DIFFERENCES BETWEEN CHILDREN WITH ASDs, THEIR NON-AFFECTED SIBLINGS, AND THE CBCL NORMATIVE SAMPLE BASED ON CBCL PROFILE SCORES

A Dissertation Presented to the
Faculty of the College of Education
University of Houston

In Partial Fulfillment
of the Requirements for the Degree

Doctor of Philosophy

by

Dena G. Buchalter

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Abstract

A sparse amount of research exists comparing individuals with autism spectrum disorders (ASDs) and their non-affected siblings regarding behaviors on a broadband measure, especially when the impact of age and IQ on the behavior of siblings is considered. According to social cognitive learning theory, non-affected siblings may be expected to be affected by the behavior of their siblings with ASD, although the directionality of that influence is unknown. This study sought to determine whether the risk of developing behavior problems differs for non-affected siblings compared to the normative population, as indicated by scores on a broad band measure of behavioral disturbance, the Parent Report Form of the Child Behavior Checklist (CBCL). Social cognitive learning theory posits that the non-affected sibling may either imitate the inappropriate behaviors of their diagnosed siblings, earning higher CBCL scores, or through observational learning they may learn to inhibit or avoid inappropriate behaviors by observing the unwelcome consequences of these behaviors applied by their parents. The study also investigated the impact of age and a proxy for IQ (the Vineland Adaptive Behavior Scales-II or Vineland-II) on the CBCL scores of the siblings with and without ASD.

The participants were individuals with ASDs (probands) from simplex families (i.e., only one child diagnosed with an ASD and without either parent diagnosed with an ASD), their non-affected siblings (ages 4-years through 17-years and 11-months), and a

third group, which consisted of the normative sample for the CBCL for ages 1.5-5, 6-11, and 12-18. Archival data were used from the Simons Foundation Autism Research Initiative (SFARI) database. Multiple Bonferroni-corrected, two-tailed t-tests with an alpha of .001 indicated that the sibling group was significantly below the normative population on the CBCL on all 9 scales examined on the age 1.5-5 version, 7 of 9 scales for the 6-11-years age group, and 8 of 9 scales for the 12-18-year age group (all significant *p* values were <.001). This suggests that the non-affected siblings may have learned vicariously to inhibit inappropriate behaviors by observing the unwelcome consequences of the probands' behavior and that they demonstrated significantly fewer inappropriate behaviors overall than did the CBCL normative sample. Consistent with prior research, the probands had significantly higher scores overall than both the non-affected siblings and the normative CBCL sample (significant *p* values were all <.001); however, on the Anxious-Depressed scale for the pre-school age group, the proband group was not significantly higher than the normative group, but the proband group was significantly higher than the non-affected sibling group.

MANCOVAs indicated that the probands also had significantly higher overall CBCL profiles than the non-affected siblings on those scales determined to be most relevant based on the research when controlling for age and adaptive behavior as a proxy for IQ. Significant differences emerged on the CBCL for the pre-school age group on the Total Problems, Anxious-Depressed, Withdrawn, Sleep Problems, Attention Problems, Aggressive Behavior, and Pervasive Developmental Problems scales ($F_{7, 721}= 23.51$, $p < .001$) and the Internalizing and Externalizing scales ($F_{2, 732}= 87.635$, $p < .001$). Significant

differences emerged on the CBCL for children and adolescents (6-18-years old) on the Anxious-Depressed, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behavior, and Total Problems scales ($F_{7, 3034}$= 681.900, $p < .001$) and the Internalizing and Externalizing scales ($F_{2, 3046}$= 130.718, $p < .001$). Limitations, including the unusual nature of the SFARI sample and the use of the Vineland-II as a proxy for IQ, and directions for future research are discussed.

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CHAPTER I

Differences Between Children With ASDs, their non-ASD Siblings, and the CBCL Normative Sample Based on CBCL Profile Scores

Autism Spectrum Disorders (ASDs) are defined by deficits in three categories: socialization, communication, and repetitive or unusual behaviors, which can range from mild to severe (American Psychiatric Association, 2000; CDC, 2007; Levy, Mandell, & Schultz, 2009). ASDs are neurodevelopmental disorders that include autistic disorder, Asperger's syndrome, and pervasive developmental disorder-not otherwise specified (CDC, 2007). In the social domain, symptoms include impaired use of nonverbal behaviors, such as eye contact, facial expression, and gestures, to facilitate social interaction, failure to form age-appropriate peer relationships, minimal to no seeking shared enjoyment or interests with other people, and limited social-emotional reciprocity. Communication deficits entail delay or absence of speech, difficulty with conversational reciprocity, idiosyncratic or repetitive language, and imitation and pretend play deficits. In the category of repetitive or unusual behaviors, there are frequently encompassing, unusual interests, inflexible adherence to nonfunctional routines, stereotyped body movements, and preoccupation with parts or sensory qualities of objects (Ozonoff, Goodlin-Jones, & Solomon, 2005).

Generally diagnosis is made after evaluating a child through observations, developmental history, parent interview, psychological testing, and speech and language assessment. Prevalence of autism has changed since the 1960s, which is when only autism was accounted for in prevalence rates. Twenty years later, rates of ASDs ranged

from 5 to 72 cases per 10,000 children in the US and Europe; this estimate was based on small sample sizes, case confirmation strategies, and screening procedures at that point in time, so the numbers were overestimated. Prevalence of autistic disorder was 10-20 per 10,000 children (Levy et al., 2009), and prevalence of ASDs was 2-6 per 1,000 people, or 1 in 166 children are diagnosed with an ASD when the Centers for Disease Control collected data in 2002 from 10 sites (CDC, 2007). Of the 10 sites who collected data in 2002, nine indicated an increase in ASD prevalence (range: 27%--95% increase; $p<0.01$) based on data collection in 2006, with increases among males in all sites and among females in four sites (CDC, 2009). In 2006, the prevalence rate of ASDs was estimated to be close to 1 in 110 children, so the average prevalence of ASDs identified in children at 8-years old increased 57% in those 10 surveillance sites who noted a change in prevalence from 2002. Although improved ascertainment accounted for some of the increased prevalence rates, a true increase in the risk for children to develop ASD symptoms cannot be dismissed (CDC, 2009).

In schools the number of children who have been identified with ASDs has increased, which may be due to changes in administrative classifications in special education and the reclassification of children from a different category to autism. Autism was added in the 1990s as a separate category for special education. Change in the wording of policy and practice may be the reason for the rise in prevalence rates (CDC, 2007; Levy et al., 2009). In 2005, 9% of special education placements were children with mental retardation (545,492 children), and 3% were children with autism (193,637 children) (IDEA, 2005). Considerable variation within a small area in regards to prevalence may be related to local healthcare and education resources. Additionally,

over-diagnosis may be occurring as a means to gain intensive services traditionally reserved for children with autism within the school setting (Levy et al., 2009).

Assessment: screening and diagnosis

In regards to age of onset, symptoms are usually present by 3-years old, but noticeable signs can be present in infancy (Charman et al., 1997; Levy et al., 2009). ASDs have specific diagnostic criteria, ages of symptoms recognition, associated medical and developmental features, standard effective treatments, and usual courses of development. Early detection and diagnosis allows for the most optimal results for that individual child. Screening measures have been developed in recent years to identify children who may be at risk for ASDs to lead to earlier diagnosis and intervention. Some screening measures are written parent questionnaires, such as the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003), based on the Autism Diagnostic Interview-Revised (ADI-R; Rutter, LeCouteur, & Lord, 2003), and Modified Checklist for Autism in Toddlers (M-CHAT; Robins, Fein, Barton, & Green, 2001), both of which should not be used for diagnostic purposes due the levels of false positives and negatives (depending on set levels of sensitivity and specificity) (Eaves, Wingert, & Ho, 2006).

Practice parameters established by the American Academy of Neurology (Filipek et al., 2000), the American Academy of Child and Adolescent Psychiatry (Volkmar, Cook, Pomeroy, Realmuto, & Tanguay, 1999), and a multidisciplinary consensus panel (Filipek et al., 1999) recommend two levels of screening and evaluation. Level 1 screening consists of routine developmental surveillance by general providers for young children, and Level 2 evaluation consists of a comprehensive diagnostic assessment by

clinicians for children who fail the initial screening. Assessment and diagnosis also needs to be viewed within a developmental framework, as the appropriate milestones differ across the lifespan (Ozonoff, Goodlin-Jones, & Solomon, 2005).

In regards to Level 2 assessment, the gold standard consists of the parent interview Autism Diagnostic Interview, Revised (ADI-R; Rutter, LeCouteur, & Lord, 2003), and the performance-based Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2002). The ADI-R, which is strongly related to the DSM-IV-TR criteria, is a semi-structured interview that takes 90 minutes to three hours to administer, depending on whether the long or short version is given. The ADI-R covers the individual's current behavior and developmental history. Its major limitations are lack of sensitivity to differences among children who are below the mental age of 20 months or who have IQs below 20 and to milder cases of ASD before the age of 3-years old, and the instrument is laborious to administer, taking more time than most practitioners can allow for assessment (Ozonoff et al., 2005). The ADOS, an observation measure, is a semi-structured interactive assessment of ASD symptoms with four separate modules, scored according to language and developmental level, allowing for flexibility in administration to a wide range of individuals. The ADOS has an empirically designed cutoff score for autistic disorder and another for the broader ASD category (Ozonoff, Goodlin-Jones, & Solomon, 2005).

In regards to diagnostic questionnaires, the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003), is a parent questionnaire based on the ADI-R and includes many of the same questions as on the ADI-R but in a "yes/no" format. The SCQ has high sensitivity and specificity in individuals with autism and other developmental

disorders. Another questionnaire, which is for parents and teachers, is the Gilliam Autism Ratings Scale-2nd edition (GARS-2; Gilliam, 2006). It contains four subscales that load onto an Autism Quotient, which measures the likelihood that a child has autism (Ozonoff et al., 2005). The GARS-2 is easy to use and it is linked to the DSM-IV-TR; however, in an earlier utility study using the GARS, by South et al. (2002), a high false negative rate was found using the GARS in comparison to the ADI-R and ADOS, and utility problems remain an issue in the GARS-2 partly because some individual items load on multiple factors, so results should be interpreted with caution (Ozonoff et al., 2005; Pandolfi, Magyar, & Dill, 2010). Lastly, the Autism Spectrum Screening Questionnaire (Ehlers, Gillberg, & Wing, 1999) is an informant checklist to assess symptoms of Asperger's disorder and high functioning autism and has good internal consistency and validity. There are specific measures that address Asperger's disorders alone, but the focus of this research is on the spectrum (Ozonoff et al., 2005).

In regards to observation instruments, besides the ADOS, another observation instrument that is widely used is the structured Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1988). The total score highly correlates with the ADI-R but overidentifies autism relative to the ADI-R because it sometimes classifies children with mental retardation as having autism. The CARS was developed before the DSM-IV-TR, so it does not measure some constructs now considered important in diagnosing autism (Ozonoff et al., 2005).

Throughout development, children with ASDs might develop symptoms and behaviors that impede their daily functioning, which can consist of factors such as changes in sleep, appetite, mood, anxiety, activity level, anger management, and

aggression. ASDs are commonly associated with psychosocial impairments and maladaptive behaviors, which may make it difficult to parse out any other psychiatric symptoms. Self reports and interviews tend to be less useful with this population because of possible lack of insight and the need to report symptoms; however, when significant behavioral problems occur outside of typical ASD symptoms, the individual's psychosocial functioning should be evaluated. The Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001), a broadband instrument, is frequently used to identify child behavioral and mental health problems but has not been used as widely in the ASD population. A broadband instrument is one that measures a broad range of behaviors, whereas a narrowband instrument measures a specific area of behaviors (Ozonoff et al., 2005). To date, five published studies that have examined CBCL profiles of children with ASDs were able to be identified (Bolte, Dickhut, & Poustka, 1999; Duarte, Bordin, de Oliveira, & Bird, 2003; Pandolfi, Magyar, & Dill, 2009; Rescorla, 1988; Sikora, Hall, Hartley, Gerrard-Morris, & Cagle, 2008).

CBCL and ASD population

The first study to examine the CBCL within the ASD population performed a cluster analysis of CBCL profiles of a sample of 204 boys who were 3 to 5-years old; 79 participants were "autistic" or "autistic-like," 82 had "reactive" (more typical emotional/behavior disorders of childhood), and 43 children were in the "other" category, which included those with fairly mild impairments who received less common clinical diagnoses (Rescorla, 1988). The archival data came from clinic patients' records in the 1960's and 70's; parent report was not available. The results of the factor analysis were

consistent with previous literature using the CBCL. There were patterns of results across 2, 3, 4, 5, and 6-cluster solutions.

From the cluster analysis, the first three factors consisted of an Autistic/Bizarre factor, an acting-out Aggressive/Destructive factor, and an Anxious/Depressed emotional factor. The Autistic/Bizzare factor does not usually appear in factor analyses of the CBCL, which seems to be a function of the large number of autistic children in Rescorla's (1998) study who exhibited symptoms with a large frequency that loaded on this factor. Other factors that emerged were Attention deficit/Hyperactivity, Immature Habits (e.g., toilet-training items), Autoerotic Behavior (masturbation and thumb-sucking), Fearful/Dependent behavior, and Antisocial/Aggressive behavior. Rescorla's research identified high and low functioning boys with autism based on their symptom profiles, and with the increased number of clusters, children on the spectrum were further differentiated, primarily on the basis of maturity of functioning and the presence of manifest anxiety. In sum, when the eight-factor (factors derived from analysis of the 73 Achenbach CBCL symptom items) CBCL symptom profiles of preschool children manifesting a wide range of psychiatric and developmental problems were submitted to cluster analysis, autistic males were differentiated easily from the sample of other disturbed preschool children (Rescorla, 1988).

