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Early pragmatic language difficulties in siblings of children with autism: implications for DSM-5 social communication disorder?

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Background: We evaluated early pragmatic language skills in preschool-age siblings of children with autism spectrum disorder (ASD), and examined correspondence between pragmatic language impairments and general language difficulties, autism symptomatology, and clinical outcomes. Methods: Participants were younger siblings of children with ASD (high-risk, n = 188) or typical development (low-risk, n = 119) who were part of a prospective study of infants at risk for ASD; siblings without ASD outcomes were included in analyses. Pragmatic language skills were measured via the Language Use Inventory (LUI). Results: At 36 months, the high-risk group had significantly lower parent-rated pragmatic language scores than the low-risk group. When defining pragmatic language impairment (PLI) as scores below the 10th percentile on the LUI, 35% of the high-risk group was identified with PLI versus 10% of the low-risk group. Children with PLI had higher rates of general language impairment (16%), defined as scores below the 10th percentile on the Receptive or Expressive Language subscales of the Mullen Scales of Early Learning, relative to those without PLI (3%), but most did not evidence general language impairments. Children with PLI had significantly higher ADOS scores than those without PLI and had higher rates of clinician-rated atypical clinical best estimate outcomes (49%) relative to those without PLI (15%). Conclusions: Pragmatic language problems are present in some siblings of children with ASD as early as 36 months of age. As the new DSM-5 diagnosis of Social (Pragmatic) Communication Disorder (SCD) is thought to occur more frequently in family members of individuals with ASD, it is possible that some of these siblings will meet criteria for SCD as they get older. Close monitoring of early pragmatic language development in young children at familial risk for ASD is warranted. Keywords: Pragmatic language, social communication, autism spectrum disorder, social (pragmatic) communication disorder, siblings, high-risk.

Introduction

Siblings of children with autism spectrum disorder (ASD) are at heightened risk for a range of developmental concerns, the most well-known being ASD, with an estimated recurrence rate of nearly 20% (Ozonoff et al., 2011). Yet it is becoming increasingly apparent that this population is also at greater risk for other atypical outcomes, including learning difficulties (Gamliel, Yirmiya, Jaffe, Manor, & Sigman, 2009), behavior problems (Schwichtenberg et al., 2013), and broader autism phenotype (BAP) characteristics (reviewed by Sucksmith, Roth, & Hoekstra, 2011), including language impairments (Drumm & Brian, 2013). Studies of language abilities in infant siblings of children with ASD who do not go on to develop ASD themselves are mixed. Some have found below average expressive and receptive language skills in a subgroup of this population (Gamliel et al., 2009; Yirmiya, Gamliel, Shaked, & Sigman, 2007), highlighting a potential heightened risk for language delay, while other studies have not (Hudry et al., 2014; Warren et al., 2012).

Pragmatic language and social communication deficits are hallmark characteristics of ASD (Tager-Flusberg et al., 2009), making them important to evaluate in siblings. In fact, the new *DSM-5* diagnosis of Social (Pragmatic) Communication Disorder (SCD), which is aimed at identifying children with

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Fewer studies have evaluated early pragmatic language development in this population, particularly not prior to the school-age years. Definitions of pragmatic language vary, but most involve the appropriate use of language in context, including the use of both verbal and nonverbal information (Bryant, 2009) to convey communicative intentions, make inferences about social partners and/or similar situations, and manage discourse (Landa, 2005). However, some definitions of pragmatic language focus more specifically on inference and nonliteral meaning via the use of linguistic context, with strong links to components of structural language, whereas others focus on aspects of social communication involving the use of language in social contexts (see Norbury (2014) for a helpful conceptualization). Although distinctions do exist between pragmatics and social communication skills, there is also clear overlap, and these terms are often used interchangeably.

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pragmatic language impairments, is described by *DSM-5* as potentially occurring more frequently in individuals with a family history of ASD (American Psychiatric Association, 2013). One study focusing on school-age outcomes of siblings of children with ASD found lower pragmatic abilities in a subgroup characterized by BAP characteristics (Ben-Yizhak et al., 2011), while another did not find evidence of impairment among siblings of children with ASD (Gillespie-Lynch et al., 2013). Thus, although several studies suggest that siblings of children with ASD are at risk for delays in pragmatic language by school-age, it remains unclear when these delays emerge and how they relate to other domains of functioning.

