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Impairments in phonological processing and nonverbal intellectual function in parents of children with autism

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Abstract

Language difficulties have been implicated to be a part of the broad autism phenotype in first-degree relatives of individuals with autism. Phonological processing difficulties in particular have been reported by some, but not all groups studying parents or siblings of probands with autism. In the present study, we examined a broad battery of language tasks and general cognitive abilities in parents of children with autistic disorder. Parents of individuals with autism ($n = 22$) were compared to matched adult controls on a series of cognitive and language measures. Parents of children with autism exhibited lower performance on the matrix reasoning subtest and total performance IQ than did controls, but did not show differences in verbal IQ measures, when tested with the Wechsler Abbreviated Scale of Intelligence (WASI). In addition, parents of children with autism had lower performance on a nonword repetition task, but did not show differences on tests of figurative language, receptive language, expressive language, and verbal fluency and on a questionnaire assessing history of reading difficulties. Results from this study are generally consistent with the cognitive profiles reported for parents of children with autism. Our finding of nonword repetition difficulties, along with others' previous findings for nonword reading in autism families, suggests that problems in phonological processing might be characteristic of the broad autism phenotype.

Keywords

Autism parents; Phonological processing; Nonverbal IQ; Broad autism phenotype; Language

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Autism is a developmental disorder characterized by impairments in social interaction and communication in conjunction with a restricted range of behaviors and interests (Rutter, 2000). Although the etiology of autism is not well understood, twin studies have shown that idiopathic autism is highly heritable. Monozygotic concordance rates range from 36% to 91% (versus dizygotic concordances of 0–23%) and overall autism heritability rates are

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estimated to be as high as 90% depending on the strictness of the definition of autism used to estimate heritability (Bailey et al., 1995; Folstein & Rutter, 1977; Ritvo, Freeman, Mason-Brothers, Mo, & Ritvo, 1985; Steffenburg et al., 1989).

In order to investigate the genetic basis of autism with molecular genetic studies, it is crucial to determine the behavioral characteristics associated with the disorder (Bishop et al, 2004a; Rutter, 2000). One approach to this issue has been to study the nonaffected parents and siblings of children with autism. The genetic liability for autism may be expressed in these individuals with behavioral and cognitive characteristics that are milder than, but qualitatively similar to, the defining features of autism. Studies have established differences between nonautistic relatives of autistic probands and the general population in personality traits (Murphy et al., 2000; Piven et al., 1994; Wolff, Sukhdev, & Moyes, 1988), rates of psychiatric illness (e.g., Bolton, Pickles, Murphy, & Rutter, 1998; Piven et al., 1991), cognitive style (Happé, Briskman, & Frith, 2001), brain structure (Peterson et al., 2006; Rojas et al., 2004), and communication and social deficits and stereotyped behaviors (Bolton et al., 1994; Piven, Palmer, Jacobi, Childress, & Arndt, 1997). These differences suggest the existence of what has come to be known as the broad autism phenotype (BAP). Although studies of nonaffected relatives mainly assess familiality, rather than genetic heritability, the presence of these systematic differences in family members of autistic probands is consistent with the idea that autism spectrum disorders (ASDs) are highly heritable.

As part of the investigation of the BAP, attempts have also been made to determine the neuropsychological profile of nonaffected relatives of people with autism. Language and intelligence testing has been carried out on these populations. In terms of intelligence, this work has suggested that parents of individuals with ASD may show patterns of intelligence scores similar to that of individuals at the high-functioning end of the autism spectrum. Several studies have found lower performance IQ scores than verbal IQ scores in these parents. For example, Folstein et al. (1999) reported that in a large sample of 90 families of children with autism, parents of the probands exhibited lower performance IQ scores than verbal IQ scores. A multiplex family study found that performance IQ and reading measures were lower in parents of probands with autism than in parents of probands with Down syndrome (Piven & Palmer, 1997). In individuals with ASD, patterns of IQ scores are also uneven, but this varies across the spectrum of IQ scores. In those who have lower full scale IQ scores, performance IQ usually exceeds verbal IQ. On the other hand, individuals with ASD who have higher full scale IQ scores (Ruhl, Werner, & Poustka, 1995) or individuals with Asperger syndrome (Gilchrist et al., 2001) have higher verbal IQ scores than performance IQ scores. It is possible that high-functioning individuals with ASD are more representative of the BAP than are low-functioning individuals with ASD, who may have a more complex etiology. Other studies fail to find differences in the intelligence scores of parents of children with autism and control samples (Bishop et al., 2004a; Freeman et al., 1989; Szatmari et al., 1993; for a review, see Bailey, Palferman, Heavey, & Le Couteur, 1998).