Another study that used the CBCL to analyze behavior problems to determine a general problem pattern in ASDs used the German form of the CBCL and conducted a MANCOVA with sex, age, and IQ as covariate predictors for all 8 CBCL syndrome scales (Bolte, et al., 1999). Of the 77 autistic individuals, diagnosed by the German versions of the ADOS and ADI-R, in Bolte et al.'s (1999) study, 38 had estimated IQ

scores due to the child's restlessness, withdrawn behavior, and noncompliance. Estimated IQs were based on scores from the German version of the Vineland Adaptive Behavior Scale and school achievement. The mean IQ was 61.3 with a SD of 25.9.

In comparison to the normative sample, the autistic sample scored more than 2 SDs higher on the Total Problem scale (Bolte et al., 1999). Compared to the clinical sample, the autistic sample had a comparable total psychopathology score on the CBCL. On the scales for social, thought, and attention problems, the average individual with autism scored at least 3 SDs higher than the normative sample, and the average individual with autism scored higher than the clinical sample, though Bolte et al. (1999) did not specify the difference between the clinical sample and the autistic sample in quantitative terms. They did include a graph with raw scores of the average scores for each of the three samples (autistic, clinical, and normative). Individuals with autism were below the clinical sample average regarding somatic complaints; the scores from the autistic sample were similar in comparison to the normative sample regarding somatic complaints. While the CBCL syndrome scales within the sample of individuals with autism were independent from sex ($F_{8, 68} = 0.89$; $p = 0.53$; effect size = 0.09), they were significantly influenced by age and level of intelligence ($F_{2, 73} = 3.1$; $p < 0.000$; effect size = 0.27) (Bolte et al., 1999).

The Brazilian version of the CBCL/4-18 was tested for validity of the identification of autism and related conditions in Duarte et al.'s (2003) study. They compared and contrasted 36 children with autism and related conditions, 31 children with other psychiatric disorders (OPD), and 34 school children, who were randomly selected and were similar in age and gender as the children with autism, in Brazil with an overall

age range of 4 to 11-years old and were mostly males. The comparison of paired group means showed a majority of the effect sizes as moderate to large. Comparison of the children with autism and the schoolchildren on the Thought Problems scale yielded the largest effect size. On the broadband scales in Duarte et al.'s sample, scores were comparable between the autistic group and the OPD group. Externalizing scores of the children with autism were also similar to the schoolchildren. Thought Problems and Autistic/Bizzare (a factor from the Rescorla, 1988 study) scales produced the best differentiation. The Thought Problem scale almost perfectly differentiated between the autistic group and the normal control group with 100% specificity and 94.3% sensitivity. In comparing autistic children with OPD children, the Thought Problems, Autistic/Bizzare, and Aggressive Behavior scales, differentiated the two groups. Autistic/Bizzare and Aggressive Behavior scales taken together showed the best discrimination of autistic versus OPD groups (sensitivity of 91.4% and specificity of 96.7%) (Duarte et al., 2003).

Sikora et al. (2008) examined the usefulness of the CBCL/1.5-5 and the GARS in identifying ASDs in a sample of 109 children with autism, 32 children with ASD, and 51 non-spectrum children based on the three different ADOS-Generic (ADOS-G) classifications (Autism, ASD, and Non-Spectrum); all participants were age 36-71 months. There was a significant positive correlation between the GARS Autism Quotient (AQ) and all CBCL scale scores, with the Withdrawn and Pervasive Developmental Problems scale scores having the strongest correlation to the GARS AQ. Using a MANOVA, significant differences, with small to moderate effect sizes, were demonstrated among the ADOS-G classification groups for the following CBCL scales:

Pervasive Developmental Problems, Anxious/Depressed, Withdrawn, and Aggressive Behavior. There also were significant differences between the autism and the non-spectrum group on these scales ($p < 0.05$), but there were no significant differences between the ASD and non-spectrum group or between the ASD and autism group. Among the CBCL scales that exhibited significant differences, the Withdrawn Scale accounted for the most variance among groups. There were no significant differences among groups for the GARS AQ (Sikora et al., 2008).

MANCOVAs also were employed to determine whether the GARS and CBCL scales were able to distinguish among ADOS-G classification groups when participant characteristics were controlled. There was a significant multivariate effect between the ADOS-G classification groups, and none for the GARS. There was a significant group effect for CBCL Withdrawn [$F_{2, 587} = 5.87$, $p = 0.01$] and Pervasive Developmental Problems [$F_{2, 307} = 3.76$, $p = 0.03$]. The CBCL Withdrawn and Pervasive Developmental Problems scales had better sensitivity than the GARS AQ in identifying children with autism, and the CBCL had better specificity than the GARS AQ, which suggests that the CBCL is a useful screener for ASD (Sikora et al., 2008).

The most recent published study examined the adequacy of the CBCL/1.5-5 factor model in a well characterized sample (N = 128) of preschoolers with ASD (Pandolfi et al., 2009). Confirmatory factor analyses revealed that the CBCL/1.5-5 measures the same constructs in children with ASD as it does in the normative population. The internal consistencies of most scales compared similarly to those reported by Achenbach and Rescorla (2000), except for the Somatic Complaints scale, in which the internal consistency was lower for the ASD sample. Except for the Anxious/Depressed scale, the

mean raw scores of the sample in this study were significantly higher than the normative sample, which supports the use of the CBCL to assess for emotional and behavioral disorders in preschoolers with ASD (Pandolfi et al., 2009).

Social Cognitive Learning Theory

While the CBCL examines behaviors, there are many explanations of the origin of various behaviors. One theory in particular, Bandura's social cognition theory (Schunk, 2004), espouses that learning occurs through actions or by vicariously observing models performing an action. Social cognition theory posits that behavioral consequences provide a source of information and motivation for learning the accuracy or appropriateness of a behavior, but people's cognitions rather than consequences, also can affect learning. Vicarious sources speed up the process of learning as opposed to if all learning had to be done firsthand. Vicarious sources also theoretically save people from personally experiencing negative consequences (Schunk).

People with ASD have deficient imitative skills, which may reflect an impaired ability to adapt their existing action repertoire as a result of observing novel actions. A functional magnetic resonance imaging (fMRI) study involving the copying of finger movements in people with ASD and controls, found that the control, but not the ASD group, activated a number of brain areas that included areas previously associated with social cognition. Controls seemed to use imitative processes to perform the task whereas the ASD group used visuomotor associative learning processes. The brain mechanisms involved in action-copying in the scanner may reflect approaches taken to social learning in daily life, and people with ASD place greater reliance on processes

involving visuomotor conditioning for social learning. This cognitive method does not facilitate the modification of an existing action repertoire through the observation of novel actions and the communication of mental states or imitation, which may lead to poor social cognitive development and rigid and repetitive patterns of behavior, characteristic of ASD (Williams, 2007).

One of the most powerful methods of socialization is imitation (Miller, 2002). Non-ASD siblings of children with ASD observe their ASD-siblings' maladaptive behavior (Williams, 2010). If the maladaptive behaviors of the ASD-siblings are being reinforced, whether negatively or positively, their non-ASD siblings may exhibit those behaviors to gain attention or other forms of positive or negative reinforcement as well; a general inclination to imitate is learned because various imitative behaviors are reinforced or not punished (Miller, 2002). For example, in the classic Bobo doll study by Bandura, Ross, and Ross (1960), subjects who were exposed to aggressive models, who were hitting the Bobo doll and verbally aggressing toward the doll, later imitated much of the physical and verbal aggression from the model. Likewise, subjects exposed to subdued and nonaggressive models, who were playing with tinker toys, and those who had no previous exposure to any models (control group) only rarely acted aggressively in play. The participants exposed to the aggressive models displayed their aggression in a manner that clearly looked like the novel patterns enacted by the models (Bandura, Ross, & Ross, 1961). Bandura would predict that observing aggression would increase the likelihood of the observer exhibiting aggressive behavior in imitation of the model, especially if there are no negative (punishment) consequences for the aggressive behavior in the model (Miller, 2002).

Furthermore, children need to pay attention to a model before the model can be influential. Salient behaviors, such as aggression, capture attention of children. Attention to a model and its behavior is most likely to occur if the model is salient and attractive, if the behavior is rather simple, and if the modeled behavior has some functional value (Miller, 2002; Schunk, 2004). Children without ASDs may see their ASD-siblings' behavior as captivating, and if the behavior is gaining attention or other forms of reinforcement from the parent, the non-ASD siblings may try to reproduce that negative behavior, particularly if there are no negative consequences for the behavior exhibited by the ASD-sibling.

In a study examining abnormal development in the social cognitive domain of siblings of children with Asperger's syndrome (AS), Dorris, Espie, Knott, and Salt (2004) compared these siblings to controls. Siblings ranged from 7-years, 6-month to 17-years, and individuals in the control group were in primary and secondary public schools in Scotland. Participants were sex-matched and matched as closely as possible for age and score on the British Picture Vocabulary Scale, version 11 (BPVS-II; Dunn, Dunn, Whetton, & Burley, 1997). Each participant was also administered the Eyes Test (Baron-Cohen, Wheelwright, Scahill, Spong, & Lawson, 2001), in which participants are asked to look at photographs of the eye area on a human face and then choose what word out of a list of four words fits best with what the person is thinking or feeling. Siblings' group score on the Eyes Test was significantly lower than the group of matched controls [$t = 1.968$, $df = 26$, $p = .030$]. Siblings scored more poorly than matched control participants in 59.3% (16 cases), while scoring higher than matched controls in 29.6% (8 cases).

Utilizing an ANCOVA, age and verbal comprehension (BPVS-II score) were a significant influence on the Eyes Test score ($p = .016$). Control participants performed significantly better than siblings of individuals with AS ($p = .0225$), and females outperformed males ($p < .0001$). Dorris et al. (2004) suggest that first degree-relatives of children with AS are affected by a milder version of the neuro-cognitive profile linked to AS/autism. This study shows that the profile of siblings of AS children in this sample have poorer social cognition than a group of matched controls and that males were significantly poorer in social cognition than females within and between groups and indicating moderately significant deficits in mental state awareness.

Dorris et al. also discussed the dearth of research into gene-environment interaction and the influence of social learning models in familial groups with the AS phenotype, in which a gene-environment interaction may exert an influence on AS relatives, but to a lesser extent than individuals with AS. In regards to the gene-environment interaction, there has been evidence of a broader phenotype of autism, even in families with one child with autism (Bolton et al., 1994; Bishop, Mayberry, Wong, Maley, & Hallmayer, 2006). Bolton found, in a sample of 99 children with autism, a familial aggregation of a lesser variant of autism, in which characteristics of family members without the diagnosed disorder were more subtle communication and social impairments or stereotypic behaviors. Approximately 12-20% of the siblings of children with autism exhibited the lesser variant/broader phenotype. Bishop et al.'s (2006) study suggested that there may be a familial relation between features of the broad phenotype across generations when looking at fathers' communicative and social behaviors.

While there is some evidence of genetic influence on behavior of non-affected siblings of children with autism, social learning theory also posits that children adopt a model's moral standards as part of their moral development (Miller, 2002). If a non-ASD sibling has an ASD-sibling who is older or close in age, as a model and consequences are not given or not given consistently to various negative behaviors exhibited by the ASD-sibling, the non-ASD sibling may learn more socially maladaptive behaviors than the average sibling with no ASD-siblings. Psychosocial behavior of non-ASD siblings may change, however, as they endure cognitive changes that come with development as an individual ages. Because they likely have different social environments outside of the home from their ASD-siblings, depending on IQ level, their psychosocial skills would likely be rated as less problematic on the CBCL, but possibly more significant than individuals from the normative population.

Several studies have examined psychosocial factors, such as general behavior problems and social competence of siblings of children with autism. The literature shows mixed findings regarding the siblings' psychosocial adjustment (Kaminsky & Dewey, 2002). Rodrigue, Geffken, and Morgan (1993) did not find significant psychosocial problems in siblings of children with autism. Rodrigue et al. (1993) compared siblings of children with autism (n= 19) to siblings of children with Down's syndrome (n = 20) and siblings of normally developing children (n = 20). They matched the participants on targeted children's mental age, gender, race, birth order, family size, and socioeconomic status. In this small sample 90% were white and middle to upper-middle class with siblings of severe developmental impairment, so results may have limited generalizability.

Rodrigue et al (1993) found significant group main effects of siblings' CBCL Internalizing score and Externalizing score. Mothers of children with autism rated siblings to have more internalizing and externalizing problems than siblings of developmentally normal children; however, these psychosocial problems of siblings of children with autism were not clinically significant. Rodrigue et al. did not find any significant main effects or interactions on perceived competence or social and behavioral adjustment. They did find, however, that older siblings in all three groups were likely to have higher internalizing problems (autism, $r = .63$, $p < .01$), and older siblings of children with autism were likely to have more externalizing problems ($r = .58$, $p < .01$). Lastly, they found marital satisfaction in families with children with autism to be highly correlated with higher levels of sibling perceived competence (Rodrigue et al.).

