPTER 5

DISCUSSION

The primary aim of the present study was to extend the current literature by increasing understanding of how specific child and maternal factors impact the various dimensions of parenting stress in three groups of high-risk mothers - mothers of boys diagnosed with idiopathic ASD, mothers of boys diagnosed with ASD and associated FXS, and mothers of boys diagnosed with FXS alone. Additionally, the current study sought to explore the relationship between maternal characteristics of the broader autism phenotype and parenting stress in mothers of children with idiopathic autism spectrum disorder.

Parenting Stress across the Three Groups of Mothers

The first research question posed in this current study involved how levels of reported parent-related, child-related, and parent-child-interaction-related parenting stress vary across these three high-risk groups of mothers. In the existing literature, type of disability has consistently been found an important variable in predicting maternal parenting stress, with mothers of children with ASD often reporting the most significant stress elevations when compared to other groups of mothers. Previous studies, however, provide few comparisons of levels of parenting stress across the three high-risk groups of mothers considered in the current study. Compared to those of mothers in the FXS alone group, parenting stress scores from mothers of boys with IA and AFXS were predicted to

be higher across all three areas of parenting stress assessed in the current study. Results provide partial support for this prediction. While mean stress scores across all three subscales of the PSI-SF were, in fact, higher for mothers of children with IA and mothers of children with AFXS, differences among the groups of mothers only reached statistical significance for one subscale, the Difficult Child scale (PSI-SF DC). For this subscale, however, the only significant difference observed was between the IA and FXS groups, with scores from mothers from the IA group significantly exceeding those of mothers from the FXS alone group.

These results are consistent with previous findings suggesting poorer outcomes for mothers of children with ASD compared to mothers of children with FXS (Abbeduto et al., 2004). In the only previous study to look at parenting stress levels across the three groups examined in the current study, Mankowski (2007) found significantly higher total stress scores on the PSI-SF for both mothers of children with IA and mothers of children with AFXS when compared to mothers of boys with FXS (and no significant difference between the IA and AFXS groups). While current results did not indicate a significant difference in reported stress between mothers from the AFXS and FXS groups, a trend of higher stress ratings for mothers of boys with AFXS compared to mothers of boys with FXS alone was observed across parenting stress domains assessed. It should be noted that the relatively small and unequal sample sizes in the current study may have impacted power to detect group differences. Current results nevertheless suggest that despite the overlapping child and maternal profiles evident in these three groups, something in the experience of parenting a child with ASD makes mothers particularly vulnerable to stress.

In addition to partially replicating previously reported results, current analyses extended previous findings by allowing for a more specific examination of the nature of these observed differences across groups. Specifically, the current investigation examined the experience of different domains of parenting stress across these groups of high-risk mothers, and identified one subscale from the PSI-SF, the Difficult-Child scale, as the primary dimension of parenting stress for which these mothers' experience of stress varies significantly. This is an important finding in that it may help to further clarify the relative contribution of environmental (i.e., child-related) and genetic (i.e., parent-related) factors influencing the excessive levels of parenting stress observed across these groups of high-risk mothers. The Difficult-Child subscale of the PSI-SF assesses for stress that is more directly tied to a mother's perceptions of her child's difficult temperament and challenging behaviors (e.g., by asking about perceptions regarding the child's fussiness, reactivity, and demandingness). That differences in reported stress were significant for this type of stress and not others (with mothers from the IA group reporting significantly higher child-related stress than mothers from the FXS group) likely reflects a couple of key factors. First, and perhaps most obviously, this finding likely reflects the increased reported severity of child problem behaviors in the IA and AFXS groups compared to the FXS alone group. This finding is consistent with previous reports of increased general behavioral complications in children diagnosed with ASD compared to other developmental disabilities (including FXS), and likely influenced the pattern of current child-related stress results. More importantly, however, this finding also serves to highlight the importance of environmental (child-related) factors on the experience of parenting stress in these three groups of mothers.

Variables Contributing to the Experience of Parenting Stress

The second research question posed in this investigation involved determining if we could account for a meaningful amount of variability in maternal parenting stress in the total sample of these three groups of high risk mothers using family, maternal, and child-level variables. Of the variables considered in the present study, only maternal IQ, child ASD symptom severity, and child problem behaviors showed significant correlations with any of the three sub-domains of the PSI-SF. Results of regression analysis only indicated significant main effects for child behavior problems (across all three parenting stress sub-domains) and maternal IQ (only for child-related parenting stress), as well as a significant interaction effect of group and child problem behaviors (only for child-related parenting stress). Results did not indicate a main effect for group or for ASD symptom severity. When looking at the final model, the total variance explained by all included variables was greatest for the child-related parenting stress domain from the PSI-SF, with nearly 58% of variance explained. In contrast, the total variances explained by the final model for the parent-related and parent-child interaction-related domains of the PSI-SF were significantly lower, reaching only approximately 20% and 30%, respectively. These results likely suggest that for all three groups of mothers, other variables not considered in the current study are exerting a considerable influence on these two facets on parenting stress that are less directly tied to child factors. Studies of other high-risk groups of parents have in fact shown a greater influence of some variables not employed in the current study (e.g., parent-reported psychological symptoms) on these two subscales of the PSI-SF when compared to the Difficult Child subscale (Reitman, Currier, & Stickle, 2002). It is therefore possible that inclusion of

these among other variables not considered in the current study would have improved the model's fit for the parent-related and parent-child interaction-related domains.