Studies of language function in the BAP have shown a wide range of language difficulties in parents of children with ASD, including both core language skills and more pragmatic language abilities. In particular, a number of studies have shown problems with pragmatic language skills in parents of individuals with ASD. Landa et al. (1992) reported that pragmatic language was impaired in parents of children with autism, albeit in milder form than in the probands themselves. Piven et al. (1994) found that parents of autism probands were more aloof and tactless on a personality interview than were the parents of Down syndrome probands, which they link to weak pragmatic language skills. Piven and Palmer (1997) have also reported higher rates of these personality characteristics, as well as pragmatic language problems, in a sample of 25 multiplex autism families.

Core language difficulties have also been implicated as a common factor among families of children with autism. Piven and Palmer (1997) reported that parents of children with autism were slower on the rapid automatized naming task for colors and objects but not for numbers and letters than the parents of Down syndrome probands. Folstein et al. (1999) found weaker performance on the Word Attack subtest (a nonword reading task) of the Woodcock–Johnson battery (Woodcock & Johnson, 1991) in ASD parents than in Down syndrome parents. Parents of individuals with ASD were three times more likely to report a history of language-related difficulties than parents of individuals with Down syndrome. Positive early language-related difficulties were correlated with lower verbal IQ and nonword reading scores.

There has been some speculation about a possible etiological overlap between specific language impairment (SLI) and autism, both of which demonstrate structural language difficulties, including phonological processing deficits (Tager-Flusberg & Joseph, 2003). However, Bishop et al. (2004b) report no deficits in parents and siblings of children with autism on a nonword repetition task that is considered to be an index of phonological processing. They suggest that phonological processing deficits are not a part of the BAP. Table 1 summarizes findings in the literature regarding core language and intelligence measures in autism.

These patterns of pragmatic and core language deficiencies reported in the literature lead us to hypothesize that the BAP language profile may include a specific weakness in phonological processing skills. The reports of poorer rapid automatized naming, a task that has a phonological component (Piven & Palmer, 1997), and phono-logical processing deficits (Tager-Flusberg & Joseph, 2003) in the BAP suggest this possibility. It may also be that phonological processing difficulties underlie the early history of language-related difficulties reported in parents of children with autism compared to controls (Folstein et al., 1999; Fombonne, Bolton, Prior, Jordan, & Rutter, 1997), as is found in other language disorders such as dyslexia (Temple et al., 2001) and specific language impairment (Gray, 2006). Although not all studies of nonword tasks in the BAP report a deficit (e.g., Bishop et al, 2004b), some of these negative reports include probands that were on the higher functioning end of the autism spectrum. We hypothesize that parents whose children are more clearly defined as autistic and who are lower functioning may show a phonological processing deficit.

The purpose of the present study was to test this hypothesis. In addition, we wished to use a larger battery of language measures in order to more widely survey the language abilities of parents of children with autism. This allowed for the comparison of phonological processing directly to a wider variety of language skills and intelligence measures. We included tests of phonological functioning and fluency as well as expressive and receptive language, which were normed for adults. We also included a test of figurative language processing since this is known to be deficient in individuals on the autism spectrum (MacKay & Shaw, 2004). These tests have not been previously used with parents of autistic probands.

METHOD

Participants

Neuropsychological data were collected on 22 biological parents of children with autism from 17 families: 14 mothers and 8 fathers. Due to the small number of fathers who participated in the study, gender differences were not investigated. A total of 2 parents from two families (9%) were non-Hispanic African Americans. The remaining 20 parents (91%) were non-Hispanic European Americans. With the exception of one family with two children who met criteria for autism, each parent had just one child who met DSM-IV (*Diagnostic and Statistical Manual of Mental Disorders—Fourth Edition*; American Psychiatric Association, 1994) criteria for autism, as determined by the Autism Diagnostic Observation Schedule (ADOS; Lord et al, 1989) and the Autism Diagnostic Interview, Revised (ADI-R; Lord, Rutter, & LeCouteur, 1994) or the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003) and by current clinical diagnosis. Despite the fact that ADOS scores were available for all of the children with autism, different modules had been used depending on the age and verbal ability of the child. In order to compare ADOS scores across modules, a severity score for each child was calculated by summing the number of symptoms endorsed as clinically significant and dividing by the total symptom score possible, consistent with methods utilized by Bailey et al. (2005). In this paper, ADOS scores transformed in this way are referred to as ADOS severity.

A total of 22 age- and gender-matched comparison participants were recruited from the Denver area. The two groups were matched based on age, handedness, IQ, and socioeconomic status (see Table 2), but not parenting status (i.e., some of the control participants were not parents). Handedness was assessed using the Annett handedness questionnaire (Annett, 1985); socioeconomic status was assessed using the Hollingshead Four-Factor Index of Social Status (SES; Hollingshead, 1975). IQ was assessed using the Wechsler Abbreviated Scale of Intelligence (WASI; Psychological Corporation, 1999). The IQ level of both groups was higher than the national average, consistent with a community-based study from the Denver metropolitan area (Willcutt et al., 2001). All participants demonstrated normal hearing (≤20 dB HL between 0.25 and 4 kHz) on a method of constant stimuli screening procedure performed in an acoustically shielded environment to rule out hearing deficiencies.