Gold (1993) also examined psychosocial factors in comparing differences between siblings of children with autism and siblings of typically developing children. The 22 siblings of boys with autism (previously diagnosed based on the DSM-III-R (APA, 1986) criteria) were recruited from agencies in Toronto servicing children with autism and their families and the 34 siblings of typically developing boys were recruited from local Toronto schools; all were between the ages of 7 and 17-years old. Gold did not find significant differences on the CBCL between the two groups in regards to Behavior Problems or Social Competence. However, Gold did find significant differences between groups on the Children's Depression Inventory (CDI; Kovacs, 1983), with siblings of boys with autism scoring significantly higher for depression than the comparison group, $F = 4.79$, $p < .04$. According to two of three possible cutoff scores ranging from 11 to 13, the siblings of boys with autism could be considered depressed.

Additionally, siblings who did not have someone to talk to about their sibling with autism scored significantly higher on the CDI than siblings who did have someone to talking to about their sibling with autism, $t = 2.56$, $p < .03$. The question regarding whether the sibling had someone to talk to about their sibling with autism was within the yes/no "Questions for Siblings" developed by Gold and listed in Appendix A (Gold, 1993). Regarding Pearson correlations, all of the correlates of higher CDI scores were moderately to highly correlated (.621-.915) to either the males with autism or the mother variables that described a sense of harassment or burden (based on the questions developed by Gold). For brothers, higher CDI scores significantly correlated with the response that there is nothing good about having a brother with autism. A higher CDI score for sisters correlated significantly and positively with specific characteristics of the child with autism. Specifically, those characteristics were the age of the sibling with autism, the length of time since diagnosis, and being younger than the sibling with autism (Gold).

In a questionnaire constructed by Gold (1993), siblings were asked "yes/no" questions that encompassed domestic work and caregiving work. Siblings of boys with autism did significantly less domestic work than other siblings, $F = 5.70$, $p < .03$, and sisters did more domestic work than brothers, $F = 4.25$, $p < .05$. In regards to correlations involving caregiving work and domestic work, sisters doing more caregiving work was significantly correlated with having a positive and realistic view of autism, a brother with moderate symptoms of autism, and parents who were not well-educated. Brothers of boys with autism who do greater amounts of domestic work had significant correlation with a

father who is dissatisfied with how he spends his days and a mother who is socially maladjusted and does relatively little caregiving work (Gold, 1993).

Similar to Rodrigue et al. (1993) and Gold (1993), siblings of children with autism (previously diagnosed based on DSM-III (APA 1980) and DSM-III-R criteria) were somewhat more negative in their perspectives regarding their sibling relationship than siblings of mentally retarded (MR) and typically developing children; however, all siblings were relatively positive in their expression of their sibling relationships (Bagenholm & Gillberg, 1991). Bagenholm and Gillberg did not find significant differences of characteristics of families across groups of typically developing siblings of children with autism, siblings of children with mental retardation without autism, and siblings of children with no apparent disorder. More than half of the siblings of children with autism were not able to describe what was wrong with their sibling with autism, and in regards to observed family stress seen in the home visit, there was an impression that more families of children with autism had high or extreme stress levels compared to the other two groups of children. Siblings of children with autism also seemed to have more problems with their sibling disturbing them and breaking their things and about what the future holds for their siblings with autism. In Bagenholm and Gillberg's sample, 35% of the siblings of children with autism reported feeling lonely. This study was conducted in Sweden and the data of Swedish parents were compared to results of American parents in Schaffer and Edgerton's (1979) study; negative feelings (e.g., hostility and embarrassment) received much higher scores in the American sample, which may be indicative of cultural differences.

Verte, Roeyers, and Buysse (2003) also found more behavior problems in siblings of children with high functioning autism spectrum disorders (ASD; n = 29) than siblings of normally developing children (n =29). The groups were matched on the target child's age and gender, siblings' age and gender, birth order, age spacing, and family size. The children with high functioning ASD, were involved in a residential treatment program and had an IQ >80 and had no co-morbid diagnoses. The mean age of siblings in the high functioning ASD group was 11.14 (SD = 3.23) and 11.31 (SD = 3.29) in the control group. Siblings of children with high functioning ASD had more internalizing problems and externalizing problems, as rated on the CBCL by parents, ($F_{2, 49} = 5.88$, $p = 0.01$). Verte et al. (2003) also found an interaction effect of group and age ($F_{2, 49} = 12.29$, $p < 0.001$). Siblings of children with high functioning ASD between 6 and 11-years old had more internalizing and externalizing problems; however, mean scores of this group did not reach clinical significance or the at-risk range, similar to the Rodrigue et al. (1993) study.

Verte et al. (2003) also examined social competence and self-concept, using self-measures of Matson Evaluation of Social Skills with Youngsters (MESSY; Matson et al., 1983) and the Self-Description Questionnaire I and II (Marsh, 1988), besides the CBCL, and found that sisters of children with high functioning ASD had rated their social competence higher than sisters of the control group. Additionally, sisters of the high functioning ASD group between ages 12 and 16-years old had a more positive self-concept. Verte et al. found a significant relationship between total self-concept and total score of the MESSY and the total competence score on the CBCL, meaning that sibling with a more negative self-concept exhibited lower social competence skills, and siblings

with a more positive self-concept scored higher in the social domain with no group effect. There was also a significant relationship between the MESSY total score and the Problem scale of the CBCL, indicating that overall ratings of siblings and parents matched regarding social competence.

Kaminsky and Dewey (2002) also examined psychosocial adjustment in siblings of children with autism but with a larger sample of 90 normally developing children of siblings with autism, Down syndrome, or no known disability. There was a trend for males to have lower Social Competence scores on the CBCL than females. Exploratory analyses showed that female siblings of children with autism had significantly higher Social Competence scores than male siblings, $F_{1, 88} = 10.5$, $p < .01$; however, overall, siblings of children with autism did not exhibit deficits in social competence. In regards to only siblings of children with autism, significantly moderate negative correlations were seen between loneliness and social support from peers, r = -.65 and significantly low negative correlations between loneliness and social support from friends, r = -.47. Additionally, social support from peers was significantly correlated with academic problems on the CBCL, r = .60. Kaminsky and Dewey also found significant negative correlations between the family size and the CBCL Total Adjustment Scale score, r = -.52, the Internalizing Factor score, r = -.46, and the Externalizing Factor score, r = -.51. Overall, Kaminsky and Dewey's study showed that siblings (ages 8-18) of children with autism are not at high risk for adjustment difficulties or loneliness, which may be confounded by the high number (77%) of participants' families who attended support groups.

In another psychosocial study examining siblings of children with autism, 22 mothers rated the adjustment of the sibling closest in age to their child with autism and MR who was attending a school for children with autism in the United Kingdom (Hastings, 2003). The children with autism ranged in age from 7 to 16-years old, and the siblings ranged in age between 4 and 16-years old. Based on mothers' ratings of the typically developing siblings on the Strengths and Difficulties Questionnaires (SDQ; Goodman, 1997), Hastings found that they were rated as having more peer problems, more overall adjustment problems, and lower levels of prosocial behavior compared to a normative sample of British children from a separate large scale study (Meltzer, Gatward, Goodman, & Ford, 2000). Hastings also found that sibling variables accounted for a significant amount of the variance (55%) in Pro-Social Behavior scores [$F_{5,6} = 3.98$, $p < .05$]. The most notable effects of sibling variables were in relation to the sibling's sex, with sisters having better adjustment compared to brothers, and relative ages of the children, with siblings who were older than their brother or sister with autism being better adjusted (Hastings).

Hastings (2003) remarked that the lack of statistical power in sibling research is an important issue to consider because samples are typically small. Hastings also noted that his findings are helpful in confirming that siblings are a vital part of the family whose mental health should be addressed by practitioners. This finding is also congruent with Gold's (1993) study, in which siblings who scored higher on the CDI were those who had no one to talk to about having a sibling with autism. Mascha and Boucher (2006) further examined the typically developing sibling's perspective from a qualitative method of analysis. Participants in Mascha and Boucher's study included 14 siblings

from 11 families recruited from an advertisement in an autism newsletter in the United Kingdom: five children were diagnosed with moderate to low functioning autism, three with high functioning autism, and three with Asperger's syndrome. They tentatively found that behavioral problems frequently related to ASDs, particularly aggression and uncontrolled anger, cause a mix of negative emotion in typically developing siblings. If these findings are confirmed in future research, there would be definite implications for support services needed for siblings of children with an ASD (Mascha & Boucher).

In a multiplex study, involving normally developing siblings of children with autism from families who have more than one child with autism, siblings of children with autism or any pervasive developmental disorder were at risk for developing deficits in social impairment (Constantino et al., 2006). Constantino et al. compared closest-in-age non-autistic brothers of children with autism from multiplex families along with an autistic sibling (n = 49 pairs), individuals with any pervasive developmental disorder and their closest-in-age brothers (n = 100 pairs), and individuals with psychopathology unrelated to autism (i.e., disruptive behavior disorders, affective disorders, or anxiety disorders) and their closest-in-age brothers. All subjects were recruited from the Autism Genetic Resource Exchange and the Washington University Child and Adolescent Psychiatry Service, ranged in age from 4 to 18-years old, and were predominantly Caucasian. Siblings of children with autism from multiplex families were the most impaired group on the Social Responsiveness Scale in comparison to the other sibling groups, followed by siblings of any pervasive developmental disorder, and then by sibling of individuals with psychopathology unrelated to autism ($F_{2,188} = 16.2$, $p < .000001$).

Having a sibling with autism would be expected to have an effect on social and emotional adjustment of a family due in part by possible aggressive and self-injurious behaviors, impulsivity, hyperactivity, rituals, severe communication deficits, and having to face the response of others. Siblings may need to cope with changes in family roles, structure, and activities, feelings of guilt and shame, loss of parental attention, and increases in parental stress. Pilowsky, Yirmiya, Doppelt, Gross-Tsur, and Shalev (2004) examined these factors of social and emotional adjustment of siblings of children with autism. They compared 30 siblings of children with autism, 28 siblings of children with mental retardation (MR), and 30 siblings of children with a developmental language delay (DLD).

Within the autism group, Pilowsky et al. (2004) found a significant correlation between siblings' chronological age and their emotional description of their sibling with autism (r = .50; p = .009). Older siblings described their sibling with autism more positively. Analyses showed significant effects based on the children with autism's verbal ability on siblings' behavior problems (CBCL: $t_{(21)} = 3.23$, p = .004) and socialization problems (VABS: $t_{(22)} = -3.80$, p = .001). Siblings of nonverbal children with autism had more behavior problems and lower socialization skills than siblings of verbal children with autism. These results are similar to Gold's (1993) findings in that severity of the children with autism seems to be related to poorer sibling adjustment. Additionally stress intensity was inversely correlated with siblings' socialization skills (VABS-Social domain: r = -.626, p = .001), and family size was correlated with siblings' socialization skills (VABS-Social domain: r = -.526, p = .006). The larger the size of the family, the greater the delay in siblings' socialization skills. Similar to previous studies (Bagenholm

& Gillberg, 1991; Gold, 1993; Kaminsky & Dewey, 2002; & Rodrigue et al., 1993), Pilowsky et al. (2004) found that most of the siblings of children with autism are fairly well adjusted; however, the small sample size and variances found on the different measures may have affected the statistical power of analyses.

In line with the social cognition theory, children with siblings who have ASDs may learn negative behaviors that receive attention or other forms of reinforcement, rather than negative consequences; therefore, children with ASDs may be providing a maladaptive model for learning positive psychosocial skills. However, if typically developing siblings see negative consequences for the sibling with an ASD exhibiting negative behaviors, then the typically developing children may inhibit exhibiting those maladaptive behaviors. The importance of the findings would be the implications they have for whether typically developing siblings of children with ASDs need behavioral or psychosocial support. An additional implication may be whether parents would benefit from parent training/psychoeducation on behavioral techniques for discipline, as well as psychosocial support, and possible family therapy.

Some research leans toward genetics influencing behavior in a broader phenotype of autism regarding non-affected siblings of siblings with ASDs (Bishop et al., 2006; Bolton et al., 1994; Doris et al., 2004), while most do not evaluate specifically for nature versus nurture (Kaminsky & Dewey, 2002). One study, Doris et al., addressed the possibility of genetic and environmental interaction and the influence of social learning models on individuals with the AS familial phenotype. Many of the studies that found significant differences in internalizing and/or externalizing behaviors between non-affected siblings and control children on parent ratings of psychosocial measures did not

find clinically significant differences (Bagenholm & Gillberg, 1991; Rodrigue et al., 1993; Verte et al., 2003). The lack of clinical significance may be due to the small sample sizes of previous psychosocial studies. Kaminsky and Dewey (2002) found a moderate negative correlation between loneliness and peer support for unaffected siblings, but a large portion of families in their study were involved in support groups. Gold (1993) found that non-affected siblings had more impairment if they reported that they had no one to talk to about their sibling with autism. Some studies found that non-affected siblings older than their sibling with autism were better adjusted than younger non-affected siblings (Hastings, 2003; Pilowsky et al., 2004). In the Hastings' study, though, the non-affected siblings with a sibling with autism who mostly lived at a boarding school were rated as having more behavior and peer problems, as rated by their mothers, than the normative sample (Hastings, 2003). In the Pilowsky et al. study, siblings of children with autism who had verbal abilities had fewer social and behavioral problems than siblings of children with autism who were nonverbal. Social cognition learning theory may view the results of these studies to indicate that older non-affected siblings have become positive role models for their younger ASD siblings. Families who attend support groups may be learning discipline strategies and positive ways to approach their children with autism that would deter maladaptive behaviors, thus reducing any negative models of behavior for the non-affected siblings. The families in the support groups may also be learning ways to direct their attention to the non-affected children, too, to aid in positive psychosocial development. On the other hand, whether or not non-affected siblings of children with ASDs have behavioral and psychosocial problems may be contingent on a broader phenotype of ASD in the family.