Consistent with previous findings from the autism literature (e.g., Ekas & Whitman, 2010; Hastings et al., 2005), current results indicate that when considered alongside ASD-related symptomatology, only general child behavior problems make a significant contribution to the prediction of parenting stress. This contribution of child problem behaviors was significant across parenting stress domains and across all groups of mothers. The fact that ASD-related symptoms failed to be a significant predictor of parenting stress across all three groups of mothers is remarkable for several reasons. First, these three groups of mothers can be thought to represent a continuum with regard to both levels of parenting stress and ASD-related symptomatolgy (IA > AFXS > FXS). Given this observed continuum, it would seem feasible to expect that the differences in ASD-related symptomatology observed across the three groups may be contributory to the disparate levels of observed parenting stress. Current results indicate, however, that differences in ASD symptom severity do not tell the whole story, and that more general behavioral challenges may exert a more powerful effect on stress levels across all three groups of mothers. Given the patterns observed in the data, its stands to reason that while increased ASD-related symptomatology may not directly account for increasing levels of parenting stress, that greater ASD-symptom severity is likely associated with higher levels of general problem behaviors, which are in turn impacting stress.

Given precedent in the existing literature, viewing current results as further evidence of a uniquely strong relationship between general child behavior problems (rather than ASD symptom severity) and parenting stress would seem a reasonable

interpretation. However, the potential impact of how these two variables were assessed in the current study is worth mentioning. Specifically, the fact that level of ASD-severity was determined via a clinician-completed measure while severity of general child problem behaviors was assessed by maternal report may have impacted current findings. An inherent limitation present in much of the ASD and FXS parenting stress literature is the frequent use of one informant (typically mothers) to complete all study measures. The potentially transactional relationship between child characteristics and maternal stress could be expected to result at times in an overestimate of the association between maternal stress and child behavior problems. Specifically, a mother experiencing more stress may be more likely to endorse more severe behavioral difficulties for her child, which could in turn impact findings. In the current study, the potential impact of maternal stress on child behavior ratings was in essence removed in the assessment of ASD symptom severity, but not for the assessment of general problem behavior severity. While it is unlikely that the effect of having differing behavioral informants could fully account for the failure of ASD symptoms to make a significant contribution to the prediction of stress in the current study, it is possible that this methodology skewed results to some degree. Specifically, results from previous investigations have noted poor agreement between parent and clinician/teacher behavioral ratings (e.g., Lecavalier et al., 2006) and have indicated that reliance on maternal ratings of child problem behaviors may artificially inflate the relationship between child behaviors and maternal stress.

Taking a closer look at the nature of the significant interaction effect found in the current study, follow-up analysis suggested that the strength of the relationship between child behavior problems and child-related parenting stress varied by group. Specifically,

for mothers in the FXS group, the strength of the relationship between child behavior problems and child-related stress was weaker than that observed in the other two groups of mothers. Though previous studies have indicated a connection between child problem behaviors and parenting stress in mothers of children with ASD (e.g., Davis & Carter, 2008; Hastings et al., 2005), mothers of children with FXS (Bailey, Sideris, Roberts, & Hatton, 2008), and mothers of children with AFXS (Mankowski, 2007), none have allowed for an examination of how the various dimensions of parenting stress may be impacted by child behavior across these three groups. Current results suggest that the impact of child behavioral problems on child-related stress is different for these three groups of mothers, with the impact appearing to be least for mothers of children with FXS alone. As previously noted, problem behavior ratings in the current study were lowest for children of mother's from the FXS alone group. So for these mothers, not only are child problem behaviors fewer than those in the other two groups, but the impact of these general problem behaviors on the experience of child-related parenting stress is less. In contrast, for the IA and AFXS groups, behavioral challenges are both more severe, and exert more of an impact on stress levels.

The third research question examined the relationship between ratings of child behavior problems to child-related parenting stress scores compared to the other domains of parenting stress across these three groups of high-risk mothers. While results did indicate a uniquely strong relationship between CBCL scores and child-related parenting stress for mothers of children with IA and mothers of children with AFXS, this pattern did not hold true for mothers of boys diagnosed with FXS. Interestingly, for mothers of children with FXS , the proportion of variance in parenting stress that was attributable to

child behavioral problems was significantly less, and remarkably similar across child-related, parent-related, and parent-child interaction-related stress domains. This finding provides further evidence of the different role that general child behavior problems play in influencing stress levels for these three groups of mothers.