All participants were screened for the existence of psychiatric disorders with the Structured Clinical Interview for DSM-IV Axis I Disorders, Research Version (SCID; First, Spitzer, Gibbon, & Williams, 2002) and/or the SCID Screen Patient Questionnaire Extended Computer Program (SSPQ-X). Participants who reported during screening that they had never seen anyone professionally for psychiatric symptoms (e.g., depression or anxiety) were first administered the SSPQ-X. If a participant's answers on the SSPQ-X indicated the possibility of a psychiatric disorder, all of the questions in the appropriate SCID section (e.g., mood disorders) were administered. Those participants who reported psychiatric histories at screening were administered the full SCID without the SSPQ-X. All participants in the comparison group reported no personal or family history of neurological or Axis I psychiatric illness and met the Research Diagnostic Criteria (Spitzer, Endicott, & Robins, 1978) for "never mentally ill." Of the 22 parents of children with autism who participated, 14 (64%) had at least one Axis I psychiatric disorder based on DSM-IV criteria. These included mood disorders (12 parents), anxiety disorders (8 parents), substance use disorders (4 parents), and eating disorders (1 parent). This pattern is consistent with reports in the literature of significantly higher rates of psychiatric disorders in parents of children with autism than in parents of children with other developmental problems (Micali, Chakrabarti, & Fombonne, 2004; Piven et al., 1991; Piven & Palmer, 1999). The presence of a psychiatric disorder did not correlate with any of the cognitive profile measures used in the study, suggesting that it did not influence our findings. Table 2 summarizes the demographic data for each group.

Testing materials and procedure

Prior to participation in the study, all participants provided written informed consent. Participants then completed a battery of language and intelligence measures. Instruments were selected for available normative data across the age ranges employed in the study. Table 3 summarizes the measures used and the domains they tested, which are more fully described below. Participants completed all testing in a single session lasting approximately two hours, with breaks given as needed.

For each trial of the Peabody Picture Vocabulary Test (PPVT-III; Dunn & Dunn, 1997) four black-and-white illustrations were presented in a multiple-choice format. The participant was required to select the picture best illustrating the meaning of a stimulus word presented orally by the examiner. The Expressive Vocabulary Test (EVT; Williams, 1997) is conormed with the PPVT-III. For each trial of this test, an orally presented word and a simple line drawing were given. Participants were required to present a synonym to the presented word that also matched the illustration. The Verbal Fluency subtest from the Delis Kaplan Executive Function System (DK-EFS; Delis, Kaplan, & Kramer, 2001) is a test of verbal fluency based on initial letters (words beginning with F, then A, then S), categories (animals, boys' names), and then a switching test where participants produced words alternatively from the categories fruit and furniture. The score for this test reflects degree of fluency as well as switching accuracy. For the Figurative Language subtest from the Test of Language Competence–Expanded Edition (TOLC-E; Wiig & Secord, 1989), participants were presented with a context (*Two students moving to a new town*) and then a metaphorical expression (*There is rough sailing ahead for us*). They were first required to give an oral interpretation of the expression and then to select the best sense of the expression in a multiple-choice format with the metaphorical meaning (*We will be facing a hard road*) presented in random order with nonrelated (*It took the wind out of our sails*), literal (*The waves are going to make it hard to sail*), and opposite meaning (*The rough times are behind us now*) foils. The Nonword Repetition subtest of the Comprehensive Test of Phonological

Processing (CTOPP; Wagner, Torgesen, & Rashotte, 1999) presented participants with 18 auditory nonwords via an audiocassette recorder. Immediately after each nonword, they were required to repeat it. They received a positive score only if the entire nonword was correctly repeated. The nonwords ranged in length from one to seven syllables and progressed with the easiest and shortest nonwords to the longest and most difficult. After three incorrect trials, the test was discontinued. The Reading History Questionnaire (RHQ; Lefly & Pennington, 2000) is a written survey consisting of 26 questions probing the parent's possible difficulties with reading from childhood (How much extra help did you need when learning to read in elementary school? Did you ever reverse the order of letter s or numbers when you were a child?) through adulthood (How much reading do you do for pleasure? How would you compare your current reading speed to that of others of the same age and education?). Responses were made on a 0–4 Likert-type scale individually designated for each question, with a higher number indicative of more reading difficulty in each case. The sum of responses to all questions was used as the RHQ score.