Pragmatic language skills have been shown to be critical to multiple developmental outcomes. For example, recent investigations have highlighted the importance of pragmatic language skills in the development of peer relations (Mok, Pickles, Durkin, & Conti-Ramsden, 2014), and have linked problems in pragmatic language to both internalizing and externalizing behavior problems (Ketelaars, Cuperus, Jansonius, & Verhoeven, 2010). Thus, assessing early pragmatic language deficits may be one important avenue for identifying those children at heightened risk for negative outcomes. Because of this, and because younger siblings of children with ASD are at greater risk for a range of negative developmental outcomes (e.g., Ozonoff et al., 2014), better understanding the nature of early pragmatic language abilities in this group could have important consequences.

The present investigation had four primary goals. First, we aimed to evaluate early (36-month) pragmatic language abilities in non-ASD siblings of children with ASD (high-risk group) relative to siblings of typically developing children (low-risk group). Second, we sought to examine the correspondence between categorically defined pragmatic language and general language impairments (i.e., receptive/expressive language). Third, we aimed to evaluate differences in autism symptomatology between children with and without pragmatic language impairment (PLI).¹ Finally, we sought to evaluate the correspondence between PLI and expert judgments of clinical outcomes. We hypothesized that: (a) the high-risk group would evidence lower pragmatic language scores and higher rates of impairment relative to the low-risk group, (b) those with PLI would evidence higher rates of general language impairments than those without pragmatic language difficulties, (c) those with PLI would display higher levels of autism symptomatology than those without PLI, and (d) those with PLI would evidence higher rates of atypical clinical best estimate outcomes by expert examiners relative to those without pragmatic language problems. Because of the very young age of the children in the present investigation, and because higher level pragmatic language abilities, including the ability to infer meaning and resolve ambiguities via linguistic context, develop later in childhood (Adams, 2002), we focus on the broader conceptualization of pragmatic language that emphasizes the use of language in social contexts (Bates, 1976; Landa, 2000). More specifically, our focus is on earlier developing aspects of pragmatic language such as the child's ability to direct the attention of others, to talk or ask about others' behaviors and mental states, to tease, and to take the listener's knowledge into account.

Method

Participants

The sample was drawn from a larger prospective longitudinal study of younger siblings of children with ASD (high-risk group) or typical development (low-risk group), recruited at two sites during two phases of data collection. The primary inclusion criterion for the high-risk group was status as a younger sibling of a child with ASD. Diagnosis of the affected older sibling was confirmed by meeting ASD criteria on both the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) and the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003). Exclusion criteria for the high-risk group included birth before 36 weeks gestation and a known genetic disorder (e.g., Fragile X syndrome) in the older affected sibling. The primary inclusion criterion for the low-risk group was status as a younger sibling of a child with typical development. Low-risk status of older siblings was confirmed by an intake screening questionnaire and scores below the ASD range on the SCQ. Exclusion criteria for the low-risk group were birth before 36 weeks gestation, developmental, learning, or medical conditions in any older sibling, and ASD in any first-, second-, or third-degree relative.

Participants were enrolled before 18 months of age (average = 8.4 months, SD = 3.93 months); 74% were enrolled by 9 months of age. Informed consent was obtained from parents. Participants were assessed by expert clinical examiners unaware of group membership; ongoing administration and scoring fidelity procedures were implemented to ensure minimal cross-examiner/cross-site differences. Although data were collected at up to five ages, the present investigation focuses only on 36-month data. At the 36-month assessment, participant outcomes were determined; participants were diagnosed with ASD if they received ADOS scores over the ASD cutoff and met DSM-IV criteria for Autistic Disorder or Pervasive Developmental Disorder-Not Otherwise Specified by an expert clinician. However, because we were interested in determining whether known pragmatic language difficulties in ASD extend to non-ASD siblings, we focus only on participants with non-ASD outcomes. Thus, all participants with non-ASD outcomes and with complete pragmatic language, cognitive, and diagnostic data at the 36-month outcome visit were included resulting in a final sample of 307 participants: 119 low-risk (n = 52 females) and 188 high-risk non-ASD (n = 95females). The study was conducted under the approval of the University's Institutional Review Boards.

Measures

Autism Diagnostic Observation Schedule (ADOS)

The ADOS (Lord et al., 2000) is a semi-structured standardized interaction and observation that measures symptoms of autism. It has two empirically derived cutoffs, one for ASD and one for Autistic Disorder. As much of these data were collected prior to the publication of the newer ADOS algorithms, we used the Communication + Social Total algorithm score (Lord et al., 2000). Psychometric studies report high inter-rater reliability and agreement in diagnostic classification (autism vs. nonspectrum). The ADOS was used for diagnostic classification purposes in both the older sibling (i.e., to verify inclusion criteria) and the participant (i.e., to determine outcome at 36 months of age). The ADOS also provided a dimensional measure of autism symptomatology.