A critical question that this set of finding raises is that of what factors might be accounting for the differential impact of behavior problems on stress in these three groups of mothers. More specifically, what is it in the experience of raising a child with autism (with or without FXS) that sets it apart from that of raising a child with FXS alone? And how might these differences be connected to how a mother perceives child behavioral challenges? One possibility for consideration in future efforts involves assessing how the process of obtaining a diagnosis of autism (in IA and AFXS) may add to the vulnerability of stress in these groups of mothers compared to mothers of children with FXS alone. For many families, the ASD evaluation process is marked by a series of long and taxing appointments and wait lists that are often one-two years long. In contrast, the diagnosis of FXS is made using a blood test, which is frequently a less time intensive process for families. The potential impact that this notable difference in the diagnostic process may have on perceptions of child behaviors and stress levels across these groups is worth exploring in the future. Another potentially contributing factor worth examining in the future involves the specific patterns of problem behaviors observed in children diagnosed with ASD (with and without FXS). Because only total problem behavior scores were considered in the current study, the possibility that children from these three groups may exhibit different patterns of difficult behaviors could not be assessed. Differing types or patterns of behavioral challenges (e.g., greater

sleep difficulties or self-abusive behaviors) in children diagnosed with ASD could account for the differential influence of general child behavior problems on stress across these three groups.

The Broad Autism Phenotype and Parenting Stress

The fourth question posed as part of the current project involved considering what the differential impact of ASD symptom severity, general child behavior problems, and maternal features of the BAP might be on the experience of parenting stress in mothers of children with ASD. Current results serve to at least partially replicate findings from Ingersoll and Hambrick (2011) which indicated a predictive relationship between maternal BAP characteristics and parenting stress. In the current study, although total scores from the BAP measure failed to make a significant contribution to the prediction of maternal stress, scores from one subscale of the measure that primarily assesses rigid and routine-oriented behaviors did significantly predict both child- and parent-related stress scores. It is worth noting that the behavioral characteristics tapped by the BAP measure in Ingersoll and Hambrick's (2011) study are quite different from those tapped by the measure employed in the current study. In fact, "rigid" behaviors (e.g., insistence on sameness and resistance to changes to one's normal routine) are minimally assessed by the Autism Quotient which was used by Ingersoll and Hambrick. In the previous investigation, the impact of specific features of the BAP was not parsed out as the AQ produces only one composite score. As the core features of the BAP have yet to be firmly established at this time in the literature, further investigation that takes into account the various dimensions of this construct will be necessary in the future to help provide a

better understanding of what specific features of the BAP are associated most with the experience of parenting stress.

As to the current finding indicating a significant effect of ASD-symptom severity rather than general child behavior problems on parenting stress (parent-child interaction-related stress), one potential contributory factor is deserving of consideration. Specifically, the measure used to assess ASD-related symptom severity in this exploratory analysis may have impacted findings. The Social Communication Questionnaire (SCQ) has rarely been used as a measure of autism severity in studies examining factors impacting parenting stress. Though there is precedent for utilizing the SCQ as an index of ASD-related symptom severity (e.g., Charman, Howlin, Berry, & Prince, 2004), very few of the studies contributing to the existing literature on the impact of ASD severity on parenting stress have employed this measure. Hence, it is possible that current results were impacted by choice of this measure. Specifically, because the SCQ was not originally designed to assess symptom severity, and only produces a summary score reflecting the number of core ASD symptoms present for a child, it is possible that the severity construct assessed in the current study differs from that typically assessed in the literature.

Limitations

Although the current study makes a contribution to the understanding of how different dimensions of parenting stress are impacted by maternal and child factors for three high-risk groups of mothers, some limitations present in the current project should be mentioned. Consideration of the following limitations may help provide direction for future research. First, limitations were evident in the current study with regard to

recruitment strategies for both primary and secondary samples of mothers. As is often the case when working with special populations, the samples utilized in the current study were not randomly selected. Instead, this project relied upon families' willingness to volunteer for participation – a strategy which comes with clear risks to external validity. Additionally, recruitment strategies were not consistent across groups of mothers, which may have biased samples to some degree. Specifically, for participants contributing to the extant dataset, mothers of children with ASD were recruited to participate in a study of maternal well-being whereas the mothers of children with FXS were recruited for a study with more general aims. Hence, potential IA recruits with greater stress levels or psychological symptoms may have been less (or potentially more) likely to participate in the study depending on their comfort level in disclosing information related to their own functioning. For newly recruited mothers comprising the secondary sample in the current study, a similar risk was present in that mothers of children with ASD were specifically recruited to participate in a study of maternal stress and parenting experiences. Given this recruitment strategy, mothers with greater levels of stress may have been more likely to volunteer for the study, which may have elevated measured stress levels. This, in fact, appears to be the case when we consider the percentage of mothers reporting clinically significant elevations in stress from the secondary sample (76% of mothers from the secondary sample versus 43% from the primary sample from the extant dataset). Additionally, it is possible that mothers with more symptoms of the BAP may have been *less* likely to participate in the current study, which may have suppressed measured levels of this variable.