Analysis

A multivariate analysis of variance (MANOVA) profile analysis was performed on 13 language and intelligence scores from the measures in Table 3: WASI Vocabulary subtest, WASI Similarities subtest, WASI Block Design subtest, WASI Matrix Reasoning subtest, CTOPP nonword repetition, TOLC figurative language, PPVT receptive language, EVT expressive language, DK letter fluency, DK category fluency, DK category switching, DK switching accuracy, and reading history. Standard scores from each subtest were converted to z-scores using the means and standard deviations of the entire sample of 44 participants before completing the analysis to facilitate statistical comparison via profile analysis. Only 1 participant, a parent, had scores beyond ±3 standard deviations; these two scores were not deleted for the analysis. Assumptions regarding normality of sampling distributions, homogeneity of variance–covariance matrices, linearity, and multicollinearity were met. SPSS MANOVA was used to conduct the profile analysis.

RESULTS

Profile analysis

Using Wilks's criterion, the profiles exhibited a trend toward deviation from parallelism, $F(12, 31) = 2.0$, $p = .06$, partial $\eta^2 = .44$, indicating a possible difference in the pattern of test performance between the two groups. For the levels test, no significant differences were found among groups when scores were averaged over all subtests, $F(l, 42) = 0.35$, $p = .56$. When averaged over groups, individual measures were not found by Hotelling's criterion to deviate from flatness, $F\{12, 31) = 0.05$, $p = 1$, partial $\eta^2 = .02$.

To further explore the results, a simple effects analysis of group was conducted holding test type constant. In addition, univariate statistics from a MANOVA were calculated on three additional measures: verbal IQ, performance IQ, and full scale IQ (see Table 4). The key finding was that ASD parents had lower performance on the non-word repetition subtest of the comprehensive test of phonological processing (CTOPP), but did not show differences on tests of figurative language, expressive language, verbal fluency, and history of reading difficulties. Additionally, parents of individuals with ASD exhibited lower performance IQ scores than controls, but did not show differences from controls in verbal IQ measures. The former is primarily due to their lower performance on the matrix reasoning subtest.

An exploratory analysis was conducted on the phonological processing (CTOPP) test results with syllabic length of the nonwords as the key variable. We followed the usual clinical practice of using a discontinue rule with our testing, such that after three incorrect trials in the nonword repetition test, it was discontinued. The nonwords varied in length from one to seven syllables, but due to the use of the discontinue rule, words longer than three syllables were completed by only a subset of participants. All but one parent completed all the three-syllable words. One control participant was randomly deleted to provide equal numbers in each of the groups, and the analysis was performed on the remaining 42 participants. A mixed 2×2 MANOVA resulted in significant effects of number of syllables, $F(2,39) = 20.6$, $p < .001$, and group, $F(1, 40) = 16.4$, $p < .001$, and an interaction, $F(2, 39) = 7.6$, $p = .002$, as seen in Figure 1.

Correlation analyses

Exploratory analyses were conducted to test relationships between the parental neuropsychological profile and the functioning of their child with autism. Pearson correlations were calculated between the IQ subtests, language measures, and the child SCQ and ADOS severity scores. The analysis revealed a significant positive correlation between the parent WASI Similarities subtest and the child SCQ score ($r = .62$, $p = .002$), a result that survived Bonferroni correction for multiple comparisons. In other words, parents whose children had a more severe SCQ rating scored higher on the Similarities subtest.

Our data generally did not show significant relationships between those participants with a history of early reading difficulties and those without such difficulties (Folstein et al., 1999; Fombonne et al., 1997). However, higher verbal fluency scores for the switching task were observed in individuals who had lower RHQ scores (i.e., had less trouble learning to read; $r = -.34$, $p = .02$).

DISCUSSION

The parents of children with autism exhibited lower performance IQ scores and exhibited more difficulty with phonological working memory from a nonword repetition task. These characteristics of the BAP are indicative of which aspects of the autism phenotype in general (i.e., not the BAP) may be familial and which may be just secondary consequences of having full autism. For example, mental retardation is not familial in relatives of a proband with autism that has mental retardation (Bailey et al., 1995, 1998), suggesting that mental retardation in autism may be secondary to autism. Theoretically, then, our findings suggest aspects of the autism phenotype that may be primary and familial rather than secondary.

The lower performance IQ scores in the parents of children with autism are comparable to other findings in the literature (Folstein et al., 1999, Piven & Palmer, 1997). The lack of verbal IQ score differences between the parents of children with autism and the control group is also consistent with many previous findings (Bishop et al., 2004a; Folstein et al., 1999; Piven & Palmer, 1997), although higher verbal IQ scores in parents of children with autism have been reported (Fombonne et al., 1997). Such a pattern has also been found in higher functioning children on the autism spectrum (Gilchrist et al., 2001). Thus this pattern of higher verbal than performance IQ scores has been reported in both high-functioning autism and first-degree relatives of children with autism. It is possible, albeit speculative, that this pattern may be more representative of phenotype heritability in autism in general, in the absence of general cognitive impairment (i.e., mental retardation).