This study sought to determine whether the risk of developing behavior problems differs for children who have a sibling diagnosed with an ASD compared to the normative population, as indicated by scores on a broad band measure of behavioral disturbance, the Parent Report Form of the Child Behavior Checklist (CBCL). Social learning theory posits that the sibling may either imitate the inappropriate behaviors of their diagnosed siblings, earning higher CBCL scores, or through observational learning they may learn to inhibit or avoid inappropriate behaviors by observing the unwelcome consequences of these behaviors applied by their parents. The study also investigated the impact of age and a proxy for IQ (the Vineland Adaptive Behavior Scales-II, or Vineland-II) on the CBCL scores of the siblings with and without ASD. Specifically, this study is asking the following research questions: Is the ASD group significantly more impaired than non-affected sibling and the normative sample in psychosocial functioning/behavior, based on CBCL profile scores? Is the non-affected sibling group more impaired than the normative group? Do age and/or IQ differentially affect CBCL scores based on the three groups? Particularly are younger children and lower IQ scores associated with significantly more impaired CBCL scores?

CHAPTER II

Methodology

Participants

In this research study there were three groups: ASD (experimental group, which will be referred to as proband), non-affected siblings (comparison group), and the normative population (control group) from the CBCL. The proband and non-affected sibling participant data were archival data from the Simons Foundation Autism Research Initiative (SFARI). The SFARI Simplex Collection (SSC) is a new resource for autism research that provides information on a highly characterized set of simplex families. Simplex families refer to only one child diagnosed with an ASD and without either parent diagnosed with an ASD. The Simons Foundation has standardized and coordinated patient evaluation at multiple medical centers across North America. There are over 2,000 simplex families in the database. The data that have been collected examine genotype, which is the set of genes a person carries, and the phenotype, which is the observable set of characteristic of an individual.

Inclusion criteria.

The proband group consisted of children with a diagnosis of Autistic Disorder, Asperger's Disorder, or PDD-NOS, according to the DSM-IV-TR, and who are between 4-years and 17-years, 11-months of age. On the ADI-R, the probands must have met one of the following four criteria: standard cutoffs on the Social and Communication domains, standard cutoff on the Social domain and within two points of Communication cutoff, standard cutoff on the Communication domain and within two point of Social cutoff, or is within one point of the standard cutoffs on both Social and Communication

domains. All ADI-R data had to be collected by a "reliable examiner," as indicated on the SFARI website. This standard is considerably higher than is the case for many studies. An examiner becomes a "reliable examiner" when that examiner has achieved at least 90% agreement with the University of Michigan Autism and Communication Disorders Center (UMACC) lab.

Examiners have to complete both the ADOS Training for Clinicians and the ADOS Training for Researchers (at UMACC) before beginning the reliability process. Examiners are required to establish reliability of at least 80% on the instrument before collecting data for SFARI. If someone training to be a reliable examiner receives training from someone who has established reliability on the ADOS, the potential examiner can complete the steps of establishing reliability with that person, as long as the potential examiner is working at the same physical site at the already established reliable examiner and have frequent (i.e., daily) contact to talk about coding and administration issues. To achieve reliability, UMACC must have evaluated the examiner's administration and coding on three different administrations of the Modules 1 / 2 and on three different administrations of Modules 3 / 4.

On the ADOS, the proband participants must have received a valid and reliable administration and must meet cutoffs for ASD or autism. Following suggestions in the ADOS manual, raters select the appropriate module for each person. Module 1 is used with children who do not consistently use phrase speech, Module 2 with those who use phrase speech but are not verbally fluent, Module 3 with fluent children, and Module 4 with fluent adolescents. New scoring algorithms for the ADOS were released in 2007 to enhance sensitivity and specificity (Gotham, Risi, Pickles, & Lord, 2007). The new

ADOS algorithms were used for Modules 1-3, and the original cutoff algorithms were allowed for Module 4.

Nonverbal cognitive ability was measured with the Mullen Scales of Early Learning (1995), Differential Ability Scales--Second edition (DAS-II; Elliott, 2007), Wechsler Intelligence Scale for Children—Fourth edition (WISC-IV; Wechsler, 2003), or Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999). Probands who were 4-years old must have had a nonverbal deviation or ratio IQ score greater than or equal to 60. Only probands were administered measures of intelligence. Probands between the ages of 5 and 8-years old must have had a nonverbal deviation score or nonverbal ratio IQ score greater than or equal to 40. Probands 8-years old or older must have had a nonverbal mental age of 36-months or older.

Exclusion criteria.

Families who met one or more of the following were excluded from the Simons Simplex database: Individuals who were fewer than 36 weeks gestation and less than 2,000 grams at birth and/or had a history of extensive pregnancy or birth complications. A history of significant prenatal/birth injury or brain damage required more extensive evaluation if the child with the ASD was in the NICU or in the hospital for greater than three days. Individuals with ASDs who also had other disorders or limitations were excluded, including those who were positive for Fragile X Syndrome or Down Syndrome. However, if individuals met the criteria for inclusion based on clinical phenotype measures, no other genetic diagnosis was excluded because the purpose of inclusion of these individuals is to identify additional risk factors or modifier genes that

may correlate with the autism phenotype. Other exclusionary factors for the probands were sensory or motor difficulties that would preclude valid use of the diagnostic instruments and a history of severe nutritional or psychological deprivation.

Any families with full or half-siblings were excluded if the children were diagnosed with or referred for ASD. Families also were excluded if siblings were identified as having mental retardation with the exception of Down Syndrome. Families with siblings diagnosed with schizophrenia or with psychiatric disorders requiring treatment with more than one psychotropic medication were not included in the research database. If siblings had an Adaptive Behavior standard score on the Vineland-II that was 70 or below or had an Individualized Education Plan for extensive special education services, families were excluded from the database. Additionally, if either biological parent was diagnosed with ASD, identified as having mental retardation, or diagnosed with schizophrenia, those families were excluded. Any second-degree or third-degree relatives diagnosed with ASD ruled out participation in the Simon Foundation database. Exclusionary criteria also included families who were participating in other autism research or in research of disorders related to autism (e.g., Obsessive Compulsive Disorder).

Instruments

Child behavior checklist (CBCL).

The CBCL is a behavioral questionnaire that produces standardized ratings of a broad range of behavioral, emotional, adaptive and social functioning. The present study will include only parent forms. The Pre-school form (CBCL 1.5-5) was used for probands

ages 4-5 and non-affected siblings age 1-year, 6-months to 5-years, 11-months and under, and the CBCL for ages 6 to 18 was used for probands and non-affected siblings who were between the ages of 6 and 18-years old. The scores of the scales are T-scores, with a mean of 50 and a standard deviation of 10, with separate norms available for 6-11 and 12-18 year-olds.

The CBCL/1.5-5 and CBCL/6-18 ask for demographic information about the child and asks parents to indicate their name and their relationship to the child, as well as their occupations to determine socioeconomic status. On the CBCL/1.5-5, the parent then rates 99 problem items as 0 for not true of the child, 1 for somewhat or sometimes true, and 2 for very true or often true, based on the previous 2 months. Some items require explanatory details, and one of the questions asks the parent to write in any additional problems that were not previously listed. Additionally, there are open-ended items that ask the parent to describe any illnesses or disabilities that the child has, what concerns the parent most about the child, and the best things about the child (Achenbach & Rescorla, 2000; Achenbach & Rescorla, 2001).

On the CBCL/6-18, the parent completes competence items, followed by open-ended items describing the child's illnesses and disabilities, what concerns the parent most about the child, and the best things about the child. Parents are also requested to rate 112 items of behavioral, emotional, and social problems as 0= not true, 1= somewhat or sometimes true, and 2= very true or often true, based on the previous 6 months. Item 113 asks the respondent to describe and rate any problems that were not listed previously, and item 56 requests raters to describe and rate any additional physical problems (Achenbach & Rescorla, 2001).

The syndrome scales on the CBCL/1.5-5 are Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Sleep Problems, Attention Problems, and Aggressive Behavior; the DSM-Oriented scales are Affective Problems, Anxiety Problems, Pervasive Developmental Problems, Attention Deficit/Hyperactivity Problems, and Oppositional Defiant Problems. The broader categories are Externalizing, Internalizing, and Total Problems. The estimated time to complete the CBCL/1.5-5 is 10 minutes (Achenbach & Rescorla, 2000). The syndrome scales on the CBCL/6-18 are Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behavior, and Aggressive Behavior, and there are also the broader categories of Externalizing, Internalizing, and Total Problems. The DSM-Oriented scales are Affective Problems, Anxiety Problems, Somatic Problems, Attention Deficit/Hyperactivity Problems, Oppositional Defiant Problems, and Conduct Problems. Additionally, there are four Competence scales: Activities, Social, School, and Total Competence. The estimated time to complete the CBCL/6-18 is 15-20 minutes (Achenbach & Rescorla, 2001). The scales chosen for this study were Anxious/Depressed, Withdrawn, Withdrawn/Depressed, Sleep Problems, Attention Problems, Aggressive Behavior, Total Problems, Pervasive-Developmental Problems, Social Problems, Thought Problems, Internalizing, and Externalizing. They were chosen based on scales used in previous studies (Bolte et al., 1999; Duarte et al., 2003; Kaminsky & Dewey, 2002; Pandolfi et al., 2009; Rodrigue et al., 1993; Sikora et al., 2008; Verte et al., 2003)

The CBCL normative, non-referred, sample excluded 13.6% of the 2, 029 children whose parents participated in the normative data collection due to the children

receiving mental health services, substance use, and special education without mental health or substance use services. Gender was fairly evenly split, with 52% males and 48% females. Regarding socioeconomic status, 33% fell in the upper income range, 51% were in the middle income range, and 16% fell in the lower income range. While a majority of the sample was White, non-Latino (60%), 20% of the participants were African American, 9% were Latino, and 12% were Mixed/Other.

Regarding psychometric properties of the CBCL/1.5-5, test-retest reliability of most scales on the CBCL/1.5-5 was high, with most Pearson r's between the .80s and .90s. The internal consistency ranges from moderate to strong, with a coefficient alpha ranging from .63 to .96. The mean stability at a 12-month interval was .61 (Achenbach & Rescorla, 2000). Regarding psychometric properties of the CBCL/6-18, test-retest reliability for 8 or 16-day intervals was very high, with the mean ranging from .88 to .90. The internal consistency was moderately high with alphas ranging from .63 to .97. For the CBCL/6-18 stability over 12 and 24 months, mean Pearson r's were between .61 and .74 (Achenbach & Rescorla, 2001).

Content validity, criterion-related validity, and construct validity were examined in the CBCL/1.5-5 and the CBCL/6-18. Content validity was examined based on previous research with the scales; items that previously were not scored higher for referred than non-referred children were replaced. Classification accuracy according to referral status was recorded using discriminant analysis at 84.2% on the CBCL/1.5-5 and at 79-85% on the CBCL/6-18. Construct validity on the CBCL/1.5-5 correlations range from .46 to .72 (Achenbach & Rescorla, 2000). Construct validity on the CBCL/6-18 correlations with the Conners' Rating Scales-Revised were very high, ranging from .71 to .85, correlations

with the BASC scales ranged from .38 to .89 (all p <.01), and correlations with the DSM-IV Checklist were all moderate, mostly in the .60's (Achenbach & Rescorla, 2000; Achenbach & Rescorla, 2001).

Vineland Adaptive Behavior Scales, Second Edition (Vineland-II).

Vineland Adaptive Behavior Scales, Second Edition (Vineland-II; Sparrow, Cicchetti, & Balla, 2005) is an adaptive behavior measure for ages birth through 90. The form used for the purposes of this research study was the Survey Interview Form for the parent. The form is administered in a semi-structured interview format. Standard scores on the Vineland-II are a mean of 100 and a standard deviation of 15. The measure examines broad domains of Communication, Daily Living Skills, Socialization, and Motor Skills; however, for the purpose of this study, only the Adaptive Behavior Composite, which incorporates all of the above domains, was utilized in analysis as a proxy for IQ (Bolte, Dickhut, & Poutska, 1999).

Regarding reliability, internal consistency, as measure through the split-half method, was high (.86-.98) for adaptive behavior. Test-retest reliability coefficients ranged from .81 to .96. Interinterviewer reliability of the sample for ages birth through 6 is .87 for the Adaptive Behavior Composite and .74 for the 7-18-year old group. Regarding validity, Scores from the Vineland-II correlated with scores from the Adaptive Behavior Assessment System, Second Edition (ABAS-II; Harrison & Oakland, 2003). From ages birth to 5-years, the adjusted correlation between the Vineland-II and the ABAS-II General Adaptive Composite is .70. The mean scores on the composite scores of both measures were very similar.

