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ORIGINAL PAPER

Minimal Risk of Internalizing Problems in Typically-Developing Siblings of Children with High-Functioning Autism Spectrum Disorder

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Abstract We examined anxiety and depression clinical symptom levels in 42 typically-developing (TD) siblings of children with high-functioning autism spectrum disorder (HFASD), compared to 42 TD siblings of TD children, matched on age, ethnicity, and gender. Participants were between the ages of 6 and 16. Groups were compared using the parent-report and child self-report versions of the Behavior Assessment System for Children, Second Edition, anxiety and depression clinical scales. Within-group differences were examined for parent versus child ratings. Multivariate procedures did not support clinically elevated levels of internalizing problems in siblings of children with HFASD. Results from both sources were consistent with the overall finding. The findings do not support an increased need for targeted intervention, though clinical and school-based practitioners should attend to clinical warning signs if presented.

Keywords Autism spectrum disorder · High-functioning · Depression · Anxiety · Siblings

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Introduction

High-functioning autism spectrum disorder (HFASD) is characterized by social interaction and social communication deficits, and restricted and repetitive behaviors and interests (American Psychiatric Association [APA] 2013), in the context of average to above-average cognitive and language ability (Klin and Volkmar 2000). Despite these strengths, children with HFASD can demonstrate behavioral problems associated with autism spectrum disorder (ASD), such as disruptive behavior, stereotyped or fixated interests, and poor social-emotional reciprocity (APA 2013). These social and behavioral problems likely increase caregivers' and other family members' stress levels (Lainhart 1999; Portway and Johnson 2005; Lee et al. 2009). For example, the increased stress and interpersonal demands on parents of a child with HFASD can put parents at-risk for internalizing problems, including anxiety and depression (for a meta-analysis including studies of children with HFASD, see Hayes and Watson 2013). Further, caregivers of children with HFASD show lower levels of adaptive coping (Lee et al. 2009). In addition, despite their cognitive and language ability, many children with HFASD continue to require support into adulthood (Portway and Johnson 2005). Thus, it is not surprising that a majority of research has focused on caregiver stress and coping. However, the potential impact of the behaviors of a child with HFASD on the emotional function of siblings should not be ignored. Current research has failed to provide a consensus regarding internalizing problems (i.e., clinically significant anxiety and depression) among siblings of children with HFASD (Maeden et al. 2010). If this is identified as a critical concern, the development of targeted interventions and an increased focus on these children should become a priority for

community and school-based mental health providers. This is also an important consideration because of the potential role siblings can play in enhancing social development for children with ASD (Ferraioli et al. 2012; Rivers and Stoneman 2008).

There is a paucity of research on internalizing difficulties of siblings of children with HFASD. Most available research focuses on siblings of children with ASD comorbid with intellectual disability (ID) or across a broad range of cognitive functioning levels. Reviews describe a conflicted field of findings (see Maeden et al. 2010; Orsmond and Seltzer 2007). For example, Maeden et al. (2010) identified 12 studies examining the social, emotional, and behavioral characteristics of typically-developing (TD) siblings of children with ASD. No consistent finding regarding internalizing difficulties of siblings of children with ASD was identified; however, demographic factors such as age, gender, and parent resources were identified as potentially important. Reviews also suggest that study specific characteristics such as type of comparison group may be contributing to the inconsistent results (Maeden et al. 2010; Orsmond and Seltzer 2007). Some studies compare TD siblings of reference children with ASD to TD siblings of other TD children, to TD siblings of children with other developmental disabilities, or to the normative sample for the measure used. Further, internalizing problems have been addressed separately as anxiety or depression problems, and as a combined construct (i.e., internalizing or emotional problems).