The lack of difference in verbal IQ scores between the parents of children with autism and the comparison participants is also consistent with their performance on all but one of the language measures we administered. These tests have not been previously administered to a sample of parents of children with autism. While it is necessary to be cautious in interpreting null results, our data suggest that higher language functions such as fluency, general expressive and receptive language skills, and understanding of figurative language are not impaired in parents of children with autism. This is consistent with previous reports that family members of children with autism do not show deficient performance on tests of reading or spelling (Folstein et al., 1999), since reading and spelling are related to these language functions. It also suggests that these types of language deficiencies are not a part of the BAP or primary to autism, although perhaps a pattern of higher verbal IQs may be.

The current findings add to the debate about phonological processing in parents of children with autism. Although the parents of children with autism did not show poor performance on most of the core language measures in our battery, they did exhibit significantly poorer performance on the nonword repetition task. This suggests that problems in phonological processing might be characteristic of the extended ASD phenotype. This is consistent with previous findings of poor performance for nonword reading in ASD families (Folstein et al., 1999). Both these nonword tasks are phonetic rather than orthographic in nature and are therefore measures of phonological processing ability. Since phonological processing is likely deficient in autism itself (Kjelgaard & Tager-Flusberg, 2001; Oram Cardy et al., 2005) it is plausible that phonological problems could be a part of the BAP, and therefore a primary deficit in ASD, rather than a secondary consequence of having an ASD.

Nonword reading and repetition problems are not uniformly found in samples of first-degree relatives of autism probands, however, as reported in two additional studies (Bishop et al., 2004b; Piven & Palmer, 1997). Bishop and her colleagues reported no differences in nonword repetition or nonsense passage reading in parents of children with autism compared to control parents.

A conceivable reason for this discrepancy is that lower scores on phonological tasks in the parents are associated with a proband sample with lower IQ scores and greater severity of the disorder. It is possible that lower functioning in the proband is related to a larger genetic liability in the parents, driving this pattern. The proband sample used by Bishop et al. (2004b) included a total of 59 children with a full-blown ASD, but also 23 children with pervasive developmental disorder not otherwise specified (PDD-NOS) who only met criteria for one or two of the three main criteria for an ASD diagnosis, with an overall average verbal IQ of 76 and performance IQ of 86. Our sample had a relatively low average IQ score in the ASD probands (approximately 60) consistent with the idea that the existence of lower IQ scores in the probands is related to a phonological processing difficulty in the parents. Additionally, the proband sample in Piven and Palmer (1997), who reported no family nonword reading difficulties, appears to have a higher mean IQ score than the sample reported by Folstein et al. (1999), who included probands with extremely low IQ scores and family members with poor nonword reading. Arguing against this inter pretation, however, is the lack of any significant relationship between proband symptom severity and any of the parental language measures in our study. Future work on the BAP could benefit from a systematic variation of level of functioning of the probands, as our sample size may have been too low to detect a small effect. Additionally, future work could examine language levels of the probands, which may be more directly related to parent language than IQ.

The current findings bear directly on the question regarding etiological overlap between autism and SLI (Bishop et al., 2004b; Tager-Flusberg & Joseph, 2003). If parents of lower functioning children with autism have phonological processing deficits, as we suggest here, this supports the idea that there could be a genetic link between autism and SLI and that phonological processing deficits are a part of the BAP, although further work is needed to clarify these claims.

Problems with nonword repetition tasks with nonwords of varying lengths can be interpreted as a phonological memory problem when there is a group by syllabic length interaction effect. For example, individuals with SLI demonstrate performance that is differentially worse on items with more syllables (Marton & Schwartz, 2003). On the other hand a main effect of group with no interaction can be interpreted as a phonological encoding or representation problem rather than a phonological memory problem. Since we obtained a strong group by syllabic length interaction, our results may suggest that the parents exhibit phonological memory difficulties rather than encoding deficits. This is consistent with other work showing a phenotypical overlap between SLI and autism (Bishop et al., 2004b). Hearing or articulatory problems could also lead to difficulties with a nonword repetition task. All of our participants in both groups had normal hearing, ruling out hearing problems as a potential explanation for our results. We did not explicitly test for articulation problems; this would be an appropriate direction for future work.

We speculate that these low-level phonological processing problems could be what are driving some of the higher level language deficits seen in ASD. The parents do not have the higher level deficits because the phonological problems they have are milder. Or, it could be that parents have devised atypical strategies for performing language tasks and are using different neural networks. This is plausible due to their high verbal intelligence and is supported by our finding of different profiles in the test of parallelism.