This study initially had proposed to use IQ as a covariate, not adaptive behavior. However, IQ scores were not available for the typically developing siblings. The Vineland-II was selected as a proxy based on its use as a proxy in an ASD study by Bolte, Dickhunt, and Poustka (1999). However, the limitations of using the Vineland-II as proxy were demonstrated by Sparrow, Cicchetti, and Balla (2005). For example, the Vineland-II was compared to the Wechsler Intelligence Scale for Children –III (WISC-III) (Wechsler, 1991), and there was low correlation between Adaptive Behavior Composite and the Verbal, Performance, and Full Scale IQ scores, indicating they measure different constructs (Sparrow et al., 2005). Nevertheless, the Vineland-II was judged to be the best proxy for IQ available in the SFARI data set.

Procedures

Once permission was obtained from the University of Houston's Committee for the Protection of Human Subjects (CPHS), the research investigator mailed the signed Research Distribution Agreement, a required document to be signed by University of Houston upon approval from CPHS, to the Simons Foundation. Additionally, the approval letter from CPHS in pdf format was uploaded onto the SFARI website. After approval was obtained from the Simons Foundation Autism Research Initiative (SFARI), the investigator submitted an online request through SFARI to obtain the archival CBCL scale scores, ages, Vineland-II, IQ scores, and demographic data. Once the data were obtained, all the data from "trio" families (no siblings) were removed from the data set, which left 1, 996 probands. Then 98 sets of sibling pairs were removed due to missing age of probands, missing Vineland-II scores from either group, and missing CBCL scores

for all scales of interest in either group. Overall, 4.9% of the sibling pairs from quad (families with two parents, one child with ASD, and one included sibling) families were removed.

In the sample of 1,898 sibling pairs, 86.8% of the probands were male, and 13.2% were female. Of the non-affected siblings, 46.4% were male, and 53.6% were female. In regards to level of education, 61% of mothers and 59.5% of fathers were educated at the baccalaureate or graduate level, and most at least completed high school. In regards to socioeconomic status, most families' income was between $36K and > $161K. A majority of the sample was white, non-Hispanic (75.2%); 4.5% were white, Hispanic, 3.3% were African American, non-Hispanic, 3.7% were Asian, 7.6% were multiracial, and the remainder fell under Other, Not Specified, Native American, African American, Hispanic, and Native Hawaiian. The mean age of the probands was 9.11 years, with a standard deviation of 3.30. The mean full scale IQ for the probands (There are only IQ data for the probands.) is 82, with a standard deviation of 27.8; two proband participants were missing IQ data. While age data for the probands were provided through the database in month format, age data had to be calculated for non-affected siblings. Data to calculate sibling ages were not included in the original data set and were requested and received after the initial data set was received. Data regarding the typically developing siblings' ages were available in the form of date of birth and the date of evaluation; the sibling ages were calculated from this information. The mean age of the typically developing siblings was 9.42 years, with a standard deviation of 3.67.

CHAPTER III

Data Analysis

To determine whether the proband group is significantly more impaired than the normative sample and the non-affected sibling group, multiple Bonferroni-corrected, two-tailed t-tests were completed comparing the means of the probands to the means of the normative sample on scales from the CBCL 1.5-5, which will be referred to as Pre-school and CBCL 6-18 (with separate norms for ages 6-11/Child and 12-18/Adolescent). To determine whether the non-affected sibling group is more impaired than the normative group multiple Bonferroni-corrected, two-tailed t-tests comparing the means of the non-affected siblings to the means of the normative sample on scales from the Pre-School, Child, and Adolescent CBCL. All t-test statistics were intended to be evaluated for significance at a Bonferroni-corrected alpha level of 0.0009, which was calculated by dividing an alpha of .05 by the number of t-tests (n=54). This calculation reduces the chances of Type I errors, which increase with multiple t-tests. Because software limitations precluded the use of the 0.0009 alpha level, it was necessary to round the alpha upwards by a value of 0.0001 to a slightly less conservative alpha level of .001. CBCL 6-18 scale means were calculated separately for males and females. Because the differences were small (0.0-0.4), the average of the means were used as the test values for the all the Child and Adolescent one-sample t-tests. The results of the multiple Bonferroni-corrected, two-tailed t-tests comparing the means of the non-affected siblings to the means of the normative sample on scales from the Pre-school, Child, and Adolescent groups are presented next.

Comparisons of Probands and Normative Sample: Pre-school

The proband data for the Pre-school t-tests included individuals ages 4-6.25 years-old (mean = 5.02; standard deviation = .63). Some parents of the probands were given the Pre-school form for children older than 5-years, 11-months of age; this is likely due to when the families were screened and then subsequently evaluated. It could take a few months to get a family into the study for a full evaluation. There were 355 probands for the Internalizing and Externalizing t-tests, and 351 probands for the t-tests on the other scales subsequently listed. The difference in sample size is due to missing data.

In comparison to the normative CBCL Pre-school sample, the probands were significantly higher on the following scales: Internalizing, Externalizing, Withdrawn, Sleep Problems, Attention Problems, Aggressive Behavior, Total Problems, and Pervasive-Developmental Problems. The probands were not significantly different from the normative sample on the Anxious-Depressed scale. Means and SDs for the probands are depicted in Table 1, and the results of the t-tests are reported in Table 2.

Table 1
Means and SDs for Probands : Preschool

Scale	Mean	SD
Internalizing	61.21	9.15
Externalizing	57.56	10.92
Anxious-Depressed	55.00	7.38
Withdrawn	70.77	9.28
Sleep Problems	57.35	9.89
Attention Problems	62.08	8.31
Aggressive Behavior	58.36	9.82
Total Problems	60.95	10.69
Pervasive-Developmental Problems	71.52	8.93

Table 2
Comparing Means of Pre-School Probands and Normative Sample

Scale	Sample size	Test Value (Standardization Mean, N = 700)	t	Sig. (2-tailed)	Mean Difference	Effect size	95% Confidence Interval of the	
							Lower	Upper
Internalizing	355	50.0	23.083	.000*	11.206	1.18	10.250	12.160
Externalizing	355	50.0	13.047	.000*	7.563	0.73	6.420	8.700
Anxious-Depressed	351	54.2	2.031	0.043	0.800	0.12	0.030	1.570
Withdrawn	351	54.1	33.643	.000*	16.666	2.15	15.690	17.640
Sleep Problems	351	54.2	5. 960	.000*	3.148	0.39	2.110	4.190
Attention Problems	351	54.1	17.983	.000*	7.977	1.13	7.100	8.850
Aggressive Behavior	351	54.2	7.937	.000*	4.159	0.51	3.130	5.190
Total Problems	351	50.1	19.031	.000*	10.854	1.05	9.730	11.980
Pervasive-Developmental	351	54.1	36.541	.000*	17.424	2.33	16.490	18.360

* p <.001

Comparisons of Probands and Normative Sample: Child

The proband data for the Child t-tests included ages 5.67-11.92-years old (mean = 8.69; SD = 1.65). Some parents of the probands were given CBCL 6-18 for children younger than 6-years old, which is likely due to when the families were screened and then subsequently evaluated. There were 1,166 participants in the t-tests for the Internalizing and Externalizing scales and 1,163 participants in the t-tests of the remainder of scales; differences in the number of participants were due to missing data.

In comparison to the normative 6-11-year old sample, the probands were significantly higher on all the scales examined: Internalizing, Externalizing, Anxious-Depressed, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behavior, and Total Problems. Means and SDs for the ASD group are depicted in Table 3, and the results of the t-tests are reported in Table 4.

Table 3
Means and SDs for Probands: Child

Scale	Mean	SD
Internalizing	59.81	9.735
Externalizing	56.77	10.561
Anxious-Depressed	59.18	9.058
Withdrawn	62.92	8.624
Social Problems	62.66	7.954
Thought Problems	67.33	8.363
Attention Problems	67.50	10.179
Aggressive Behavior	60.01	9.289
Total Problems	63.07	8.680

Table 4
Comparing Means of Child Probands and Normative Sample

Scale	Sample size	Test Value (Standardization Mean, N = 777)	t	Sig. (2-tailed)	Mean Difference	Effect size	95% Confidence Interval of the Lower	Upper
Internalizing	1166	50.15	33.870	.000*	9.656	1.00	9.10	10.22
Externalizing	1166	50.05	21.735	.000*	6.722	0.67	6.12	7.33
Anxious-Depressed	1163	54.20	18.755	.000*	4.981	0.66	4.46	5.50
Withdrawn	1163	54.30	34.073	.000*	8.617	1.18	8.12	9.11
Social Problems	1163	54.40	35.412	.000*	8.260	1.20	7.80	8.72
Thought Problems	1163	54.15	53.740	.000*	13.179	1.87	12.70	13.66
Attn Probs	1163	54.45	43.733	.000*	13.054	1.59	12.47	13.64
Aggress Beh	1163	54.20	21.312	.000*	5.805	0.75	5.27	6.34
Total Probs	1163	49.80	52.135	.000*	13.275	1.43	12.78	13.77

* p <.001

Comparisons of Probands and Normative Sample: Adolescent

The proband data for the Adolescent t-tests included 374 probands. The range of ages for all the Adolescent t-tests for the probands was 12-17.92-years old with a mean = 14.33 and SD = 1.62. In comparison to the Adolescent normative sample the probands were significantly higher on all the scales examined: Internalizing, Externalizing, Anxious-Depressed, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behavior, and Total Problems. Means and SDs for the proband group are depicted in Table 5, and the results of the t-tests are reported in Table 6.

Table 5
Means and SDs for Probands: Adolescent

Scale	Mean	SD
Internalizing	60.82	9.511
Externalizing	54.01	10.810
Anxious-Depressed	60.55	9.359
Withdrawn	62.82	9.089
Social Problems	65.97	8.418
Thought Problems	67.47	8.323
Attention Problems	64.74	9.569
Aggressive Behavior	58.76	9.220
Total Problems	62.02	8.379

Table 6
Comparing Means of Adolescent Probands and Normative Sample

Scale	Sample size	Test Value (Standardization Mean, N = 976)	t	Sig. (2-tailed)	Mean Difference	Effect size	95% Confidence Interval of	
							Lower	Upper
Internalizing	374	50.15	21.702	.000*	10.674	1.11	9.71	11.64
Externalizing	374	50.1	6.991	.000*	4.908	0.48	2.81	5.01
Anxious-Depressed	374	54.05	13.423	.000*	6.495	0.84	5.54	7.45
Withdrawn	374	54.5	17.694	.000*	8.316	1.10	7.39	9.24
Social Problems	374	54.05	27.379	.000*	11.918	1.66	11.06	12.77
Thought Problems	374	54.15	30.945	.000*	13.318	1.89	12.47	14.16
Attention Problems	374	54.5	20.692	.000*	10.238	1.31	9.27	11.21
Aggressive Behavior	374	54.25	9.453	.000*	4.507	0.58	3.57	5.44
Total Problems	374	49.85	28.092	.000*	12.171	1.32	11.32	13.02

* p <.001

Comparisons of Non-Affected Sibling Group and Normative Sample: Pre-school

The non-affected sibling data for the Pre-school t-tests included siblings ages 3.42-6.5-years old (mean = 4.86; SD = 0.66). Some parents of the non-affected siblings were given the Pre-school form for children older than 5-years, 11-months of age, which is likely due to when the families were screened and then subsequently evaluated. There were 382 individuals in the non-affected sibling group for the Internalizing and Externalizing t-tests, and 380 individuals in the non-affected sibling group for the t-tests on the other scales subsequently listed. The difference in sample score is due to missing data.

In comparison to the normative Pre-school sample, the non-affected sibling group was significantly lower on all scales examined: Internalizing, Externalizing, Anxious-Depressed, Withdrawn, Sleep Problems, Attention Problems, Aggressive Behavior, Total Problems, and Pervasive-Developmental Problems. Means and SDs for the non-affected sibling group are depicted in Table 7, and the results of the t-tests are reported in Table 8.

Table 7

Means and SDs for Non-Affected Sibling Group: Pre-School

Scale	Mean	SD
Internalizing	43.09	10.197
Externalizing	43.76	9.840
Anxious-Depressed	52.09	4.704
Withdrawn	51.67	4.654
Sleep Problems	52.45	4.544
Attention Problems	51.96	4.244
Aggressive Behavior	52.02	4.880
Total Problems	42.58	9.821
Pervasive-Developmental Problems	51.47	4.281

Table 8

Comparing Means of Pre-School Non-Affected Sibling Group and Normative Sample

Scale	Sample size	Test Value (Standardization Mean, N = 700)	t	Sig. (2-tailed)	Mean Difference	Effect size	95% Confidence Lower	95% Confidence Upper
Internalizing	382	50.0	-13.252	.000*	-6.914	-0.69	-7.94	-5.89
Externalizing	382	50.0	-12.395	.000*	-6.214	-0.63	-7.23	-5.25
Anxious-Depressed	380	54.2	-8.746	.000*	-2.111	-0.40	-2.59	-1.64
Withdrawn	380	54.1	-10.175	.000*	-2.429	-0.46	-2.90	-1.96
Sleep Problems	380	54.2	-7.496	.000*	-1.747	-0.34	-2.21	-1.29
Attention Problems	380	54.1	-9.814	.000*	-2.137	-0.43	-2.56	-1.71
Aggressive Behavior	380	54.2	-8.693	.000*	-2.176	-0.40	-2.67	-1.68
Total Problems	380	50.1	-14.923	.000*	-7.518	-0.76	-8.51	-6.53
Pervasive-Developmental Problems	380	54.1	-11.996	.000*	-2.634	-0.52	-4.20	-3.07

* p <.001

Comparisons of Non-Affected Sibling Group and Normative Sample: Child

The non-affected sibling data for the Child t-tests included siblings ages 4.17-11.92-years old (mean = 8.73; SD = 1.75). Some parents of the non-affected siblings

were given CBCL 6-18 for children younger than 6-years old, which is likely due to when the families were screened and then subsequently evaluated. There was one more participant in the Child t-tests of Internalizing and Externalizing (n = 1018) than the remainder of scales (n = 1017) due to missing data with other scales.