Recent studies specifically examining anxiety in siblings of children with ASD have shown minimal evidence of elevated concerns. For example, Pollard et al. (2013) used a self-report measure of anxiety (the Multidimensional Anxiety Scale for Children; March et al. 1997) in siblings of children with ASD. Compared to TD siblings of children with Down's syndrome, while siblings of children with ASD reported more problems in the sibling relationship, they did not show elevated levels of anxiety. Consistent with this finding, Shivers et al. (2013) used a parent-reported anxiety subscale from the Child Behavior Checklist (CBCL; Achenbach and Rescorla 2001) in a large sample from the Simons Simplex Collection database. While they also found no heightened anxiety overall, they found male siblings were more likely to be in the borderline or clinical range than females and that anxiety levels correlated to a parental history of anxiety and behavior problems in the reference sibling with ASD.

While recent studies focused on depression are largely absent, older studies have shown a mixed risk of depression in TD siblings of children with ASD. For example, Gold (1993) and Macks and Reeve (2007) both compared TD siblings of children with ASD to comparison groups of TD siblings of TD children. Gold (1993) found elevated levels of depression in the siblings of children with ASD, though no specific link to a gender, behavior, or family factor were identified. In contrast, Macks and Reeve (2007) did not identify an elevated risk of depression and was unique in using both self-report (Children's Depression Inventory-Short Form; Kovacs 1992) and parent-report (Behavior Assessment System for Children; Reynolds and Kamphaus 1992) indicators of depression. This study also identified a heightened positive self-concept in TD siblings of children with ASD, relative to the comparison siblings, when demographic risk factors (such as low economic resources) were minimal.

In addition to studies looking at anxiety and depression specifically, other researchers have examined internalizing problems broadly using composite scores. Three recent studies used the internalizing problems composite from the CBCL (Barak-Levy et al. 2010; Dempsey et al. 2012; Tomeny et al. 2012). None of these studies found an increased mean level of internalizing problems among TD siblings of children with ASD, compared to either the normative sample (Dempsey et al. 2012) or a comparison group of TD children with TD siblings (Barak-Levy et al. 2010; Tomeny et al. 2012). However, Barak-Levy et al. (2010) identified reduced participation in social and extracurricular activities in the TD siblings of children with ASD and interpreted this as a tentative indication of problems with emotional adjustment.

The emotional symptoms composite from the Strengths and Difficulties Questionnaire (SDQ; Goodman 1997) has also been used to identify internalizing problems in siblings of children with ASD. This measure has an advantage over the CBCL as it can be completed by either a parent or the child. Hastings and Petalas (2013) found no elevation for self-reported levels of emotional symptoms of siblings of children with ASD compared to the normative sample for the SDQ. In contrast, using the parent report of the SDQ, Meyer et al. (2011) found elevated emotional symptoms in siblings of children with ASD compared to the normative sample. This difference in results across informants highlights a possible informant contrast that supports the use of multiple sources of information when evaluating for these concerns (Weisbrot et al. 2005).

In contrast to studies comparing TD siblings of reference children with ASD and ID, or in mixed-function samples, studies focusing exclusively on siblings of children with HFASD are largely absent. Only three studies in the current literature focus on this group and these vary in findings and are limited in quality. All three are broadly limited by only using parent report information from the internalizing problems composite of the CBCL (Achenbach and Rescorla 2001). Verté et al. (2003) examined the psychosocial functioning of 29 TD siblings of children with HFASD compared to 29 TD children with TD siblings.

Reference children with HFASD had average cognitive ability (IQ > 80), but a consistent measure was not used and descriptive values were not reported. Results indicated higher average levels of internalizing problems for both male and female siblings aged 6–11 compared to the TD group. However, the authors did not conclude there was an elevated risk of internalizing problems because these scores did not fall into the clinical range (identified in this study as CBCL internalizing problems T-score > 67). In contrast to the interpretation of Verté et al. (2003), Ross and Cuskelly (2006) concluded there was an elevated risk of internalizing problems in a sample of 25 TD siblings of children with HFASD. In this study, despite not finding an elevated mean score difference, 40 % of the sample exceeded the clinical range (identified in this study as CBCL internalizing problems T-score > 60). While identified as high functioning, no specific information on cognitive ability of the reference children with HFASD was provided. Finally, Rao and Beidel (2009) examined a subsample of seven TD siblings of children with HFASD in comparison to eight TD siblings of TD children. While the main focus of the study was parenting stress and factors related to family function, sibling internalizing problems were reported to be close to the population mean but higher than those of the TD siblings of TD children. A single measure of cognitive function was used to screen for reference child cognitive ability (inclusion required IQ > 85) though no specific mean score was reported.