Piven and Palmer (1997) found one additional language related task that showed deficient performance in parents of children with autism, the rapid automatized naming task—parents were slower than controls on the color and object subtests, but not slower on number and letter subtests. This could be one potential area where problems with higher level tasks (i.e., naming) may be explained by problems with lower level functions such as phonological processing. (See Brizzolara et al., 2006, for data supporting the distinctiveness of naming and phonological processing.) It is possible that the generally more highly automated grapheme-to-phoneme conversion required for reading numbers and letters presents parents of children with autism with fewer problems relative to control participants than the generally less auto mated process of viewing a color or object and lexically accessing a word and then its phonological representation. Both our nonword repetition task and the nonword reading task reported by others (Folstein et al., 1999) involve processing unfamiliar phoneme strings. Performance on the color and object subtests also involve processing less familiar phoneme strings than do the letter and number sub tests. Thus it may be that the less automatic processing required for both these situations is what presents a challenge for parents of children with autism. All this points to the need for further investigation of phonological processing problems in autism and the BAP and their link to higher language deficits.

The profile analysis indicates that while parents of children with autism differ on only a few measures of intelligence and language functioning, these differences represent an overall different pattern of performance. It is possible to provisionally suggest that what is detectable with behavioral tests does not reveal the full differences in neurological functioning in the parents of children with autism. This is supported by recent work from our laboratory, which has uncovered substantial differences in the brain structures of a similar group of such parents (Peterson et al., 2006). It is thus also conceivable that there are substantial functional differences as well. These could be explored in situations where neural differences in language functioning are found even when there are no overt behavioral differences in children with autism and other develop mental disorders (Flagg, Oram Cardy, & Roberts, 2006; Roberts, Flagg, & Gage, 2004). Further neuroimaging work is needed to explore these differences in children with autism as well as the BAP as manifest in relatives of the probands.

One important feature of the current study is the participation of families with a single child with autism (only one multiplex family member participated). The majority of autism family studies reported to date have been conducted with multiplex families—that is, those with more than one proband with autism (e.g., Piven & Palmer, 1997). Multiplex families probably have higher genetic loading than nonmultiplex families, which could result in a higher likelihood of finding differences between the groups of parents of individuals with ASD and controls. These findings strengthen already-published findings in this area, such as the language and IQ findings of Folstein et al. (1999). It is possible, however, that the use of multiplex families may have yielded additional findings on other measures in the study. A strength of this study was the inclusion of a number of language measures that had not previously been used with parents of children with autism. Finding normal functioning on these tests in the parents of children with autism extends the findings of other investigators who report no core language difficulties in first-degree relatives (e.g., Folstein et al., 1999). However, we were not able to include some language testing that would have added valuable information, such as tests of pragmatic language abilities. We also did not include a broader assessment of other aspects of the BAP, such as measures of personality, social functioning, or narrow interests.

A number of implications emerge from the findings presented here. In both parents of children with autism and the probands themselves, low level phonological difficulties, as we report here, could lead to reading comprehension difficulties. There is evidence that both phonological decoding difficulties, as demonstrated by childhood reading and spelling problems (Folstein et al., 1999), and passage comprehension difficulties (Piven & Palmer, 1997; see Table 1) are evident in parents of children with autism. Thus it is plausible to suggest that language difficulties on the autism spectrum and in the BAP may be due to phonological processing difficulties. From a clinical standpoint, this leads to the suggestion that intervention at the phonological level may benefit children with autism who have language difficulties.