In comparison to the normative Child sample, the non-affected sibling group was significantly lower on the following scales: Externalizing, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behavior, and Total Problems. Differences were not significant for the Internalizing and Anxious-Depressed scales. Means and SDs for the non-affected sibling group are depicted in Table 9, and the results of the t-tests are reported in Table 10.

Table 9

Means and SDs for Non-Affected Sibling Group: Child

Scale	Mean	SD
Internalizing	49.39	9.619
Externalizing	47.72	9.482
Anxious-Depressed	54.26	6.160
Withdrawn	53.02	4.819
Social Problems	53.09	4.544
Thought Problems	52.66	4.582
Attention Problems	53.41	5.251
Aggressive Behavior	53.16	5.219
Total Problems	47.36	9.895

Table 10
Comparing Means of Child Non-Affected Sibling Group and Normative Sample

Scale	Sample size	Test Value (Standardization Mean, N = 777)	t	Sig. (2-tailed)	Mean Difference	Effect size	95% Confidence	
							Lower	Upper
Internalizing	1018	50.15	-2.521	0.012	-0.760	-0.08	-1.35	-0.17
Externalizing	1017	50.05	-7.843	.000*	-2.331	-0.24	-2.91	-1.75
Anxious-Depressed	1017	54.20	0.303	0.762	0.059	0.01	-0.32	0.44
Withdrawn	1017	54.30	-8.499	.000*	-1.284	-0.24	-1.58	-0.99
Social Problems	1017	54.40	-9.235	.000*	-1.307	-0.26	-1.58	-1.03
Thought Problems	1017	54.15	-10.392	.000*	-1.493	-0.30	-1.78	-1.21
Attention Problems	1017	54.45	-6.333	.000*	-1.043	-0.19	-1.37	-0.72
Aggressive Behavior	1017	54.20	-6.329	.000*	-1.036	-0.19	-1.36	-0.71
Total Problems	1017	49.80	-7.873	.000*	-2.443	-0.25	-3.05	-1.83

* p <.001

Comparisons of Non-Affected Sibling Group and Normative Sample: Adolescent

The sibling data for the Adolescent t-tests included non-affected siblings ages 12-18.92-years old (mean = 14. 35 and SD = 1.78). There was one more participant in the Adolescent t-tests of Internalizing and Externalizing (n = 492) than the remainder of scales due to missing data with other scales (n = 491).

In comparison to the normative Adolescent sample, the non-affected sibling group was significantly lower on the following scales: Internalizing, Externalizing, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behavior, and Total Problems. The difference was not significant for the Anxious-Depressed scale. Means and SDs for the non-affected sibling group are depicted in Table 11, and the results of the t-tests are reported in Table 12.

Table 11

Means and SDs for Non-Affected Sibling Group: Adolescent

Scale	Mean	SD
Internalizing	47.92	9.856
Externalizing	45.94	9.249
Anxious-Depressed	53.43	5.971
Withdrawn	53.26	4.959
Social Problems	52.81	4.813
Thought Problems	52.78	4.939
Attention Problems	53.47	5.196
Aggressive Behavior	52.65	4.896
Total Problems	46.02	10.259

Table 12

Comparing Means of Adolescent Non-Affected Sibling Group and Normative Sample

Scale	Sample size	Test Value (Standardization Mean, N = 976)	t	Sig. (2-tailed)	Mean Difference	Effect size	95% Confidence Interval of Lower	Upper
Internalizing	492	50.15	-5.026	.000*	-2.233	-0.23	-3.11	-1.36
Externalizing	492	50.10	-9.969	.000*	-4.157	-0.44	-4.98	-3.34
Anxious-Depressed	491	54.05	-2.302	0.022	-0.620	-0.11	-1.15	-0.09
Withdrawn	491	54.50	-5.556	.000*	-1.243	-0.23	-1.68	-0.80
Social Problems	491	54.05	-5.725	.000*	-1.243	-0.24	-1.67	-0.82
Thought Problems	491	54.15	-6.128	.000*	-1.366	-0.26	-1.80	-0.93
Attention Problems	491	54.50	-4.391	.000*	-1.030	-0.18	-1.49	-0.57
Aggressive Behavior	491	54.25	-7. 252	.000*	-1.602	-0.29	-2.04	-1.17
Total Problems	491	49.85	-8.276	.000*	-3.832	-0.38	-4.74	-2.92

* p <.001

Comparisons of Proband Group and Sibling Group: CBCL, All ages

The ASD group was significantly higher than the normative sample on all the scales for all age groups, with the exception of the Pre-School group on the Anxious-Depressed scale. The non-affected sibling group was significantly lower than the normative sample on the CBCL except for the Anxious-Depressed scale for the Child and Adolescent groups and the Internalizing scale for the Child group. These findings indicate that, as expected based on previous research, the proband group scored significantly higher on almost all scales than the non-affected sibling group.

Covariate Analyses

To determine whether age and/or adaptive behavior (as a proxy for IQ) differentially affect CBCL scores of the experimental (proband) group and the comparison (non-affected sibling) group, a MANCOVA with a between groups design and an alpha level of 0.05 was conducted. The covariates age and Vineland-II Composite score were considered to account for dependent variable (DV) variance that is not attributable to the independent variables (IVs). The IVs were the proband group and the non-affected sibling group, subdivided into age groups based on the two CBCL forms, 1.5-5 (Pre-school) and 6-18 (Child and Adolescent).

The analyses for each of the Pre-school and Child and Adolescent age groups will be presented in two phases. First, the multivariate and the univariate analyses for the individual scales are presented. Then, the multivariate and univariate analyses for the composite scales (Internalizing and Externalizing) are presented. The individual scales included the following scales: Anxious-Depressed, Withdrawn, Sleep Problems, Social Problems, Thought Problems, Attention Problems, Aggressive Problems, Total Problems, and Pervasive Developmental Problems. The Sleep Problems and Pervasive Developmental Problems scales are only included in the Pre-school analyses. The Social Problems and Thought Problems scales are only included in the Child and Adolescent analyses. The Anxious-Depressed, Withdrawn, Social Problems, Thought Problems, and Attention Problems scales load on the Internalizing scale. The Aggressive Behavior, Social Problems, Thought Problems, and Attention Problems load on the Externalizing scale.

Despite the Pre-school and Child and Adolescent subdivisions based on the CBCL forms, some of the participants who were as young as age 4-years old were included in the older category (6-18), and some of the participants up through age 6.5 years-old were included in the younger category (1.5-5), based on the form given to the parent as reported in the SFARI database. Removal of these subjects from the data set would have resulted in a >15% loss of the data set so they were included. Overall, all analyses retained 95% of the available data.

MANCOVA: Pre-school

When comparing the Pre-school proband group with the non-affected sibling group for the multivariate analysis, the DVs were the CBCL Total Problems, Anxious-Depressed, Withdrawn, Sleep Problems, Attention Problems, Aggressive Behavior, and Pervasive Developmental Problems scales. The effects of the IV of group (proband versus non-affected sibling) membership on each Pre-school scale was statistically significant, even after controlling for the effects of age and adaptive behavior as a proxy for IQ on the above mentioned scales ($F_{7, 721}$= 23.51, p <.001). Group membership had a medium size of .721. When examining the univariate analyses, group membership also had a significant effect on Anxious-Depressed, Withdrawn, Attention Problems, Total Problems, and Pervasive Developmental Problems scales. Age had a significant effect on Sleep Problems, Aggressive Behavior, and Total Problems scales. Adaptive behavior had no statistically significant effect on scale scores in this analysis. The results are depicted in Table 13.

Table 13

Between-Subjects Effects: Pre-School Individual Scales

IV	Scale	F	Sig.	Effect size
Age	Anxious-Depressed	0.016	0.900	0.005
	Withdrawn	2.876	0.090	0.000
	Sleep Problems	6.813	0.009*	0.007
	Attention Problems	2.407	0.121	0.001
	Aggressive Behavior	11.130	0.001*	0.003
	Total Problems	16.954	0.000*	0.002
	Pervasive-Developmental	3.437	0.064	0.000
VABSII	Anxious-Depressed	0.145	0.703	0.003
	Withdrawn	0.320	0.572	0.007
	Sleep Problems	0.021	0.886	0.001
	Attention Problems	0.001	0.975	0.006
	Aggressive Behavior	0.076	0.782	0.006
	Total Problems	1.425	0.233	0.018
	Pervasive-Developmental	2.493	0.115	0.011
Group	Anxious-Depressed	5.124	0.024*	0.050
	Withdrawn	99.649	0.000*	0.633
	Sleep Problems	1.067	0.302	0.100
	Attention Problems	28.255	0.000*	0.373
	Aggressive Behavior	1.986	0.159	0.152
	Total Problems	30.358	0.000*	0.453
	Pervasive-Developmental	136.368	0.000*	0.678

*p< .05

When examining only the Pre-school Internalizing and Externalizing scales on the multivariate analysis, the effects of the IV of group (proband versus non-affected sibling) membership on both scales were statistically significant, even after controlling for the effects of age and adaptive behavior as a proxy for IQ on the Internalizing and Externalizing scales ($F_{2, 732}$= 87.635, $p < .001$). The effect size for the multivariate analysis was small, .193. When examining the univariate analyses, group membership and adaptive behavior had statistically significant effects on the Internalizing and Externalizing scale scores, but age did not produce a significant effect. See Table 14.

Table 14

Between Subjects Effects: Pre-School Internalizing and Externalizing Scales

Source	Dependent Variable	F	Sig.	Effect size
Age	Internalizing	0.606	0.437	0.001
	Externalizing	1.650	0.199	0.002
Vineland-II	Internalizing	23.612	0.000*	0.031
	Externalizing	33.753	0.000*	0.044
Group	Internalizing	175.089	0.000*	0.193
	Externalizing	62.809	0.000*	0.079

*p< .05

MANCOVA: Child and Adolescent

When comparing the Child and Adolescent proband group and the non-affected sibling group for the multivariate analysis, the DVs were Anxious-Depressed, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behavior, and Total Problems. The effects of the IV of group (proband versus non-affected sibling) membership on each Child and Adolescent scale were statistically significant, even after controlling for the effects of age and adaptive behavior as a proxy for IQ on the above mentioned scales ($F_{7, 3034}$= 681.900, p < .001), with a small effect size, .079. When examining the univariate analyses, group membership had a significant effect on the all the scales in this analysis, with the probands being more impaired. Age had a significant effect on all the DVs, except for Thought Problems, and adaptive behavior had a significant effect on all DVs except for Anxious-Depressed. See Table 15 for results.

Table 15

Between-Subjects Effects: Child & Adolescent Individual Scales

IV	Dependent Variable	F	Sig.	Effect size
Age	Anxious-Depressed	4.865	0.027*	0.002
	Withdrawn	13.944	0.000*	0.005
	Social Problems	25.813	0.000*	0.008
	Thought Problems	1.297	0.255	0.000
	Attention Problems	9.914	0.002*	0.003
	Aggressive Behavior	12.227	0.000*	0.004
	Total Problems	8.490	0.004*	0.003
VABSII	Anxious-Depressed	2.866	0.091	0.001
	Withdrawn	21.601	0.000*	0.007
	Social Problems	11.749	0.001*	0.004
	Thought Problems	5.293	0.021*	0.002
	Attention Problems	16.372	0.000*	0.005
	Aggressive Behavior	12.146	0.000*	0.004
	Total Problems	43.184	0.000*	0.014
Group	Anxious-Depressed	386.476	0.000*	0.113
	Withdrawn	1479.794	0.000*	0.327
	Social Problems	1918.024	0.000*	0.387
	Thought Problems	3536.198	0.000*	0.538
	Attention Problems	2073.749	0.000*	0.406
	Aggressive Behavior	589.649	0.000*	0.162
	Total Problems	2193.589	0.000*	0.419

*p< .05

When examining only the Child and Adolescent Internalizing Problems and Externalizing Problems scales on the multivariate analysis, the effects of the IV of group (proband versus non-affected sibling) membership on both scales were statistically significant, even after controlling for the effects of age and adaptive behavior as a proxy for IQ on Internalizing and Externalizing scale scores ($F_{2, 3046}$= 130.718, p < .001), with a medium effect size, .611. When examining the univariate analyses, age had a statistically significant effect only on the Externalizing scale scores. See Table 16.