In summary, the existing research on internalizing problems of siblings of children with ASD/HFASD has been mixed. Some researchers have described an elevation of internalizing problems, potentially warranting intervention, while others have found no elevation. Given limited resources for school and community-based mental health providers, this risk needs to be clearly determined. In addition, the research on siblings of children with HFASD is especially limited.

The current study improves on the existing literature by including a comparison group of participants matched on demographic variables, using a well-validated measure of internalizing problems, describing the functional level of reference siblings with HFASD, and consulting multiple informants. The purpose of the current study is to evaluate the clinical need for targeted psychosocial interventions for siblings of children with HFASD by testing the hypothesis that internalizing symptoms (anxiety and depression) of TD siblings are elevated compared to those of TD children with TD siblings. To test this hypothesis, between-group comparisons were conducted for parent ratings and child self-reports. In addition, we expect within-group differences (parent-report vs. self-report) to be consistent.

Method

Participants

A total of 84 children, ages 6–16, were target participants in the current study. Within the total sample were two groups: 42 TD siblings of children with HFASD (HFASD-siblings group) and 42 TD siblings of TD children (TD-siblings group). For the purpose of clarity, the participants will be referred to as "target siblings" and the brother or sister allowing them to qualify for the study (with or without HFASD) as "reference children." The target siblings across the two groups were individually matched on age (within 2 years), gender, and ethnicity (majority/minority). Matching was confirmed on these variables. Target sibling age was not different between groups ($t[82] = 0.60$, $p = 0.551$) and averaged 9.52 ($SD = 2.80$) years in the HFASD-siblings group and 9.86 ($SD = 2.28$) years in the TD-siblings group. Target siblings were 57.1 % male and 97.6 % Caucasian in both groups. Both the target sibling and reference child in each pair attended school in the same school district. In addition, none of the reference children with HFASD attended specialized facilities for individuals with developmental disabilities.

To qualify, target siblings had to have a reference brother or sister between the ages of 5 and 18 with either a diagnosis of ASD and cognitive function in the average range (confirmatory procedures are described below) or no identified developmental, psychiatric (except anxiety or depression), or educational conditions.

The target siblings for the HFASD-siblings group were recruited from families of children with HFASD who participated in various treatment trials. Potential target siblings were screened by parent questionnaire and excluded if there was evidence of developmental delays, psychiatric conditions (other than anxiety or depression), or educational needs. Inclusion criteria and procedures for the recruitment of the reference children with HFASD have been described in previous publications (for example, see Lopata et al. 2010 or 2015). At minimum, a prior diagnosis of an ASD (autism, Asperger's, or PDD-NOS; APA 2000) confirmed from a licensed physician or psychologist and a Wechsler Intelligence Scales for Children-Fourth Edition (WISC-IV; Wechsler 2003) short-form IQ score >70 (with a Verbal Comprehension Index [VCI] or Perceptual Reasoning Index [PRI] score ≥80) were required. In addition to the minimum requirements, thirty-three (78.6 %) of the reference children with HFASD also had their ASD diagnoses confirmed via the Autism Diagnostic Interview-Revised (ADI-R; Rutter et al. 2003). A summary of the reference children with HFASD's WISC-IV and ADI-R scores are provided in Table 1.