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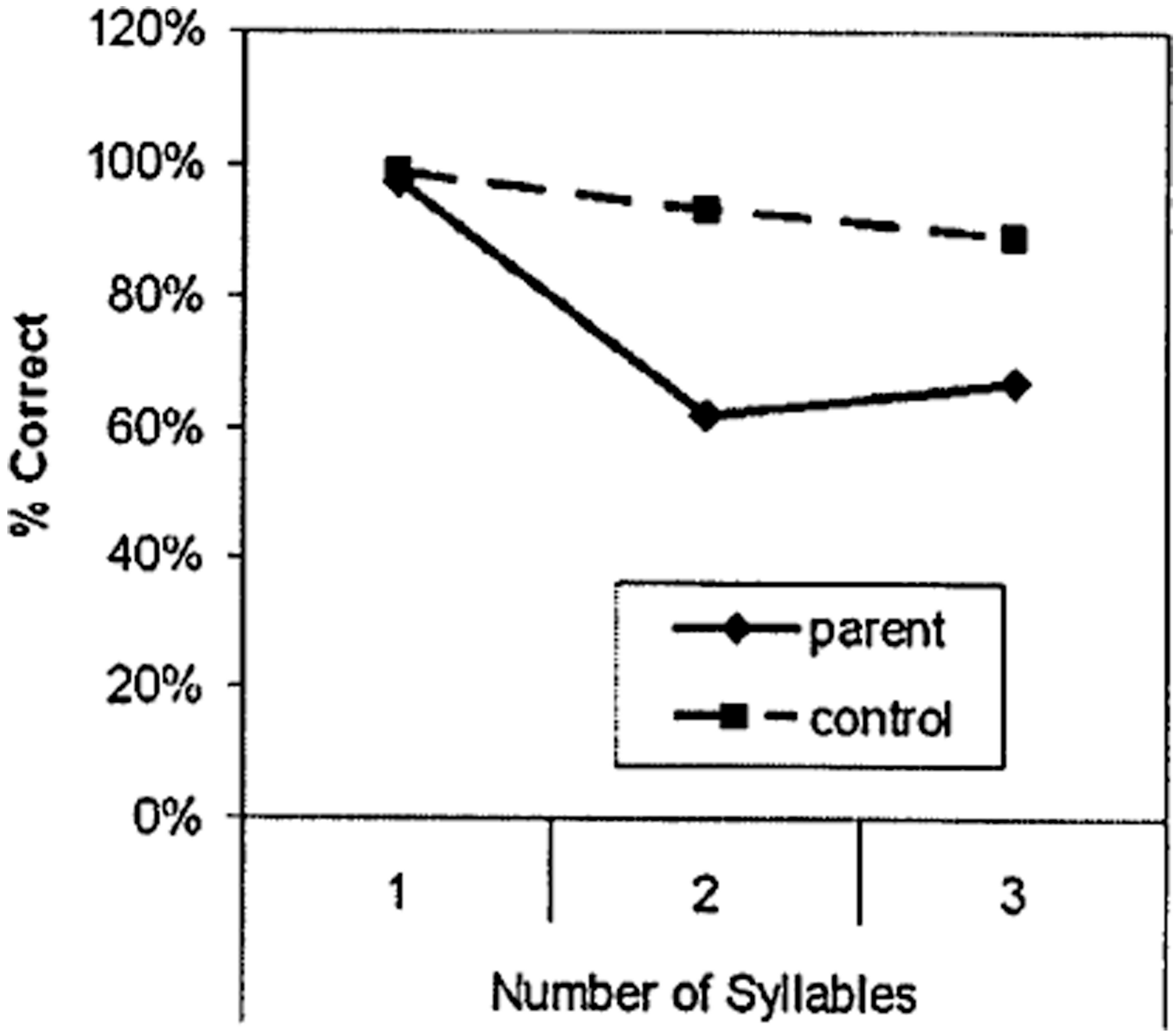

Figure 1.
Percentage of errors in the parent and control groups based on number of syllables for the Comprehensive Test of Phonological Processing (CTOPP) nonword repetition test.

TABLE 1

Intelligence and core language findings in the literature

Measure	Finding	References
Full Scale IQ	Autism parents lower than DS parents	Folstein et al., 1999
	No difference between autism & DS parents	Fombonne et al., 1997
Performance IQ	Autism parents lower than DS parents	Folstein et al., 1999; Piven & Palmer, 1997
	No difference between autism & DS parents	Fombonne et al., 1997
	No difference between autism & normal control parents	Bishop et al., 2004a
Verbal IQ	Autism parents higher than DS parents	Fombonne et al., 1997
	No difference between autism & DS parents	Folstein et al., 1999; Piven & Palmer, 1997
	No difference between autism & normal control parents	Fombonne et al., 1997
Picture Arrangement WAIS–R subtest	Autism parents lower than DS parents	Folstein et al., 1999
Picture Completion WAIS–R subtest	Autism parents lower than DS parents	Folstein et al., 1999; Piven & Palmer, 1997
Object Assembly WAIS–R subtest	Autism parents lower than DS parents	Piven & Palmer, 1997
Verbal subtests—Digit Span, Vocabulary, Comprehension, Similarities	Autism parents higher than DS parents	Fombonne et al., 1997
Verbal IQ—Performance IQ difference	VIQ > PIQ for autism parents, No difference for DS parents	Folstein et al., 1999
	VIQ > PIQ for autism parents, PIQ > VIQ for DS parents	Fombonne et al., 1997
Woodcock Johnson nonsense word reading	Autism parents lower than DS parents	Folstein et al., 1999
	No difference between autism & DS parents	Piven & Palmer, 1997
Nonword repetition & nonsense passage reading	No differences between autism parents & normal control parents	Bishop et al., 2004b
Early language related cognitive deficits—late onset of phrase speech, articulation, reading, or spelling problems	Autism parents more than DS parents	Folstein et al., 1999
Rapid automatized naming task—color & object	Autism parents slower than DS parents (but not slower on number & letter)	Piven & Palmer, 1997
Woodcock Johnson passage comprehension	Autism parents lower than DS parents	Piven & Palmer, 1997

Note. DS = Down syndrome. WAIS-R = Wechsler Adult Intelligence Scale–Revised. VIQ = Verbal IQ. PIQ = Performance IQ.