Table 16

Between-Subjects Effects: Child and Adolescent Internalizing and Externalizing Scales

IV	Dependent Variable	F	Sig.	Effect size
Age	Internalizing	1.643	0.200	0.001
	Externalizing	59.696	0.000*	0.019
Vineland-II	Internalizing	10.850	0.001*	0.004
	Externalizing	59.873	0.000*	0.019
Group	Internalizing	261.245	0.000*	0.079
	Externalizing	59.696	0.000*	0.021

*p< .05

CHAPTER IV

Discussion

The current study compared individual scale and composite scale scores in a sample of probands and their non-affected siblings to the normative sample of the CBCL to determine if the probands were significantly more impaired than the non-affected siblings and the normative sample in psychosocial functioning/behavior based on CBCL profile scores. In line with most of the existing research, the multiple Bonferroni corrected ($p < .001$) two-tailed *t*-tests indicated that the ASD group means were significantly higher than the normative sample on all the scales for all age groups, with the exception of the Pre-school group on the Anxious-Depressed scale. An unexpected finding, but one that is consistent with social cognitive learning theory, was that the non-affected sibling group displayed significantly fewer maladaptive behaviors than did the normative sample on the CBCL, except for the Anxious-Depressed scale in the Child and Adolescent groups and the Internalizing scale in the Child group. Thus, the probands were significantly more impaired on almost all scales than the non-affected siblings, and the non-affected sibling group was significantly less impaired than the CBCL normative sample on almost all scales. This study also examined whether age and/or adaptive behavior, as a proxy for IQ, differentially affect CBCL scores for the proband and non-affected sibling groups. The multivariate analysis revealed that the proband group had significantly higher CBCL profile scores in comparison to their non-affected siblings, even when controlling for age and adaptive behavior (as a proxy for IQ). The univariate analyses in the Pre-school group revealed age was a significant factor for Sleep Problems,

Aggressive Behavior, and Total Problems scale scores, and impaired adaptive behavior scores were significantly related to impaired Internalizing and Externalizing scores. Group membership (proband versus non-affected sibling) was significantly associated with scores on Internalizing, Externalizing, Withdrawn, Attention, Total Problems, and Pervasive Developmental Problems scales. The univariate analyses in the Child and Adolescent group revealed age as a significant factor to the Externalizing, Anxious-Depressed, Withdrawn, Social Problems, Attentions Problems, Aggressive Behavior, and Total Problems scales. Impaired adaptive behavior scores were significantly associated with impaired Internalizing, Externalizing, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behaviors, and Total Problems scale scores. There also were significant differences between groups on all nine scale scores in relation to group membership (non-affected sibling versus proband).

Next, the findings for each of the three research questions are discussed in the context of the comparability of these findings to the available research. Future research directions are interspersed throughout the next portion of the Discussion. Several important limitations of this study are then discussed.

Research Question 1

This question asked whether the proband group was significantly more impaired than the non-affected sibling group and the normative sample in psychosocial functioning/behavior based on CBCL profile scores. Results are reported in sequence for the Pre-school, Child, and Adolescent groups.

For the Pre-school group, the proband group had significantly higher scores than either group (non-affected sibling and normative) except on the Anxious-Depressed scale. The overall proband group means were higher than the normative sample by 3.15 to 17.42 points, with half of the significant differences in means > 1SD (SD = 10). Because these are large differences, they may have clinically as well as statistically important implications. The proband group Pre-school mean scores for the Withdrawn and Pervasive-Developmental Problems were more than 1.5 SDs higher than the normative sample. This is consistent with expectations because many of the characteristics of ASD fall on these scales, according to the DSM-IV-TR (ApA, 2000).

The proband Pre-school group means were significantly higher than the normative sample on all the scales, with the exception of the Anxious-Depressed scale. This may reflect the established finding that parents report lower levels of depression and other internalizing symptoms in children than the children/adolescents report themselves (Klein, Dougherty, & Olino, 2005); examining psychosocial self-report of children with siblings of autism may be a future direction of research.

A supplemental analysis indicated that the non-affected sibling group scored significantly lower on the Anxious-Depressed scale than the normative sample and the proband sample ($p < .001$). These findings indicate that the proband group was significantly more impaired than the non-affected sibling group. These findings are fairly consistent with the findings of Bolte et al. (1999), in which most of the autistic group means (Total Problems, Social Problems, Thought Problems, Attention Problems, Withdrawn, Anxious/Depressed, Delinquency, and Aggressive Behavior) were

significantly higher than the normative CBCL sample (based on the German version of the CBCL 4-18).

For the Child group, the proband group had significantly higher scores than either group (non-affected sibling and normative). The proband group overall obtained significantly higher means than the normative sample by 4.98 to 13.28 points, with 1/3 of the differences in means > 1SD (SD = 10). Because these are large differences, they may have clinically as well as statistically important implications. Mean score differences in the 1-1.5 SD range emerged on the Child Thought Problems and Attention Problems scales. This is consistent with expectations because many of the characteristics of ASD fall on these scales, according to the DSM-IV-TR.

The proband Child group means were significantly higher than the normative sample on all the scales, and the non-affected sibling group was significantly lower than the normative sample on the CBCL except for the Anxious-Depressed scale and the Internalizing scale. Because the sibling group mean was only .06 points higher than the normative mean on the Anxious-Depressed scale, and lower than the normative mean on the Internalizing scale, we can infer that the proband group was significantly more impaired than the non-affected sibling group on all scales. These findings are consistent with the findings of Bolte et al. (1999), in which most of the autistic group means (Total Problems, Social Problems, Thought Problems, Attention Problems, Withdrawn, Anxious/Depressed, Delinquency, and Aggressive Behavior) were significantly higher than the normative CBCL sample (based on the German version of the CBCL 4-18).

For the Adolescent group, the proband group had significantly higher scores than either group (non-affected sibling and normative). The proband group obtained

significantly higher means than the normative sample by 4.51 to 13.32 points, with 55.6% of the significant differences in means > 1SD (SD = 10). Because these are large differences, they may have clinically as well as statistically important implications. Mean score differences in the 1-1.5 SD range for the Adolescent proband group emerged on the Social Problems and Thought Problems scales. These scales contain items that encompass characteristics of ASD, according to the DSM-IV-TR.

The Adolescent proband group means were significantly higher than the normative sample on all the scales, and the non-affected sibling group was significantly lower than the normative sample on the CBCL except for the Anxious-Depressed scale. Thus, the proband group also was significantly more impaired than the non-affected sibling group on all scales. These findings also are consistent with the findings of Bolte et al. (1999), in which most of the autistic group means (Total Problems, Social Problems, Thought Problems, Attention Problems, Withdrawn, Anxious/Depressed, Delinquency, and Aggressive Behavior) were significantly higher than the normative CBCL sample (based on the German version of the CBCL 4-18).

In applying social cognitive learning theory to the non-affected siblings, the results from the current study suggest that non-affected siblings may have learned to inhibit negative behaviors from observing the probands exhibiting maladaptive behaviors and the consequences that follow. This information could be useful in clinical practice and will be discussed in greater detail later in this Discussion. On the other hand, the results could be an artifact of the criteria of the study. Families were screened in a very conservative way that would exclude several youth that may have some type of disability. The genetics component of the data collection was attempting to get a “clean” family

sample, so if any non-affected siblings were referred for anything, they were not represented in this sample.

Research Question 2

This question asked whether the non-affected sibling group was more impaired than the normative sample in psychosocial functioning/behavior based on CBCL profile scores. Results are reported for the Pre-school, Child, and Adolescent age groups.

For the Pre-school group, the non-affected sibling group had significantly lower scale scores in comparison to the normative group on all scales. The mean differences between the non-affected sibling group and the normative sample ranged from -7.52 to -1.75 (< 1SD), with non-affected sibling scores lower than the normative scores. This finding indicates that the non-affected sibling group actually exhibited fewer behavior problems than did the CBCL normative sample. In applying social cognitive learning theory to non-affected siblings, the results from the current study suggest that non-affected siblings may have learned to inhibit negative behaviors from observing probands exhibiting maladaptive behaviors and the consequences that follow.

For the Child group, the non-affected sibling group had significantly lower scale scores in comparison to the normative group on all scales, with the exception of the Anxious-Depressed and Internalizing scales. As noted previously, parent report tends to be lower in levels of depression and other internalizing symptoms in children than children/adolescents report themselves (Klein et al., 2005); examining psychosocial self-report of non-affected children with siblings with ASDs may be a future direction of research to further elucidate this possibility.

The mean differences between the non-affected Child sibling group and the normative sample ranged from -2.44 to -1.04 (< 1SD), with non-affected sibling scores lower than the normative scores. This finding indicates that the non-affected sibling group exhibited fewer behavior problems than did the CBCL normative sample. In applying social cognitive learning theory to non-affected siblings, the results from the current study suggest that non-affected siblings may have learned to inhibit negative behaviors from observing probands exhibiting maladaptive behaviors and the consequences that follow.

For the Adolescent group, the non-affected sibling group had significantly lower scale scores in comparison to the normative group on all scales, with the exception of the Anxious-Depressed scale. There was no difference between the non-affected Adolescent sibling group and the normative group on the Anxious-Depressed scale, possibly due to the aforementioned tendency of parents to report lower levels of depression and other internalizing symptoms than do children/adolescents report themselves (Klein, Dougherty, & Olino, 2005).

The mean differences between the Adolescent non-affected sibling age group and the normative sample ranged from -4.16 to -1.03 (< 1SD), with non-affected sibling scores lower than the normative scores. This finding indicates that the non-affected sibling group actually exhibited fewer behavior problems than did the CBCL normative sample. As noted previously, non-affected siblings may have learned to inhibit negative behaviors from observing probands exhibiting maladaptive behaviors and the consequences that follow, consistent with the observational and vicarious learning components of social cognitive learning theory.

These findings also provide suggestive support for the outcomes reported by Kaminsky and Dewey (2002). They found that higher levels of social support of siblings of children with autism were related significantly to lower levels of loneliness and higher academic functioning (CBCL School Performance scale). Additionally, siblings of children with autism whose parents were involved in a support group exhibited less internalizing, externalizing, and total adjustment problems on the CBCL than siblings whose parents did not attend support groups. Despite these significant results, none of the mean scores on the CBCL scales were clinically significant for siblings of children with autism, so the results may not have generalizable clinical significance. Yet, the results are similar to the current study in that the siblings had no clinically significant scores. The siblings of children with autism in Kaminsky and Dewey's study seemed to be psychosocially well-adjusted to their environment and did not internalize or externalize the negative behaviors that their sibling with autism may be exhibiting.

Rodrigue et al. (1993) also did not find significant CBCL Internalizing and Externalizing scores for siblings of children with autism, even with their sample only including siblings of children with severe autism from predominantly middle to upper-middle class. However, in contrast to the current findings, the siblings of children with autism in the Rodrigue et al. study had significantly higher scores (i.e., more impaired) than the siblings of mentally age-matched, typically developing children.

Like Rodrigue et al. (1993) and the current study, Gold (1993) did not find clinically significant CBCL Internalizing and Externalizing scores for non-affected siblings. However, the sampling and demographic differences between the Gold study and the current study are worth noting. Gold had a small sample size and older subjects

(22 siblings of boys with autism, with a mean age of 13.52, and 34 siblings of nondisabled boys with a mean age of 12.78). The mean age for the current study was between 9 and 9.5-years old for the ASD and sibling groups. Level of parent education in Gold's study also differed from the current study, with 31-38% for Gold's group having baccalaureate or higher, whereas the current study had about 60% at baccalaureate or higher. In regards to parental income, Gold's study had 42% of families < $39K in the siblings of boys with autism group and 13% of families < $39K in the siblings of nondisabled boys, whereas most families' income was above $36K in the current study. Also, the individuals with autism were diagnosed using the criteria of the DSM-III-R, rather than the ADOS and ADI-R, which were based on DSM-IV (1994) criteria; this difference is due to the date of the research and the availability of resources at the time. These sample differences reduce the comparability between the studies, thus reducing the comparability of the results.

Despite the fact that the CBCL scales used in the Gold (1993) and Rodrigue et al. (1993) studies were not the same as those used in the current study, taken together the results suggest that non-affected children may learn to avoid exhibiting the maladaptive behaviors that their siblings are exhibiting, as social cognitive learning theory would predict. This may be due to parental discipline approaches that include appropriate consequences for maladaptive behaviors, modifying inappropriate behaviors, and modeling and shaping positive replacement behaviors. One direction for future research would be to study parental discipline/behavior in families with children with ASD to determine whether parental disciplinary approaches correlate with increased or decreased levels of maladaptive sibling behavior. According to social cognitive learning theory

comparatively more positive sibling behavior would be expected to be associated with increased parental efficacy and consistency of discipline in addressing maladaptive behavior in children with ASD.

Although the current findings were consistent with several prior studies, inconsistencies also are apparent. Pilowsky et al. (2004), in another study that utilized the CBCL with siblings of children with autism, looked at a summed general behavior problems scores of a Hebrew version of the CBCL. Most (86.7%) of the small sample of non-affected siblings were high-functioning (did not have clinically significant scores). They found that siblings of nonverbal children with autism had more behavior problems and lower socialization skills, based on the Vineland (Sparrow, Balla, & Cicchetti, 1984) in the Social Domain, than siblings of verbal children with autism. Pilowsky et al.'s small sample had a much lower mean IQ for the probands (mean = 56.32, SD = 21.78) in comparison to the current study (mean = 82; SD = 27.8). When looking at nonverbal and verbal probands, it would be helpful in future research to also examine IQ of the non-affected siblings and the probands if the data are available.

An implication of Pilowsky et al.'s (2004) finding about nonverbal children with autism and the behavior problems of their non-affected siblings from the standpoint of social cognitive learning theory would be that the typically developing siblings who have a nonverbal sibling with autism have learned negative ways of coping with their environmental and self-imposed demands (Thomas, 2005). Future studies with a large sample size could examine whether there is a difference in the psychosocial development of the non-affected siblings from the normative population when examining verbal versus nonverbal siblings with autism/ASD. If similar results are obtained, then it would be

important for parents of nonverbal children with autism/ASD to be aware of this information for psychological treatment purposes of their non-affected children.