Table 1 Functional characteristics of the reference children with HFASD

WISC-IV ($n = 42$)	
Short-form IQ	101.24 (13.88)
Short-form VCI	101.01 (13.25)
Short-form PRI	101.21 (16.64)
ADI-R ($n = 33$)	
Reciprocal social interaction	21.94 (5.44)
Social communication	15.55 (4.42)
Restricted, repetitive, and stereotyped behaviors	5.85 (1.97)

WISC-IV Wechsler Intelligence Scale for Children-4th Edition; *VCI* Verbal Comprehension Index; *PRI* Perceptual Reasoning Index; *ADI-R* Autism Diagnostic Interview-Revised

The TD-siblings group was recruited through public advertisements and interested families were interviewed for the presence of developmental, psychiatric, or educational conditions among all children present in the home. Inclusion criteria were no child in the household identified with developmental, psychiatric (other than anxiety or depression) or educational conditions.

Though not evaluated in order to test the hypotheses of the current study, additional characteristics of the reference children and participating families were identified and compared. Reference child age did not differ between groups ($t[82] = 1.73$, $p = 0.088$) and averaged 10.52 ($SD = 1.82$) years in the HFASD-siblings group and 9.42 ($SD = 3.67$) years in the TD-siblings group. The average age-difference between target and reference siblings ranged from one to six years. Age-difference did not differ between groups ($t[82] = 0.51$, $p = 0.611$) and averaged 2.64 ($SD = 1.30$) years in the HFASD-siblings groups and 2.79 ($SD = 1.26$) years in the TD-siblings group. However, while average age-difference was not statistically significant, proportion of birth orders did differ between groups (Exact test two-tailed $p = 0.028$), with 35.7 % of target siblings older in the HFASD-siblings group and 61.9 % of the target siblings older in the TD-siblings group. Gender proportion also differed for the reference children (Exact test two-tailed $p < 0.001$), with 85.7 % male in the HFASD-siblings group and 45.2 % male in the TD-siblings group. While differing across groups, this gender disparity is to be expected given the gender difference in the prevalence of ASD, with males substantially outnumbering females (APA 2013).

Family characteristics of socioeconomic status and family size were also collected and compared. Socioeconomic status was identified using average parent education as a proxy for the availability of economic resources. Socioeconomic status did not differ significantly between groups ($t[82] = 1.02$, $p = 0.311$), and averaged 16.23 ($SD = 2.17$) years of education in the HFASD-siblings group and 15.82 ($SD = 1.46$) years in the TD-siblings group. Total number of children in the family also did not differ between groups ($t[82] = 0.29$, $p = 0.774$), and averaged 2.50 ($SD = 0.77$) children in the HFASD-siblings group and 2.55 ($SD = 0.74$) children in the TD-siblings group.

Procedures

The current study was approved by a university institutional review board and conducted in accordance with the approved protocol. Target siblings for the HFASD-siblings group were recruited from a pool of families participating in a psychosocial therapeutic intervention with children with HFASD. Behavior Assessment System for Children, Second Edition-Parent Rating Scales (BASC-2 PRS) and Self-Report of Personality (BASC-2 SRP) measures were collected as close to initiation of treatment as feasible for the family. For those cases where the reference child with HFASD had an ADI-R ($n = 33$, 78.6 % of sample), the same parent completed the BASC-2 PRS as completed the ADI-R. Target siblings and their parents completed the rating forms on the same day. All assessments were administered by doctoral-level psychologists or advanced graduate students trained in their administration. Rating forms were reviewed for missing or multiply marked items, and corrections were completed as soon as possible after the assessment. BASC-2 ASSIST was used for all scoring and all measures were double-entered and verified by a senior researcher.

Measures

Wechsler Intelligence Scales for Children-Fourth Edition (WISC-IV)

Cognitive functioning level of the qualifying HFASD child was assessed using a 4-subtest short form of the WISC-IV (Wechsler 2003) consisting of Block Design, Similarities, Vocabulary, and Matrix Reasoning subtests. Methods described by Tellegen and Briggs (1967) were used to calculate short-form reliability and validity coefficients from information supplied in the technical manual. The short-form IQ correlated 0.92 with the full-scale IQ and showed a calculated internal consistency estimate of 0.95. The short-form VCI (Similarities and Vocabulary) correlated 0.82 with the original 3-subtest VCI and showed a calculated internal consistency estimate of 0.93. The short-form PRI (Block Design and Matrix Reasoning) correlated 0.66 with the original 3-subtest PRI and showed a calculated internal consistency of 0.92.