TABLE 2

Participant characteristics

Variable	Autism parent Mean	Autism parent SD	Control Mean	Control SD	p (2-tailed)
Age	38.6	6.2	37.6	5.8	.59
Handedness[a]	.81	.35	.77	.32	.68
SES (Hollingshead)[b]	49.6	8.0	50.4	6.0	.71
Full Scale IQ	116.1	7.7	119.7	9.4	.17
Presence of a mood disorder	N = 14 64%		0%		
Child SCQ scores	22.2	4.2	n/a		
Child ADOS severity score	58.5	21.7	n/a		

Note. SCQ = Social Communication Questionnaire. ADOS = Autism Diagnostic Observation Schedule. n/a = not applicable.

[a]Handedness scores are derived from Annett (1985) where +1 represents completely right handed, and −1 represents completely left handed.

[b]Hollingshead Four-Factor Index of Social Status (SES). This tool is commonly used in research to assess familial SES using both parents' education and occupation, if available.

TABLE 3

IQ and language measures

	Measure	Reference
General intellectual ability	Wechsler Abbreviated Scale of Intelligence (WASI); includes these subtests:	Psychological Corporation, 1999
	Vocabulary	
	Similarities	
	Block Design	
	Matrix Reasoning	
Receptive language	Peabody Picture Vocabulary Test (PPVT-III)	Dunn & Dunn, 1997
Expressive language	Expressive Vocabulary Test (EVT)	Williams, 1997
	Verbal Fluency subtest from the Delis Kaplan	Delis, Kaplan, & Kramer, 2001
	Executive Function System (DK-EFS)	
Figurative language	Figurative Language subtest from the Test of Language	Wiig & Secord, 1989
	Competence-Expanded Edition (TOLC-E)	
Phonology	Nonword Repetition subtest of the Comprehensive Test of Phonological Processing (CTOPP)	Wagner, Torgesen, & Rashotte, 1999
Difficulty learning to read	Reading History Questionnaire (RHQ)	Lefly & Pennington, 2000

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TABLE 4

Results of univariate statistics from MANOVA and simple effects analysis

	Original scores				Z-scores					
	Autism parent		Control		Autism parent		Control			
Measure	Mean	SD	Mean	SD	Mean	SD	Mean	SD	F	p (2-tailed)
Verbal IQ	115.1	8.7	114.4	9.7	(not part of simple effects analysis)				0.06	.81
Performance IQ	113.8	11.7	120.5	9.3	(not part of simple effects analysis)				4.3 4	. 0 4
Full scale IQ	116.1	7.7	119.7	9.4	(not part of simple effects analysis)				1.91	.17
WASI—Vocabulary	61.6	6.7	60.0	6.3	0.13	1.03	−0.1 6	0.99	0.69	.41
WASI—Similarities	57.2	4.6	57.9	6.0	−0.06	0.88	0.03	1.15	0.16	.70
WASI—Block Design	57.8	7.9	60.7	5.1	−0.22	1.17	0.23	0.77	2.11	.15
WASI—Matrix Reasoning	58.6	6.5	62.6	5.2	−0.33	1.05	0.31	0.86	5.37	.03
CTOPP Nonword repetition	6.9	1.8	8.5	1.8	−0.41	0.93	0.3 2	0.84	8.52	.01
TOLC Figurative Language	12.4	1.6	11.9	1.9	0.14	0.92	−0.15	1.11	0.88	.35
PPVT Receptive Language	1 0 8.0	10.0	111.4	10.4	−0.17	0.98	0.21	1.02	1.26	.27
EVT Expressive Language	110.8	11.4	109.4	14.8	0.05	0.87	−0.04	1.16	0.12	.73
Letter Fluency	11.7	2.5	10.6	3.0	0.20	0.89	−0.21	1.11	1.85	.18
Category Fluency	12.6	2.7	11.7	2.3	0.1 5	1.08	−0.08	0.86	1.03	.32
Category Switching	11.6	2.7	12.1	3.5	−0.0 8	0.8 6	0.13	1.1 4	0.28	.60
Switching Accuracy	12.5	2.4	13.1	2.9	−0.10	0.91	0.16	1.09	0.45	.51
Reading History	23.9	10.0	20.7	8.3	0.17	1.08	−0.12	0.88	1.31	.26

Note. WASI = Wechsler Abbreviated Scale of Intelligence. CTOPP = Comprehensive Test of Phonological Processing. TOLC = Test of Language Competence. PPVT = Peabody Picture Vocabulary Test. EVT = Expressive Vocabulary Test.