Contrary to the current study, and from several other studies reviewed (Bagenholm & Gillberg, 1991; Gold, 1993; Pilowsky et al., 2004; Kaminsky & Dewey, 2002; Rodrigue et al., 1993), Hastings (2003) found that a sample of non-affected siblings of children with autism were rated as having more peer problems, more overall adjustment problems, and lower levels of prosocial behavior as compared to a normative sample of British children on the SDQ. The sample size of the non-affected siblings of children with autism was small (n = 22); the children with autism attended a school for children with autism. One caveat is that 11 children with autism were weekly boarders at the school so they did not have as much family contact. Another caveat is that all the children with autism had MR. Additionally, not all families were of 2-parent households.

Research Question 3

This question asked whether age and/or IQ differentially affect CBCL scores for the proband and non-affected sibling groups. Results are reported for the Pre-school MANCOVA multivariate effect, followed by the Pre-school MANCOVA univariate effects. Then the results for the Child and Adolescent multivariate effect will be reported, followed by the Child and Adolescent univariate effects.

The Pre-school multivariate analysis revealed that the proband group had significantly higher CBCL profile scores in comparison to their non-affected siblings, even when controlling for age and adaptive behavior (as a proxy for IQ). When looking at Pre-school univariate effects, age was a significant finding for Sleep Problems,

Aggressive Behavior, and Total Problems scale scores. Young children may exhibit aggressive, nonverbal behavior when they do not have sufficient insight and vocabulary to express themselves verbally or have not learned yet how to control their emotions. Additionally, in the Pre-school grouping, adaptive behavior was significantly associated with Internalizing and Externalizing scale scores; the higher (or the more impaired) the adaptive behavior scores are, the higher (or the more impaired) the Internalizing and Externalizing scores. Future research could examine the differences between verbal and nonverbal probands and their non-affected siblings to determine if significant differences exist between non-affected siblings of these two proband groups.

In the Pre-school grouping, group membership (proband versus non-affected sibling) was significantly associated with scores on the following scales: Internalizing, Externalizing, Withdrawn, Attention, Total Problems, and Pervasive Developmental Problems. Similarly, Sikora, Hall, Hartley, Gerrard-Morris, and Cagle (2008) found that the CBCL scales distinguished among the ADOS-G classification groups. Sikora et al. found a significant group effect for CBCL 1.5-5 Withdrawn and Pervasive Developmental Problems, but not for Aggressive Behavior or Anxious-Depressed. They also used the GARS as a DV, besides the CBCL scales, and found that the Preschool CBCL Withdrawn and Pervasive Developmental Problems scales had better sensitivity than the GARS in distinguishing children with autism or without autism. Additionally, the CBCL Withdrawn scale had better specificity than the GARS. Coupled with the Sikora et al. findings, as well as findings from other studies (Bolte et al.,1999; Pandolfi, Magyar, & Dill, 2009; Rescorla, 1988), the current study findings suggest that the CBCL could be a useful screener in schools, where the ADOS and ADI-R are not so readily

available due to financial reasons and the amount of time it takes to complete the ADOS and ADI-R. Future studies with a more representative sample should further examine the validity of the CBCL as a screener for probands.

Adaptive behavior was not significantly related to any of the scales except for Internalizing and Externalizing scales for the Pre-school age grouping in the current study. Age had no relation to Internalizing and Externalizing scale scores. Additionally, age was not significantly associated with Attention Problem scores, which may be due to younger children being more easily distracted as they are beginning to grow and explore their environments.

The multivariate analysis for the Child and Adolescent group revealed that the probands had significantly higher CBCL profile scores in comparison to their non-affected siblings, even when controlling for age and adaptive behavior (as a proxy for IQ). When examining the univariate effects in the older age grouping (Child and Adolescent), age was significantly related to the Externalizing, Anxious-Depressed, Withdrawn, Social Problems, Attentions Problems, Aggressive Behavior, and Total Problems scales. Adaptive behavior was significantly associated with Internalizing, Externalizing, Withdrawn, Social Problems, Thought Problems, Attention Problems, Aggressive Behaviors, and Total Problems scale scores. There were significant differences between groups on all nine scale scores in relation to group membership (non-affected sibling versus proband).

Age was not related to the Internalizing and Thought Problems scale scores, which is contrary to Rodigue et al.'s (1993) findings that older non-affected siblings of children with autism were likely to have more internalizing and externalizing problems

than younger non-affected siblings based on CBCL scores. Rodrigue et al.'s small sample size and inclusion of only non-affected siblings of children with severe autism (as opposed to a range of symptom severity) may be linked to this difference in results.

In comparison to the current study, Verte, Roeyers, and Buysse (2003), who subdivided their small sample of non-affected siblings of a child with high functioning autism (HFA) and the control group of siblings of children without a disorder into two age groups of 6-11-years old and 12-16-years old, found the non-affected siblings of children with HFA between 6 and 11-years old had more internalizing and externalizing behavior problems (significant interaction effect of group and age) than those non-affected siblings who were 12-16-years old. However, the CBCL scores were not clinically significant. These results were contrary to Hastings (2003) and Rodrigue et al. (1993), in which older children were more likely to have internalizing and externalizing problems than the younger children. The children with HFA in Verte et al.'s (2003) sample were enrolled in a semi-residential treatment program, where they spent weekends and holidays at their families' homes, and they all had IQ >80. It might have been helpful to know which non-affected children were older versus younger than their sibling with autism, which may impact their self-efficacy and how they adapt to their environmental demands. Future research could examine whether the proband being older or younger than the non-affected sibling affects the social cognitive development of the non-affected sibling. Future studies with larger samples could also compare two school-age groups of non-affected siblings and probands (i.e., 6-11 and 12-18) to further examine if there are statistical differences. Additionally, use of IQ scores would be

helpful in looking at psychosocial and behavioral differences between probands and their non-affected siblings; IQ data were not available for the non-affected siblings.

Similar to the current study's multivariate findings on the Pre-school and Child and Adolescent groups, Bolte et al., (1999) found group differences (between an autistic sample, clinical sample, and German CBCL 4-18 normative sample), when age and IQ were covaried. In the Bolte et al. study, however, the generalizability of the IQ findings should be considered suspect because only one-half of the subjects had actual IQ scores available. The other half (n = 38) of the participants' IQ scores were derived from a 1996 German version of the Vineland and school achievement. Additionally, how the 1996 German version of the first version of the Vineland compares to the Vineland-II is not known.

Another difference to note from the current study and Bolte et al.'s (1999) study is that the three groups in Bolte et al.'s study were the autistic sample, clinical sample, and the normative sample, whereas the current study's three groups were ASD (the spectrum of disorders as opposed to only autistic disorder), non-affected siblings of the individuals with ASD, and the normative sample, with only the ASD and non-affected sibling groups included in the multivariate analyses (MANCOVA). The autistic sample was diagnosed with autism using a German version of the ADI-R (Poutska, Lisch, Ruhl, Schmotzer, & Werner, 1996) for Bolte et al.'s study, whereas the ADOS and the ADI-R were used for diagnosis in the current sample. The differences in the sampling between the two groups reduce the external validity of the studies, meaning that the comparability of the results is reduced more than if the samples were more closely matched.

While Bolte et al. (1999) and the current study found a significant group effect, the differences between the normative sample (of the 1996 German version of Vineland) and the autistic sample were greater (>2 SD) with Bolte et al.'s study than with the current study. Another important difference to examine in comparing the current study to the Bolte et al. (1999) study is the demographic details. Bolte et al.'s study was smaller in sample size with the autistic sample including 54 (70.1%) males and 23 (29.9%) females between 4-18-years old, as opposed to 86.8% males and 13.2% females in the proband sample in the current study of 1,898 probands. Bolte et al. only used the German version of the CBCL 4-18, as opposed to the current study that used the current (2001) versions of the CBCL: 1.5-5 and 6-18. The IQ range (20-128; Mean = 61.3; SD = 25.9) in the Bolte et al. study was different than in the current sample, which required probands who were 4-years old to have a nonverbal deviation or ratio IQ score greater than or equal to 60. Participants between the ages of 5 and 8-years old were required to have a nonverbal deviation score or nonverbal ratio IQ score greater than or equal to 40. Participants 8-years old or older needed a nonverbal mental age of 36-months or older. The differences in the sampling between the two groups reduce the external validity of the studies, meaning that the comparability of the results is reduced more than if the samples were more closely matched.

Lastly, another important aspect to examine in comparing the current study to Bolte et al.'s study is the scales used to compare the groups within each study. Bolte et al. did not use all of the same scales as the current study, although most of the scales were similarly named. They used Withdrawn, Somatic Complaints, Anxious-Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, and Aggressive

Behavior. Two of the scales (Somatic Complaints and Delinquent Behavior) Bolte et al. used were not used in the current study, and the additional scales that were used in this study were Internalizing, Externalizing, and Total Problems for the Child and Adolescent group. The Child group in this study also used Sleep Problems and Pervasive Developmental Problems scales. Because the normative samples are different between the Bolte et al. study and the current study (two versions of the CBCL) and there was a slight difference in the scales used between the two studies, the external validity and comparability of the two groups are reduced more than if the versions of the CBCL and scales used were the same.

Additionally in regards to the significance of age and contrary to the current study, Hastings (2003), who used a small sample of 4-16-year old siblings of 7-16-year old children with autism, found non-affected siblings older than their sibling with autism had better social adjustment based on the SDQ. While the SDQ domains were significantly higher than the proportion expected, the scores were not clinically significant, just like the CBCL mean scores of the current study. The difference between the non-affected sibling group older than their sibling with autism and the non-affected sibling group younger than their sibling with autism may be reflective of the non-affected siblings' adjustment to their environment and possibly to differences in parental discipline and modeling and shaping of prosocial behaviors. The non-affected siblings who are older than their siblings with autism, depending on the age difference, may have already learned many prosocial skills prior to the child with autism's birth, and the non-affected siblings who are younger than their sibling with autism may be more likely to pick up modeled behavior of the sibling with autism.

Limitations

One of the limitations of the study was the demographic constitution of the SFARI sample. Socioeconomic status (SES) and level of parent education was not representative of the general population. Over half of the parents were college educated (baccalaureate or graduate level). Most parents at least completed high school. The high level of education characteristic of the parents in this sample limits the external validity (i.e., generalizability) of the findings of this study. Parents who are college educated are more apt to know about resources or where to seek resources for their child with an ASD. To add to this, most families' income was between $36K and >$161K, also a not representative of the general population. Families with middle to high income and college educated parents are also more likely to expose the probands, besides their other children, to more educational resources, (e.g., museums, vacations, book, and toys) which may impact the child's intellectual and adaptive development. Also, families were screened in a very conservative way that would exclude several youth that may have some type of disability. The genetics component of the data collection was attempting to get a "clean" family sample, so if any non-affected siblings were referred for anything, they were not represented in this sample.

Another limitation was that IQ scores were not available for the non-affected sibling group. IQ scores would have been a preferable covariate to adaptive behavior as a proxy. IQ may have a different relation to CBCL scores between groups than adaptive behavior scores, as Sparrow, Balla, and Cicchetti (1996) found a low correlation between the WISC-III and the Vineland-II. If IQ data are available in the future for the non-

affected sibling group through SFARI SSC, those data should be substituted for adaptive behavior scores as covariates in future studies.

Another limitation is in regards to the CBCL normative population. The CBCL manual indicated that participant eligibility criteria were children at least18-months old and to have no major physical disabilities or mental retardation. Additionally, children whose parents reported that the child had received mental health services, services for substance abuse problems, and/or special education services in the past 12 months were excluded from the norm sample. Thus, although some children with diagnosable conditions and significant maladaptive behaviors were excluded from the norm group, others that exhibited maladaptive behaviors but had not received services likely were included. However, the CBCL normative sample was more representative of the normative population in relation to demographics, which is contrary to the non-representative demographics of this study. Thus, the normative sample scores may have been higher because they included a higher proportion of children with maladaptive behavior, compared to those in the SFARI sample. It is possible that the significantly lower CBCL scores for the non-affected siblings may have been an artifact of the CBCL sampling method rather than reflecting the influence of the observational and vicarious learning components of social cognitive learning theory on maladaptive behavior.

Summary

The current study compared a sample of probands and their non-affected siblings, to the normative sample of the CBCL to determine if the proband group was significantly more impaired than the non-affected sibling group and the normative sample in

psychosocial functioning/behavior based on CBCL profile scores. The ASD group means were significantly higher than the normative sample on all the scales for all age groups, with the exception of the Pre-School Anxious-Depressed scale, and the non-affected sibling group was significantly lower than the normative sample on the CBCL except on the Anxious-Depressed scale for the Child and Adolescent groups and the Internalizing scale for the Child group. Thus, the proband group was also significantly more impaired on almost all scales than the non-affected sibling group.

Lastly, this study examined whether age and/or IQ differentially affect CBCL scores for the proband and non-affected sibling groups. The multivariate analysis revealed that the proband group, overall and across the age range, had significantly higher CBCL profile scores in comparison to their non-affected siblings, even when controlling for age and adaptive behavior (as a proxy for IQ).

These findings for the all three research questions, coupled with previous studies, support the role of social cognitive learning theory in influencing sibling behavior of non-affected siblings in families of children with ASD. Siblings from the SFARI sample seem to be learning prosocial behaviors despite, or perhaps because of, having a sibling with an ASD. However, the unique characteristics of the SFARI sample, coupled with the methodological limitations of this study limit the generalizability of the findings.

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