Autism Diagnostic Interview-Revised (ADI-R)

The ADI-R (Rutter et al. 2003) is the gold-standard, semi-structured diagnostic interview consisting of 93-items administered to a parent or caregiver of an individual being assessed for the presence of an autism spectrum disorder. The three domains of the ADI-R match those defined by the DSM-IV-TR (APA 2000), i.e., Reciprocal Social Interaction, Communication, and Restricted, Repetitive, and Stereotyped Patterns of Behavior. Item level inter-rater reliability has been demonstrated to be between 0.63 and 0.89 (Lord et al. 1994).

Behavior Assessment System for Children, Second Edition-Parent Rating Scales (BASC-2 PRS) and Self-Report of Personality (BASC-2 SRP)

The BASC-2 (Reynolds and Kamphaus 2004) is a well-supported survey/questionnaire with multiple forms (depending on age). It assesses broad dimensions of clinical and adaptive characteristics of children, allowing multiple informants to provide perspectives using a consistent format and standardization sample. Used in conjunction with additional clinical instruments the BASC-2 can assist in the identification of developmental, behavioral, and psychiatric conditions affecting children (Reynolds and Kamphaus 2004).

The current study used the BASC-2 PRS and BASC-2 SRP forms appropriate to the age of the participating child. For the BASC-2 PRS the PRS-Child (ages 6–11; 160 items) or the PRS-Adolescent (ages 12–21; 150 items) were administered by giving the form to the parent to complete independently. The PRS-Child and PRS-Adolescent are consistent in the format of responses, with parents rating a variety of items on a four point scale (Never, Sometimes, Often, and Almost Always). For the BASC-2 SRP the SRP-Interview form (SRP-I; ages 6–7; 65 items), SRP-Child (ages 8–11; 139 items), and the SRP-Adolescent (ages 12–21; 176 items) all were administered by reading the items to the participant and having them state their responses aloud. This was done to correct for any unidentified reading difficulties or comprehension problems. The PRS-Interview form consists of all yes or no questions. The SRP-Child and SRP-Adolescent are consistent in the format of responses, beginning with true or false questions and then proceeding to items rated on a four point scale (Never, Sometimes, Often, and Almost Always). All forms of the BASC-2 were entered into the BASC-2 Scoring Assistant program and normalized T-scores were derived. T-scores have a mean of 50 and a standard deviation of 10. Clinical ranges for the BASC-2 are normal (T-score < 60), at-risk (T-score between 60 and 69), and clinically significant (T-score > 69).

The current study focused on the anxiety and depression clinical scales of the BASC-2 only. The BASC-2 operationalizes anxiety and depression consistently on all response forms. The depression scale assesses symptoms such as withdrawal, feelings of loneliness, hopelessness, anhedonia, sadness, self-reproach, and general dysphoric mood. The anxiety scale assesses symptoms such as consistent and excessive worry, nervousness, negative rumination, and fearfulness (Reynolds and Kamphaus 2004). The clinical scales were developed to represent diagnostic symptoms for anxiety and mood (depression) disorders from the DSM-IV-TR (APA 2000).

The BASC-2 is well-supported with strong psychometric characteristics. The test manual reports median (across the age-determined forms used) coefficient alpha reliabilities of PRS-anxiety scale (0.84), PRS-depression (0.87), SRP-anxiety (0.86) and SRP-depression (0.84). Median test–retest reliabilities are reported as PRS-anxiety scale (0.79), PRS-depression (0.83), SRP-anxiety (0.72) and SRP-depression (0.71). Evidence of concurrent validity is well established with measures such as the CBCL (Achenbach and Rescorla 2001; see Reynolds and Kamphaus 2004).