A second limitation present in the current study involves the lack of genetic data for mothers, particularly FXS status information for mothers in the ASD only groups. For mothers from both the extant dataset and the secondary sample, although attempts were made to rule out the possibility of co-morbid FXS (e.g., by discussing family history of possible FXS or intellectual disability), a chance still exists that a subset of mothers from the ASD alone group also had FXS. Future studies of these groups of high-risk mothers would benefit from increased efforts to collect genetic screening data from all participants, including mothers of children with ASD. This type of data will be critical not just for ensuring proper group assignment, but also for possibly gaining a better understanding of the complex maternal-gene-behavior interactions impacting these groups of mothers.

Sample size is another potentially limiting factor in the current study. For the extant dataset, relatively small sample sizes, particularly for the AFXS group, may have impacted power to find significant effects. For example, given observed trends, the impact of ASD-related symptomatology on maternal stress should not be ruled out until larger samples are included. Future attempts to disassociate maternal parenting stress in these three groups of mothers would benefit from larger and more even participant groups. With regard to the secondary sample of mothers recruited to examine BAP-parenting stress relationship in mothers of children with ASD, sample size was again a significant limitation. Though meant to be exploratory, the small sample for this secondary analysis may have impacted power to detect significant effects reported in the existing literature.

As was previously noted, the current study was also limited by the use of single informants to gather information on key constructs. Due to the reciprocal nature of maternal and child factors and outcomes, reliance on maternal reports of both child behaviors and stress in the current study may have impacted results. A majority of studies contributing to the existing literature on developmental disability and parenting stress has relied upon maternal reports. With the exception of ASD severity data which was assessed via clinician observation, data for all other constructs in the current study was collected from mothers. Future efforts to better understand the complex relationships among child and maternal factors and parenting stress would benefit from taking a multi-rater approach. Collecting information from multiple informants for both maternal and child behavioral variables (e.g., maternal BAP, child problem behaviors) and for maternal outcome variables (i.e., parenting stress) would serve to minimize the potential risk of finding exaggerated relationships between variables.

Another, and somewhat related, limitation to current study involves its cross-sectional design. Few longitudinal investigations have been conducted in this literature to allow for development of a solid model concerning the direction of causality between child characteristics and maternal stress in these populations. Future studies that employ a multi-rater approach with data collected over multiple time points will permit stronger conclusions regarding causal relationships among maternal and child variables.

Failure to employ other potentially important variables which may impact the experience of maternal parenting stress, such as presence of social support, access to diagnostic and treatment services, and coping style, represent another limiting factor in the current study. As a whole, existing studies have inconsistently controlled for parent,

child, and family variables with established links to maternal parenting stress. While the current study took steps to take into account variables such as maternal age and IQ, child age, and family income, other variables that may exert a critical influence on the experience of maternal parenting stress were not employed. Given evidence from previous research indicating that factors such as coping strategies and social support may at least partially mediate child behavior-stress and maternal BAP-stress relationships (Ingersoll & Hambrick, 2011), future efforts would benefit from inclusion of measures capturing these constructs. Also, as mentioned previously, future studies would benefit from consideration of what specific types of more general (non-ASD-related) behavioral problems (e.g., sleep difficulties, aggression) may be differentially impacting parenting stress in these high-risk groups.

Implications for Practice

The current study provides critical information which can be used to help inform screening and intervention efforts for those working with families of children diagnosed with ASD and FXS. Current findings also have important implications for helping to disentangle the relationships among maternal traits, child factors, and parenting stress in these three relevant clinical groups. Potential implications and applications of current results are discussed below.

First, nearly half of the mothers from the primary sample, and approximately three quarters of mothers from the secondary sample, reported levels of parenting stress exceeding the PSI-SF clinical cut-off score. These significant elevations in parenting stress were observed across all three groups of mothers. Due to the host of known negative outcomes associated with high levels of parenting stress, including increased

maladaptive parenting behaviors, greater incidence of maternal psychopathology, poorer engagement with services, and decreased benefit from intervention services for children (Osborne et al., 2008; Robbins et al., 1991), current stress findings cannot easily be ignored. As providers of primary care and specialty care and intervention are making increasing efforts to apply family-centered principles to their practices, meaningful assessment and management of parenting stress should be a critical component. For mothers of children in these high risk groups in particular, assessing and addressing high levels of parenting stress may be key for improving child, parent, and family outcomes. Although a number of barriers are currently present which hinder professionals working with these families from incorporating parent well-being into treatment plans (e.g., time limitations, lack of brief assessment tools, reimbursement issues), accurate screening and management of parenting stress in these and other high-risk clinical populations seems an important goal.