Mullen Scales of Early Learning (MSEL)

The MSEL (Mullen, 1995) is a standardized developmental test for children birth to 68 months and was used to evaluate cognitive functioning. Four subscales were administered: Fine Motor, Visual Reception, Expressive Language, and Receptive Language. Raw scores convert to T-scores and an overall composite score of all four subscales, the Early Learning Composite (ELC), is obtained. MSEL subscales have excellent internal consistency (median 0.91) and test-retest reliability (median 0.84). Because pragmatic language difficulties could be related to lower overall cognitive ability, and because we were primarily interested in identifying pragmatic language difficulties over and above overall cognitive ability, the ELC at 36 months was used as a covariate in relevant analyses. General language impairment was defined as scores at or below the 10th percentile on either the Expressive or Receptive Language subtests of the MSEL; the 10th percentile is a commonly used clinical cutoff for defining language impairment (Paul & Norbury, 2012).

Language Use Inventory (LUI)

The LUI is a standardized, norm-referenced parent report that was designed to evaluate pragmatic language development in children 18-47 months (O'Neill, 2009). It focuses on the use of language in social contexts in daily life, highlighting the inherently social nature of language. The LUI was developed with a focus on very young children's language use/pragmatic competence as influenced by their developing theory of mind (O'Neill, 2005), with a primary focus on communicative function. Because of the young age of children for whom this measure was developed, there is less focus on the laterdeveloping aspects of pragmatic language such as making inferences about indirect meanings and higher level metapragmatic skills. Instead, the LUI indexes the pragmatic and social communication skills that develop between 18 and 47 months of age. The LUI has been identified as a relevant tool to assess pragmatic language skills in very young children with or at risk for ASD (Tager-Flusberg et al., 2009).

The LUI has good internal consistency (0.79-0.99) and testretest reliability (0.85-0.96). There are 10 scored subscales that are aggregated to yield a Total Score (range 0–161), which can then be converted to age- and gender-based percentile ranks. Examples of skills assessed include requests for help, declarative use of words, using language to comment and/or gain information, using language to regulate the actions of others, teasing and sense of humor, adapting conversations to others, and more sophisticated discourse and narrative skills. In addition to differentiating children with language impairment from those without (O'Neill, 2007), LUI scores have been shown to predict later structural and pragmatic language outcomes (Pesco & O'Neill, 2012).

Parents completed the LUI at the 36-month visit. Total scores were log transformed to meet normality assumptions; higher scores equate to better pragmatic language ability. Categorical impairment in pragmatic language was identified using a cutoff of <10th percentile for the LUI Total Score; this cutoff has been shown to yield high sensitivity and specificity for predicting later language impairment (Pesco & O'Neill,

2012) and the 10th percentile is a commonly used clinical cutoff for defining language impairment (Paul & Norbury, 2012).

Clinical Best Estimate (CBE) outcome classification

At the end of the 36-month visit, expert clinical examiners classified each child into one of six clinically defined outcome categories based on clinical judgment: ASD, BAP, ADHD Concerns, Global Developmental Delay, Speech-Language Problems, or Typical Development. Children classified with ASD met DSM-IV criteria for Autistic Disorder or PDD-NOS; as noted previously, these children were excluded from analyses. All other clinical outcome categories were not intended to correspond with specific DSM diagnoses, but instead reflected clinical concerns. Specifically, children classified with BAP displayed social communication difficulties below the ASD threshold. Children classified as having ADHD Concerns displayed developmentally atypical levels of hyperactivityimpulsivity, inattention, and/or disruptive behavior. Children classified with Global Developmental Delay had consistently low scores across cognitive and motor domains. Children classified as having Speech-Language Problems displayed immature speech patterns or low language levels based on standardized testing. All other participants were classified as Typically Developing.

