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Language-related abilities in 'unaffected' school-aged siblings of children with ASD

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ABSTRACT

Siblings of children with autism spectrum disorder (ASD) who do not have ASD outcomes are more likely than their peers to experience delays in language acquisition as preschoolers. However, less is known about how these siblings are faring when they are school-aged. We examined language-related abilities of 18 siblings with non-ASD outcomes, aged 8–11. On average, siblings performed more poorly than the normative sample in phonological memory and phonological awareness. In contrast, word-level reading was unimpaired. No deficits relative to norms were found on a direct child assessment and parent-report measure of pragmatic language; however, recommendations for further research in this area are made. Comparing two direct child assessments, this sample performed more poorly in pragmatics than receptive/expressive language. Language abilities in siblings of children with ASD is an area ripe for further investigation with larger sample sizes and appropriate comparison groups.

1. Introduction

Younger siblings of children with ASD are over 13 times more likely to develop ASD than their same-aged peers—prospective longitudinal studies place the recurrence risk at almost 1 in 5 siblings (Centre for Disease Control and Prevention, 2014; Ozonoff et al., 2011). Researchers often use the term unaffected to describe the 4 out of 5 siblings of children with autism spectrum disorder (ASD) who do not have ASD outcomes; however, emerging evidence suggests that the developmental trajectories of these siblings are often atypical (Brian et al., 2014; Gamliel, Yirmiya, Jaffe, Manor, & Sigman, 2009; Gross, Stuart, & Faherty, 2013). Many display subclinical ASD traits, referred to as the Broader Autism Phenotype (BAP; Georgiades et al., 2013; Sucksmith et al., 2011; Sucksmith, Roth, & Hoekstra, 2011; Szatmari et al., 2000), as well as poorer outcomes in cognition and differences in temperament and behaviour (Brian et al., 2008, 2014; Clifford et al., 2013; Constantino, Zhang, Frazier, Abbacchi, & Law, 2010; Garon et al., 2009; Schwichtenberg et al., 2013; Stone et al., 2007; Warren et al., 2012).

These ostensibly unaffected siblings are also at greater risk of delayed acquisition of language and other communication impairments during infancy and preschool (Hudry et al., 2014; Landa, Gross, Stuart, & Bauman, 2012; Messinger et al., 2013), but less is known about their language abilities later in childhood. To date, few published studies have followed siblings past age three, and significant methodological limitations have been identified with their case definitions and measurement

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(Drumm & Brian, 2013). It is crucial to extend studies beyond early childhood in order to determine whether early language delays persist or resolve, as well as to examine later-developing language skills such as pragmatics, phonological processing, and reading. As children age and experience increasing psychosocial demands, these language abilities become critical to academic and occupational success and social well-being.

Inherited genetic risk for ASD and shared genetic susceptibility across neurodevelopmental and psychiatric disorders may partly account for the broad array of developmental difficulties in siblings of children with ASD (Constantino et al., 2010; Lichtenstein et al., 2010; Jiang et al., 2013; Devlin & Scherer, 2012; Ronald & Hoekstra, 2011). It is also possible that environmental input and modelling from their older sibling with ASD may play a role in the language development of younger siblings of children with ASD; however, research is limited in this area. If such effects exist, we would expect that they would be strongest in the early developmental period; as the younger siblings grow older, begin school, and experience a wider array of peer interactions, any input effects may diminish as peers become more salient models.

In the past decade, research has begun to uncover the developmental risks and potential etiology of these risks for siblings of children with ASD. The present study will extend the literature by examining a range of language and language-related learning abilities in school-aged siblings with non-ASD outcomes. This study, as well as many of the studies described herein, is part of a series of prospective longitudinal studies of younger siblings of children with ASD recruited during infancy. Research from these studies provides a long-awaited opportunity to examine the risks and outcomes for siblings paired with solid methodology and growing sample sizes (see Zwaigenbaum et al., 2007). Here, we examine whether school-aged siblings[1] display deficits in receptive/expressive language, pragmatics, phonological processing, or word-level reading.

1.1. Receptive/expressive and pragmatic language

When comparing siblings of children with ASD to same-aged peers with no family history of ASD, the high-risk siblings are more likely to have early delays in language acquisition and demonstrate a lack of receptive-over-expressive advantage in early language development (Hudry et al., 2014; Landa et al., 2012; Messinger et al., 2013; Mitchell et al., 2006; Toth et al., 2007; Yirmiya et al., 2006; Yirmiya, Gamliel, Shaked, & Sigman, 2007; Zwaigenbaum et al., 2005). This same pattern is found in children with ASD: some children with ASD have very well-developed receptive/expressive language (Tager-Flusberg et al., 2005), but on the whole they are more likely have early delays in language acquisition and often fail to show the superior receptive-over-expressive language pattern that is typical of early childhood (Ellis Weismer, Lord, & Esler, 2010; Howlin, 2003; Volden et al., 2011). Children with ASD may struggle with higher-level aspects of syntax and morphology (Eigsti, Bennetto, & Dadlani, 2007), and this is another area that has yet to be explored in their siblings.

The majority of evidence that siblings with non-ASD outcomes are at greater risk for language delays comes from longitudinal studies that extend to age 3. We do not yet have sufficient research to conclude whether these siblings catch up to their same-age peers or continue to have receptive/expressive language difficulties beyond preschool. A prospective longitudinal study of high-risk siblings age 4–7 (Warren et al., 2012) and another of high-risk siblings age 9–12 (Ben-Yizhak et al., 2011) found unimpaired receptive/expressive language abilities in siblings compared to low-risk controls. In contrast, Gamliel et al. (2009) found deficits in receptive/expressive language in a subset of high-risk siblings. However, this study divided the siblings into two groups based on difficulties in language, cognition, or academic skills, rendering their result tautological.

Analogous to research on receptive/expressive aspects of language development, few conclusions can be drawn regarding the pragmatic language abilities of school-aged siblings. Pragmatics is the appropriate use of language in social contexts, and it is universally impaired in children with ASD (Ben-Yizhak et al., 2011; Bishop, 1989; Volden, Coolican, Garon, White, & Bryson, 2009), making it a natural candidate for investigation in siblings. Measurement limitation is a barrier to research on pragmatics; it can be challenging to collect standardized and quantifiable data while capturing the variety of appropriately contextualized pragmatic abilities that transpire in the complex social world (Drumm & Brian, 2013).

Bishop, Mayberry, Wong, Maley, and Hallmayer (2006) found no group differences between siblings with non-ASD outcomes and low-risk controls from parent-reports on the pragmatic scales of the Children's Communication Checklist (CCC-2; Bishop, 2003). However, a substantial 24% of the 42 siblings scored more than two standard deviations below the mean of the low-risk controls. In a longitudinal study, Ben-Yizhak et al. (2011) found pragmatic difficulties in a subset of siblings. This finding should be interpreted with caution as the ADOS was used to both divide the siblings into two groups (high or low algorithm scores) and to compare pragmatic abilities. As well, the ASD status of siblings was not systematically examined through diagnostic testing or clinical judgment. If any of the siblings in the analysis met criteria for ASD, this could have artificially inflated the scores.

1.2. Phonological processing and reading

Children with ASD are more likely than typically developing children to have deficits in phonological awareness, phonological memory, and rapid naming (Lindgren, Folstein, Tomblin, & Tager-Flusberg, 2009; Losh, Esserman, & Piven,

[1] For the purposes of this paper, when siblings is used as a stand-alone term it will always refer to children who do not have ASD themselves, but do have an older brother or sister (proband) with ASD.