An additional implication of current results for intervention involves the type of child variable identified as contributing most to parenting stress across all three groups of mothers. While observed trends likely indicate some effect of ASD-symptomatology on the experience of stress, results more clearly indicate a critical influence of more general child behavioral problems on maternal stress levels. The implications of this literature for intervention are potentially positive in that the types of behaviors identified as most stressful for mothers are also the types of behaviors generally thought to be most amenable to intervention. While, certainly, efforts to improve core deficits of autism remain an essential component of interventions for children with ASD and their families (with or without fragile X); results would indicate that efforts to decrease problem-

behaviors unrelated to ASD are also likely essential for improving outcomes for families. Given the noted reciprocal relationships between maternal stress, child behavior, and even child responsiveness to intervention (e.g., Osborne et al., 2008), child and family-based treatment programs incorporating treatment of general maladaptive behaviors would likely enhance benefits for both mothers and their children.

Current results related to the BAP-parenting stress relationship may also have important implications for professionals working with families of children diagnosed with ASD. Present findings point to certain subclinical characteristics of ASD as possibly predisposing mothers to increased levels of parenting stress. While additional research will be necessary to fully parse out this effect and to determine what mediating/moderating variables may also be at work, current findings suggest that professionals should be aware of how parental characteristics of the BAP may influence the experience of stress. Given the very early stages of research in this area, however, it should be noted that results may ultimately suggest both liabilities *and* benefits associated with expression of the BAP in parenting a child diagnosed with ASD. While certain characteristics of the BAP (such a rigid tendencies) may predispose some parents to increased stress, other characteristics may serve as protective factors by increasing insight and understanding into child behavioral characteristics. Specifically, it is possible that a parent's overlapping traits with her child on the spectrum may provide much needed perspective for understanding the child's social, communication, and behavioral challenges, and hence serve to improve coping with diagnosis-related stress. Keeping a strengths-based perspective in exploring the BAP-parenting stress relationship will be an

important goal for future efforts seeking to improve outcomes for families impacted by ASD.

REFERENCES

Abbeduto, L., Seltzer, M.M., Shattuck, P., Krauss, M.W., Orsmond, G., & Murphy. M. M. (2004). Psychological well-being and coping in mothers of youths with autism, Down syndrome, or fragile X syndrome. *American Journal on Mental Retardation, 109,* 237-254.

Abidin, R. R. (1995). *Parenting stress index professional manual (3rd ed.).* Odessa, FL: Psychological Assessment Resources, Inc.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (5th ed.).*

Archer, L., & Szatmari, P. (1991). Assessment and treatment of food aversion in a four-year-old boy: A multidimensional approach. *The Canadian Journal of Psychiatry, 35(6),* 501-505.

Bailey, D. B., Hatton, D. H., Skinner, M., & Mesibov, G. (2001). Autistic behavior, FMR1 protein, and developmental trajectories in young males with fragile X syndrome. *Journal of Autism and Developmental Disorders, 31,* 165-174.

Bailey DB, Raspa M, Bishop E, Holiday DB. No change in age of diagnosis for fragile X syndrome: Findings from a national parent survey. Pediatrics. 2009; 124:527–533.

Bailey, D.B., Sideris, J., Roberts. J., Hatton, D. (2008). Child and genetic variables associated with maternal adaptation to fragile X syndrome: A multidimensional analysis. *American Journal of Medical Genetics, 146(A),* 720-729.

Bauminger, N., Solomon, M., & Rogers, S.J. (2010). Externalizing and internalizing behaviors in autism spectrum disorders. Autism Research. 3(3):101-12.

Bebko, J.M., Konstantareas, M.M., & Springer, J. (1987). Parent and professional evaluations of family stress associated with characteristics of autism. *Journal of Autism and Developmental Disorders, 17,* 565-576.

Belsky, J. (1984). The determinants of parenting: A process model. *Child Development, 55(1),* 83-96.

Bouma, R., & Schweitzer, R. (1990). The impact of chronic childhood illness on family stress: A comparison between autism and cystic fibrosis. *Journal of Clinical Psychology, 46,* 722-730

Bowen, M. (1978). *Family therapy in clinical practice.* New York: Jason Aronson.fdc

Bravo, K. (2006). Severity of autism and parental stress: The mediating role of family environment. Dissertation Abstracts International: Section A: The Humanities and Social Sciences, 66 (10), 3.821.

Brereton, A.V., Tonge. B.J., & Einfeld, S.E. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36,* 863 – 870.

Centers for Disease Control and Prevention. (2009). MMWR weekly: Prevalence of autism spectrum disorders—Autism and Developmental Disabilities Monitoring Network, United States, 2006. Retrieved from http://www.cdc.gov/mmwr/preview/mmwrhtml/ss5810a1.htm

Constantino, J.N., Zhang, Y., Frazier, T., Abbacchi, A.M., Law, P. (2010). Sibling recurrence and the genetic epidemiology of autism. *The American Journal of Psychiatry, 167(11),* 1349-1356.

Creasey, G., & Reese, M. (1996). Mothers' and fathers' perceptions of parenting hassles: Associations with psychological symptoms, nonparenting hassles, and child behavior problems. *Journal of Applied Developmental Psychology, 17,* 393–406.