Data analytic plan

Analyses were performed using SPSS for Macintosh, Version 21 (IBM Corporation, 2013). Continuous variables were first standardized and outliers Winsorized at ± 3.00 (i.e., 3 SD; Tabachnick & Fidell, 2001). Table 1 contains descriptive statistics, presented as unadjusted scores for ease of interpretation. First, group differences in continuous LUI Total Scores were evaluated via Analysis of Covariance (ANCOVA), with risk group (high vs. low) as the independent variable, and covariates of site, phase, gender, and 36-month MSEL ELC (i.e., cognitive ability). Next, risk group differences in rates of PLI were evaluated using a chi-square test. In evaluating the remainder of our research questions, analyses were first conducted across the entire sample, and then within the high-risk group only to clarify the effect of pragmatic language problems within the high-risk group alone. To address our second goal, we examined the correspondence between PLI and general language impairments using a chi-square test. Next, we used ANCOVA to evaluate group differences in autism symptomatology (i.e., ADOS scores) in those children with and without PLI, including the covariates of site, phase, gender, and 36-month MSEL ELC. Finally, CBE outcomes for those identified as impaired were examined descriptively and via a chi-square test.

Results

Risk group differences in pragmatic language and categorical impairment

The ANCOVA revealed significantly lower LUI Total Scores in the high-risk group relative to the low-risk group, accounting for covariates of phase, site, gender, and MSEL ELC, F(1, 301) = 18.41, p < .001. Using a 10th percentile cutoff for the LUI to examine rates of PLI (impaired vs. unimpaired), the high-risk group showed a higher rate of impairment (35%, n = 65) relative to the low-risk group (10%, n = 12), $\chi^2 = 23.26$, p < .001, odds ratio (OR) = 4.71 (CI_{0.95} = 2.42, 9.19).

Table 1	Unadjusted	descriptive	characteristics	for high- and	low-risk groups	at 36-months
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	Low-Risk (<i>n</i> = 119)		High-Risk (<i>n</i> = 188)			
	Mean	SD	Mean	SD	t	Cohen's d
Mullen Scales of Early Learning						
Visual Reception	61.38	10.97	61.12	12.56	0.19	0.02
Fine Motor	52.36	12.70	51.92	13.04	0.29	0.03
Expressive Language	56.06	7.27	53.06	8.86	3.09**	0.37
Receptive Language	53.82	9.59	50.07	8.59	3.55**	0.41
Early Learning Composite	111.76	15.23	108.19	16.88	1.87	0.22
ADOS communication + social	2.29	1.97	3.10	2.38	-3.26**	0.37
Language Use Inventory total	134.72	18.25	118.32	28.51	5.59***	0.69

****p* < .001; ***p* < .01.

Note: Mullen Scales of Early Learning subscale scores are T-scores (mean = 50, SD = 10); Mullen Early Learning Composite scores are Standard Scores (mean = 100, SD = 15). Language Use Inventory Total scores are raw scores.

Correspondence between PLI and general language impairment

Given that criteria for the new diagnosis of SCD require that pragmatic deficits are not solely accounted for by general language impairments, we examined the proportion of those children identified with PLI in our sample who also had impairments in general language skills. Across the entire sample, children identified with PLI showed a higher of rate of general language impairment (16%, n = 12) relative to those without PLI (3%, n = 7), $\chi^2 = 15.63$, p < .001, OR = 5.88 (CI_{0.95} = 2.23, 15.55), although, notably, the vast majority (84%, n = 65) of children with PLI did not evidence general language impairment. This finding was virtually identical when restricting analyses to just the high-risk group: Those children in the high-risk group with PLI showed a higher rate of general language impairment (17%, n = 11) relative to those without PLI (3%, n = 4), $\gamma^2 = 10.83$, p = .001, OR = 6.06 (CI_{0.95} = 1.85, 19.89); 83% of children in the high-risk group (n = 54) identified with PLI *did not* evidence general language impairment.

Pragmatic language and autism symptomatology

To determine whether children with PLI evidenced higher levels of autism symptomatology, we compared ADOS scores in those with and without PLI, accounting for relevant covariates. Across the whole sample, ADOS algorithm scores were significantly higher in those with PLI (mean = 4.04, SD = 2.67) relative to those without PLI (mean = 2.37, SD = 1.94), F(1, 301) = 16.81, p < .001, Cohen's d = 0.72. Similarly, within the high-risk group alone, those with PLI had higher ADOS algorithm scores (mean = 3.94, SD = 2.70) relative to those without PLI (mean = 2.66, SD = 2.61), F(1, 182) = 5.72, p = .018, Cohen's d = 0.48.