Data Analyses

Multivariate procedures were used to test hypotheses because of the two factor composition of the dependent variable and to maximize statistical power. Multivariate normality for the dependent variables was assessed using Mahalanobis distance estimates. No violations of multivariate normality were identified. Bivariate correlations between dependent variables indicated acceptable collinearity for MANOVA: BASC-2 PRS, $r = 0.41$ and BASC-2 SRP, $r = 0.45$. Differences between groups in internalizing symptoms (BASC-2 anxiety and BASC-2 depression clinical scales) were assessed using MANOVAs. Source differences within each group were also assessed using MANOVAs (BASC-2 PRS vs. BASC-2 SRP).

Results

Mean anxiety and depression scores are summarized in Table 2. Separate between group MANOVAs were conducted for parent report (BASC-2 PRS) and child self-report (BASC-2 SRP). These are summarized in Table 3. Results of the parent-report analyses indicated no significant multivariate effect between groups (Wilk's $\lambda = 0.997$, $F[1,82] = 0.119$, $p = 0.888$). Results of the child self-report analyses indicated no significant multivariate effect between groups (Wilk's $\lambda = 0.982$, $F[1,82] = 0.724$, $p = 0.488$). Because of the lack of multivariate effects, no univariate follow-up analyses were conducted.

Table 2 Mean BASC-2 anxiety and depression clinical scales by group

Composite	HFASD-siblings Mean (SD)	TD-siblings Mean (SD)	Total Mean (SD)
BASC-2-PRS			
Anxiety	51.43 (9.67)	50.57 (8.83)	51.00 (9.21)
Depression	47.67 (7.27)	47.05 (6.95)	47.36 (7.07)
BASC-2-SRP			
Anxiety	48.93 (8.43)	50.36 (8.93)	49.64 (8.66)
Depression	46.55 (5.32)	45.95 (5.77)	46.25 (5.52)

Total $N = 84$, with $n = 42$ in each group

BASC-2 PRS Behavior Assessment System for Children, 2nd Edition, Parent Rating Scale; *BASC-2 SRP* Behavior Assessment System for Children, 2nd Edition, Self-Report of Personality

Table 3 MANOVA results for between-groups (HFASD-siblings group versus TD-siblings group) comparison for internalizing symptoms

Effect	Wilks' λ	*F* value (*df*)	*p* value	Partial η^2
BASC-2 PRS				
Multivariate test	0.997	0.119 (1, 82)	0.888	0.003
BASC-2 SRP				
Multivariate test	0.982	0.724 (1, 82)	0.488	0.018

Total $N = 84$, with $n = 42$ in each group. Partial η^2 presented as a measure of effect size

BASC-2 PRS Behavior Assessment System for Children, 2nd Edition, Parent Rating Scale; *BASC-2 SRP* Behavior Assessment System for Children, 2nd Edition, Self-Report of Personality

To examine source differences in internalizing problems, MANOVAs were used comparing parent report (BASC-2 PRS) to child self-report (BASC-2 SRP) for each group. These are summarized in Table 4. Results of the within-group analyses indicated no significant multivariate effect for the HFASD-siblings group (Wilk's $\lambda = 0.980, F[1,82] = 0.829, p = 0.440$) or for the TD-siblings group (Wilk's $\lambda = 0.992, F[1,82] = 0.336, p = 0.715$). Because of the lack of multivariate effects, no univariate follow-up analyses were conducted.

Discussion

The current study did not support an increased risk of internalizing problems (anxiety and depression) in the TD siblings of children with HFASD. Specifically, while we hypothesized that there would be an elevated level of internalizing problems, we found no higher mean difference relative to a matched comparison group. This finding was supported by consistent levels of internalizing problems reported across informants. While past results have varied, the results of the current study are consistent with recent work in siblings of children with ASD and ID or mixed functional levels (e.g., Hastings and Petalas 2013; Pollard et al. 2013; Tomeny et al. 2012). However, the available research specific to siblings of children with HFASD has been minimal. While improving upon the limitations of the existing research, the current study also supports a minimal risk finding for TD siblings of children with HFASD.