Crnic, K. A., Gaze, C., & Hoffman, C. (2005). Cumulative parenting stress across the Preschool period: Relations to maternal parenting and child behaviour at age five. *Infant and Child Development, 14,* 117-132.

Davis, N., & Carter, A. S. (2008). Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism and Developmental Disabilities, 38, 1278*-1291.

Deater-Deckard, K. (2006). Parenting stress and child adjustment: Some old hypotheses and new questions. *Clinical Psychology Science and Practice, 5(3),* 314-332.

Deater-Deckard, K., & Scarr, S. (1996). Parenting stress among dual-earner mothers and fathers: Are there gender differences? *Journal of Family Psychology, 10,* 45-59.

Delong, G.R., & Dwyer, J.T. (1988). Correlation of family history with specific autistic subgroups: Asperger's syndrome and bipolar affective disease. *Journal of Autism and Developmental Disorders, 18,* 593-600.

Dumas, J.E., Wolf, L.C., Fisman, S.N., & Culligan, A. (1991). Parenting Stress, child behavior problems, and dysphoria in parents of children with autism, Down syndrome, behavior disorders, and normal development. *Exceptionality, 2,* 97-110.

Ekas, N., & Whitman, T.L. (2010). Autism symptom topography and maternal socioemotional functioning. *American Journal on Intellectual and Developmental Disabilities, 115(3),* 234-249.

Eisenhower, A.S., Baker, B.L., & Blacher, J. (2005). Preschool children with intellectual disability: Syndrome specificity, behavioural problems, and maternal well-being. *Journal of Intellectual Disability Research, 49,* 657-671.

Franke, P., Leboyer, M., Gansicke, M., Weiffenbach, O., Biancalana, V., Cornillet-Lefebre, P., et al. (1998). Genotype-phenotype relationship in female carriers of the premutation and full mutation of FMR-1. *Psychiatry Research, 80,* 113-127.

Freeman, N.L., Perry, A., & Factor, D.C. (1991). Child behaviors as stressors; Replicating and extending the use of the CARS as a measure of stress; A research note. *Journal of Child Psychology and Psychiatry, 32(6),* 1025-1030.

Gelfand, D. M., Teti, D. M., & Fox, C. R. (1992). Sources of parenting stress for depressed and nondepressed mothers of infants. *Journal of Clinical Child Psychology, 21,* 262-272.

Gilliam, J. E. (1995). *Gilliam Autism Rating Scale.* Austin, TX: Pro-Ed.

Goldstein, D.S. (1995). Stress as a scientific idea: A homeostatic theory of stress and distress. *Homeostasis in Health and Disease, 36,* 177-215.

Gray, D. E., & Holden, W. J. (1992). Psycho-social well-being among parents of children with autism. *Australia and New Zealand Journal of Developmental Disabilities, 18,* 83–93.

Griffith, G.M., Hastings, R.P., Nash, S., & Hill, C. (2010). Using matched groups to explore child behavior problems and maternal well-being in children with Down Syndrome and autism. *Journal of Autism and Developmental Disorders, 40,* 610-619.

Hall, S.S., Lightbody, A.A., & Reiss, A.L. (2008). Compulsive, self-injurious, and autistic behavior in children and adolescents with fragile X syndrome. *American Journal on Mental Retardation, 113,* 44–53

Hartley, S.L., Seltzer, M.M., Hong, J., Greenberg, J.S., Almeida, D., Coe, C., & Abbeduto, L. (2012). Cortisol response to behavior problems in FMR1 premutation mothers of adolescents and adults with fragile X syndrome: A diathesis-stress model. *International Journal of Behavioral Development, 36,* 53-61.

Hastings, R.P. (2003). Parental stress and behavior problems of children with developmental disability. *Journal of Intellectual and Developmental Disability, 27,* 149-160.

Hastings, R.P. & Johnson, E. (2001) Stress in UK families conducting intensive home-based behavioral intervention for their young child with autism. *Journal of Autism and Developmental Disorders, 31,* 327-336.

Hastings, R.P., Kovshoff, H., Ward, N.J., Espinosa, F.D., Brown, T., & Remington, B. (2005). Systems analysis of stress and positive perceptions in mothers and fathers of pre-school children with autism. *Journal of Autism and developmental disorders, 35,* 635-644.

Hatton, D.D., Hooper, S.R., Bailey, D.B., Skinner, M.L., Sullivan, K.M., & Wheeler, A. (2002). Problem behavior in boys with fragile X syndrome. *American Journal of Medical Genetics, 108,* 105-116.

Herring S, Gray KM, Taffe J, Tonge B, et al. (2006). Behaviour and emotional problems in toddlers with pervasive developmental disorders and developmental delay: Associations with parental mental health and family functioning. *Journal of Intellectual Disability Research 50*(12):874-882.