Correspondence between PLI and expert clinical judgment

To identify how PLI classifications corresponded with examiner-rated outcome classifications, we examined CBE outcomes for those with PLI in the highand low-risk groups (see Table 2). For the 65 children in the high-risk group who evidenced impairment, 54% (n = 35) received atypical CBE outcome ratings and 46% (n = 30) received CBE outcomes of typical development. In contrast, of the 12 low-risk children identified with PLI, only 25% (n = 3) received atypical CBE outcome ratings, while 75% (n = 9) were rated by examiners as having CBE outcomes of typical development. Across the entire sample, children with PLI, based on parent-reported LUI scores, had a significantly higher rate of atypical CBE outcomes (49%, n = 38) relative to those without PLI (15%, n = 34), $\chi^2 = 38.40$, p < .001, OR = 5.62 ($CI_{0.95} = 3.16$, 10.00). This finding was similar when restricting the analysis to just the highrisk group: Those in the high-risk group with PLI had higher rates of atypical CBE outcomes (54%, n = 35)

Table 2 Clinical best estimate outcomes for those children identified with pragmatic language impairment by risk group

	Low-risk		High-risk		
	Pragmatic Language Unimpaired (<i>n</i> = 107)	Pragmatic Language Impaired (n = 12)	Pragmatic Language Unimpaired (n = 123)	Pragmatic Language Impaired (<i>n</i> = 65)	
Typically developing	96	9	100	30	
Broader autism phenotype	4	3	14	16	
Speech-language problem	3	0	3	10	
ADHD concerns	4	0	5	6	
Global developmental delay	0	0	1	3	

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relative to those without PLI (19%, n = 23), $\chi^2 = 24.63$, p < .001, OR = 5.07 (CI_{0.95} = 2.61, 9.87).

Discussion

In the present investigation, we sought to characterize early pragmatic language abilities in young children with family histories of ASD, and to evaluate correspondence between deficits in pragmatic language and other aspects of functioning (i.e., general language abilities, autism symptomatology, and clinical outcome). Comparisons revealed significantly lower parent-rated pragmatic language scores in the high-risk group, consistent with previous work focusing on school-age outcomes of siblings of children with ASD (e.g., Ben-Yizhak et al., 2011) and indicating that vulnerabilities in pragmatic language are evident as early as 36 months of age in younger siblings of children with ASD. Additionally, over three times as many children in the high-risk group (35%) were identified with PLI at 36 months than in the low-risk group (10%), consistent with a study by Bishop, Maybery, Wong, Maley, and Hallmayer (2006) which found that 24% of a non-ASD sibling sample displayed scores more than 2 SD below the control groups' mean on the Children's Communication Checklist. In the present investigation, more than half of high-risk children with PLI evidenced clinician-rated atypical outcomes, versus one-quarter of low-risk children. Notably, across risk groups, approximately half of the children identified with PLI were judged by examiners to be typically developing. There are several possible reasons for this. First, these PLI cases may be "false positives," in that using a 10th percentile cutoff on the LUI may capture a proportion of children who do not actually have pragmatic language problems. Alternatively, it may be that the pragmatic language difficulties experienced by these children are not prominent enough to be noted by examiners in the context of a one-time laboratory visit, but are more apparent to parents, who experience their child's language skills on a daily basis. These are important questions for future research.

We also examined how PLI corresponded with general language impairments. The vast majority (over 80%) of children identified with PLI did not evidence general language impairments, suggesting that early pragmatic and social communication impairments are not easily accounted for by general language deficits among younger siblings of children with ASD. Additionally, we found significantly higher ADOS scores in the pragmatically impaired group relative to those without PLI, indicating that, perhaps not surprisingly, children with parent-rated pragmatic language deficits also had greater examiner-rated impairment in social communication. Analyses focused solely on the high-risk group yielded similar results. The agreement between methods supports the validity of the parent reports

and further highlights the links between pragmatic language and social communication skills (Norbury, 2014). Importantly, because restricted and repetitive behaviors were not examined in the present study, whether those children with PLI also show higher levels of this aspect of ASD symptomatology remains a question for future research.

A timely issue relates to the new *DSM-5* diagnosis of SCD, which (per DSM-5) may be more common in individuals with a family history of ASD. Yet there are substantial concerns about the validity of this diagnosis (see Norbury, 2014), including whether it will serve as a "catch-all" for children with subthreshold ASD symptoms. Indeed, the most frequently rated CBE outcome in our subsample of high-risk non-ASD children identified with PLI was BAP (which, we emphasize, is not a diagnosis itself and, by definition, does not entail clinically significant impairment). Thus, those with clinician-categorized subthreshold ASD symptoms comprised the largest proportion of the high-risk group with PLI. Although it has previously been questioned whether PLI and ASD are diagnostically distinct (e.g., Gagnon, Mottron, & Joanette, 1997), recent research has found that, in fact, it is possible to distinguish children with PLI from children with other ASD-related characteristics (Gibson, Adams, Lockton, & Green, 2013). Our findings highlight that although the group of children with PLI evidenced higher rates of atypical clinical outcomes and higher levels of autism symptomatology, pragmatic language difficulties were also present in a proportion of children who were identified as typically developing. Notably, by design, none of the children in this sample met criteria for ASD, further emphasizing that difficulties with pragmatic language do occur outside of ASD in children as young as 36 months.