Table 4 MANOVA results for within-groups (parent vs. child report) source comparisons for internalizing symptoms

Effect	Wilks' λ	*F* value (*df*)	*p* value	Partial η^2
HFASD-siblings				
Multivariate test	0.980	0.829 (1, 82)	0.440	0.020
TD-siblings				
Multivariate test	0.992	0.336 (1, 82)	0.715	0.008

Total $N = 84$, with $n = 42$ in each group. Partial η^2 presented as a measure of effect size

There are clinical implications to the current results. A generally increased risk of internalizing problems would have indicated a need for targeted intervention and therapeutic support in the community and school environments (Tudor and Lerner 2015). Though the current result does not support targeting resources to this problem in siblings of children with HFASD, mental health practitioners should maintain awareness of these families and attend to other potential clinical concerns if presented. Other concerns such as academic or behavioral problems may be present in siblings that were not evaluated in the current study.

There are three factors likely contributing to the current results. First is the functional level of the reference children with HFASD. Strong cognitive ability in their siblings with HFASD may allow shared communication and interests, promoting a positive sibling relationship and minimizing the impact of the behavioral and social deficits of the child with HFASD. A second explanatory factor involves the sample characteristics in this study. The average parent education in both groups was close to 16 years, meaning that parents in both groups had approximately a college education. This level of resources may serve as a protective factor from life stressors unrelated to the presence of a child with HFASD, attenuating the potential for internalizing problems (Lee et al. 2009). Third, the children with HFASD were participating in various related intervention trials, and though the measures for the current study were conducted very early in the treatment programs, some early intervention effects may have had an ancillary influence on the internalizing problems of the TD siblings. Likewise, by enrolling their children with HFASD in intervention, the families demonstrated a willingness to seek and participate in psychotherapeutic support, a positive indicator for

family function and outcomes. This factor is also a limitation of the current study.

The current study benefited from a variety of strengths. It was the first to explore levels of internalizing problems using both anxiety and depression in TD siblings of children with HFASD. It was also the first to provide specific documentation of cognitive ability for the reference sibling and use both parent-report and child self-report measures from a single well-validated instrument (BASC-2). In addition, the diagnoses of reference siblings with HFASD were established prior to enrollment in the study, and a majority of these diagnoses (78.6 %) were confirmed with the ADI-R. Finally, the TD comparison group was strongly analogous to the HFASD-siblings group in that it was screened using the same procedures; matched on age, gender, and ethnicity; demonstrated equivalence in reference child age, age-difference between target sibling and reference child, family socio-economic status, and total family size; and composed of children with TD siblings.

The current study has some limitations. First, while the target participants were screened for diagnosed developmental, psychiatric, and educational concerns; the current study did not specifically quantify possible sub-threshold characteristics of ASD in TD participants. Some past researchers (e.g., Orsmond and Seltzer 2009) have proposed a diathesis-stress model to sibling internalizing symptoms wherein sub-threshold ASD symptoms of the TD child themselves, interacting with family stressors, accounts for increased risk of anxiety or depression. However, the current study was intended to demonstrate a link between the presence of a sibling with diagnosed HFASD and internalizing problems of their TD sibling as a clinical consideration. Thus, the unidentified characteristics of TD siblings were not germane to the primary question. Future studies should consider including a measure of ASD symptoms for both the siblings of children with HFASD and TD comparison children to account for this proposal. Another limitation is the lack of matching between groups on the birth order of participants relative to reference siblings and the gender of the reference children. As discussed in the previous limitation, because the primary consideration was identifying an elevated clinical risk for the target participant, these characteristics were not directly relevant to the hypothesis. In addition, the research exploring these factors has not identified a consistent configuration of family or reference sibling demographic characteristics to be uniquely important (Maeden et al. 2010). Future research could explore the unique contribution of these factors in matched samples. In addition, information on the prior utilization of psychological or social services for members of the participating families were not documented. Future research should consider this as an additional factor. Finally, the overall sample is limited in that it was composed of primarily Caucasian (97.6 %) and highly educated families. This limits the generalizability of the results. Future studies should include larger and more diverse samples.

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Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in this study.

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