Hoffman, C.D., Sweeney, D.P., Lopez-Wagner, M.C., Hodge, D., Nam, C.Y., & Botts, B.H. (2008). Children with autism: Sleep problems and mothers' stress. *Focus on Autism and Other Developmental Disabilities, 23(3),* 155-165.

Holden, E. W., & Banez, G. A. (1996). Child abuse potential and parenting stress within maltreating families. *Journal of Family Violence, 11,* 1–12.

Ingersoll, B., & Hambrick, D.Z. (2011). The relationship between the broader autism phenotype, child severity, and stress and depression in parents of children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 5,* 337-344.

Johnston, C., Hessl, D., Blasey, C, Eliez, S., Erba, H., Dyer-Friedman, J., et al. (2003). Factors associated with parenting stress in mothers of children with fragile X syndrome. *Developmental and Behavioral Pediatrics, 24,* 367-275.

Kasari, C., & Sigman, M. (1997). Linking parental perceptions to interactions in young children with autism. *Journal of Autism and Developmental Disorders, 27,* 39-57.

Kaufmann, W.E., Cortell, R., Kau, A.S.M., Bukelis, I., Tierney, E., Gray, R.M., & Stanard, P (2004). Autism spectrum disorder in fragile X syndrome: Communication, social interaction, and specific behaviors. *American Journal of Medical Genetics, 129,* 225–234.

Koegel, R.L., Schreibman, L., Loos, L.M., Dirlich-Wilheim, H., Dunlap, G., Robbins,

F.R., & Plienis, A.J. (1992). Consistent stress profiles in mothers of children with autism. *Journal of Autism and Developmental Disorders, 22,* 205-216.

Konstantareas, M.M., & Homatidis, S. (1989). Assessing child symptom severity and stress in parents of autistic children. *Journal of Child Psychiatry, 30,* 459-470.

Konstantareas, M.M., & Papageorgiou, V. (2006). Effects of temperament, symptom severity and level of functioning on maternal stress in Greek children and youth with ASD. *Autism, 10,* 593-607.

Kotchick, B.A., Dorsey, S., Heller, L. (2005). Predictors of parenting among African American single mothers: Personal and contextual factors. *Journal of Marriage and Family, 67 (2),* 448-460.

Krug, D. A., Arick, J., & Almond, P. (1993). *Autism Screening Instrument for Educational Planning*. Austin, TX: Pro-Ed.

Lecavalier, L., Leone, S. & Wiltz, J. (2006). The impact of behaviour problems on caregiver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research, 50,* 172-183.

Leigh, B., & Milgrom, J. (2008). Risk factors for antenatal depression, postnatal depression and parenting stress. *BMC Psychiatry 8,* 24.

Lewis, P., Abbeduto, L., Murphy, M., Richmond, E., Giles, N., Bruno, L., Schroeder, S., Anderson, J., & Orsmond, G. (2006). Psychological well-being of mothers of youth with fragile x syndrome: syndrome specificity and within-syndrome variability. *Journal of Intellectual Disability Research, 50,* 894-904.

Mankowski, J.B. (2007). Mood, anxiety, and stress in mothers of children with Fragile X syndrome, autism, and Fragile X syndrome and autism. *Dissertation Abstract International,* (UMI No. AAT 3444113) Retrieved June 10, 2011, from Dissertations and Theses database.

Mash, E.J., & Johnston, C. (1983). Parental perceptions of child behavior problems, parenting self-esteem, and mothers' reported stress in younger and older hyperactive and normal children. *Journal of Consulting and Clinical Psychology, 51,* 86-99.

McCarthy, A., Cuskelly, M., van Kraayenoord, C.E., & Cohen, J. (2005) Predictors of Stress in mothers and fathers of children with fragile X syndrome. *Research in Developmental Disabilities.*

Micali, N., Chakrabarti, S., & Fombonne, E. (2004). The broad autism phenotype: Findings from an epidemiological survey. *Autism, 8,* 21-37.

Milgram, N.A., & Atzil, M. (1988). Parenting stress in raising autistic children. *Journal of Autism and Developmental Disorders, 18,* 415-424.

Milgrom, J. & McCloud, P. I. (1996). Parenting stress and postnatal depression. *Stress Medicine, 12,* 177–186.

Noh, S., Dumas, J. E., Wolf, L. C., & Fisman, S. N. (1989). Delineating sources of stress in parents of exceptional children. *Family Relations, 38,* 456-461.

Osborne, L.A., McHugh, L., Saunders, J., & Reed, P. (2008). Parenting stress reduces the effectiveness of early teaching interventions for autistic spectrum disorders. *Journal of Autism and Developmental Disorders, 3,* 1092-1103.

Osborne, L.A., & Reed, P. (2009). The relationship between parenting stress and behavior problems of children with autistic spectrum disorders. *Exceptional Children, 76.*

Ozonoff, S., & Rogers, S. (2003). From Kanner to the millennium. In S. Ozonoff, S. J. Rogers, & R. L. Hendren (Eds.), *Autism spectrum disorders: A research review for practioners* (pp. 3-33). Washington, DC: American Psychiatric Press.

Phetrasuwan, S., & Miles, M.S. (2009). Parenting stress in mothers of children with autism spectrum disorders. Journal for Specialists in Pediatric Nursing, 14(3), 157-165.

Piven, J. (2001). The broad autism phenotype: A complementary strategy for molecular genetic studies of autism. *American Journal of Medical Genetics, 105,* 34-35.

Piven, J., Palmer, P., Jacobi, D., Childress, D., & Arndt, S. (1997b). Broader autism phenotype: Evidence from a family history study of multiple-incidence autism families. *American Journal of Psychiatry, 154* (2) 185-190.

Reitman, D., Currier, R.O., & Stickle, T.R. (2002). A critical evaluation of the PSI-SF in a Head Start population. Journal of Clinical Child and Adolescent Psychology, 31(3), 384-392.

Richardson, L.L. (2010). The relationship of repetitive behavior and sensory behavior to parenting stress in mothers of boys with autism and mothers of boys with fragile X syndrome. Theses and Dissertations. Paper 250, http://scholarcommons.sc.edu/etd/250

Robbins, F. R., Dunlap, G., & Plienis, A. J. (1991). Family characteristics, family training, and the progress of young children with autism. *Journal of Early Intervention, 15,* 173–184.

Roberts, J., Bailey, D., Mankowski, J., Ford, A., Sideris, J., Weisenfeld, L., & Golden, R., (2009). Mood and anxiety disorders in females with the FMRI premutation. *American Journal of Medical Genetics, Part B, Neuropsychiatric Genetics: The*

Official Publication of the International Society of Psychiatric Genetics, 150B(1), 130-139.

Rogers, A. Y. (1993). The assessment of variables related to the parenting behaviors of mothers with young children. *Child and Youth Services Review, 15,* 385–402.

Sanders, J.L., & Morgan, S.B. (1997). Family stress and management as perceived by parents of children with autism or Down Syndrome: Implications for intervention. *Child and Family Behavior Therapy, 19,* 15-32.

Schopler, E., Reichler, R., & Renner, B. (1988). *The childhood autism rating scale (CARS).* Los Angeles: Western Psychological Services.

Seltzer, M., Barker, E.T., Greenberg, J.S., Hong, J., Coe, C., & Almeida, D. (2011). Differential sensitivity to life stress in FMR1 premutation carrier mothers of children with fragile X syndrome. *Health Psychology.*

Sharpley, C.F, Bitsika, V., & Efremidis, B. (1997). Influence of gender, parental health, and perceived expertise of assistance upon stress, anxiety, and depression among parents of children with autism. *Journal of Intellectual & Developmental Disability, 22,* 19-29.

Sivberg, B. (2002). Family system and coping behaviors: A comparison between parents of children with autistic spectrum disorders and parents with non-autistic children. *International Journal of Research and Practice, 6,* 397-409.

Smalley, S.L., McCracken J., & Tanguay, P. (1995) Autism, affective disorders, and Social phobia. *American Journal of Medical Genetics (Neuropsychiatric Genetics) 60,* 19-26.

Stoddart, K. P. (2003). Reported stress, personality, and mental health in parents of children with pervasive developmental disorders. *Dissertation Abstracts International: Section A: Humanities and Social Sciences, 64*(4-A), 1411.

Tobing, L.E., & Glenwick, D.S. (2002). Relation of the Childhood Autism Rating Scale – Parent version to diagnosis, stress, and age. *Research in Developmental Disabilities, 23,* 211-223.

Tomanik, S., Harris, G.E., & Hawkins, J. (2004). The relationship between behaviours exhibited by children with autism and maternal stress. *Journal of Intellectual and Developmental Disability, 29(1),* 16-26.

von Gontard, A., Backes, M., Laufersweiler-Plass, C., Wendland, C., Lehmkuhl, G., Zerres, K., & Rudnik-Schöneborn, S. (2002). Psychopathology and familial stress—comparison of boys with fragile X syndrome and spinal muscular atrophy. *Journal of Child Psychology and Psychiatry, 43,* 947-957.

Wheeler, A.C., Hatton, D.D., Reichardt, A., & Bailey, D.B. (in press). Correlates of Maternal behaviors in mothers of children with fragile X syndrome. *Journal of Intellectual Disability Research.*

Wolf, L. C., Noh, S., Fisman, S. N., & Speechley, M. (1989). Brief report: Psychological effects of parenting stress on parents of autistic children. *Journal of Autism and Developmental Disorders, 19,* 157–166.

Wolff, S., Narayan, S., & Moyes, B. (1988). Personality characteristics of parents of autistic children. *Journal of Psychology and Psychiatry, 29,* 143-153.

Yau, M. K. S. and Li-Tsang, W. P. C. (1999). Adjustment and adaptation in parents of children with developmental disability in two-parent families: a review of the characteristics and attributes. *The British Journal of Developmental Disability, 45,* 38-51.