A related issue pertains to the age at which SCD can be identified. The DSM-5 indicates that SCD may not be detectable before the age of 4 years, as some pragmatic abilities may not be developed enough even in typically developing children to capture deficits at this young age. Indeed, several of the SCD criteria describe deficits in skills that would generally not be expected to develop until the early school-age years (e.g., inferring indirect meanings, refined conversational turn-taking skills, higher level meta-pragmatic skills; see Adams, 2002). The LUI's intentional focus on a broader conceptualization of pragmatic language incorporates aspects of social communication (as is also done in the SCD criteria). Based on this, our data suggest that, on average, pragmatic vulnerabilities in siblings of children with ASD are detectable as early as 36 months of age. However, given that SCD criteria require that a child demonstrate all four symptoms (i.e., difficulties in using communication for social purposes, changing communication to fit the context or needs of the listener, following rules of conversation and

narrative, and using and understanding nonliteral language), including those that develop later in childhood, it is unlikely that any of our participants would have met criteria, even had they been available at the time of data collection. A crucial question is whether there is developmental continuity between the classifications of PLI by the LUI in this sample of 3-year-old children with a family history of ASD, and PLI classifications at later ages using instruments like the Children's Communication Checklist (Bishop, 2003), which focus on laterdeveloping aspects of pragmatic language. Additionally, whether those children identified as pragmatically impaired in the present sample actually evidence clinically significant impairment, and/or would meet criteria for SCD at older ages, remain important questions. Continued follow-up of infant sibling samples will allow for clarification of these points.

Limitations of the present study include the use of indirect measurement strategies to evaluate pragmatic skills. The use of multiple informants and direct measurement strategies in future investigations will provide a more comprehensive view of these abilities. We also did not include those children with ASD outcomes; future research should examine similar questions within this population. Additionally, pragmatic language abilities are just beginning to develop at 36 months of age (Bryant, 2009) and few measures exist to assess these abilities early. Thus, the LUI is unique in this respect, but because of its focus on very young children between the ages of 18 and 47 months, it cannot evaluate aspects of later-developing pragmatic abilities such as inferring indirect meanings, understanding and using metaphors and idioms, and other more complex narrative and discourse skills (see Adams, 2002). This complicates the discrimination of pragmatic language from social communication, potentially resulting in higher rates of pragmatic language impairment in our high-risk sample, who may be more vulnerable to social communication deficits by virtue of having a family history of ASD. In addition, the sampling method used in the present study may have impacted our finding of higher rates of PLI in the high-risk group. For example, parents of children with ASD may be more inclined to enroll their new infant in the study if they have concerns about the infant's development, thus biasing the sample toward atypical outcomes. While community-based sampling methods (e.g., Sandin et al., 2014) are preferable in reducing selection biases, the very large scale of such an approach makes standardized direct assessments of development (which were central to the present investigation), using gold-standard tools, impossible. Given the early ages at which infants were enrolled in our study (74% enrolled by 9 months), we do not believe that this

issue significantly influenced our findings. Because early signs of ASD and other developmental delays do not tend to emerge until 12 months of age or later (Ozonoff et al., 2010, 2014), few parents of children in the high-risk group were likely to have been concerned about their infant at time of enrollment in the study.

Our findings have several implications for SCD. First, pragmatic language/social communication deficits (which comprise the SCD criteria) do appear to be more common in unaffected family members of children with ASD as early as 36 months of age and occur in the absence of cooccurring general language deficits in most cases. However, given that the criteria for SCD focus on later-developing aspects of pragmatic language, it is unlikely that any of the children in our sample would have met criteria for SCD, had the criteria been available at the time of the 36month outcome assessments. A primary challenge in applying these criteria will likely be the requirement that all four symptoms be present, suggesting that only substantially affected children will meet criteria for SCD, not unlike children with ASD themselves (Ozonoff & Miller, in press). Our findings highlight the potential importance of evaluating the early development of pragmatic language abilities in very young children with family histories of ASD. Indeed, a recent study showed that children with pragmatic language difficulties were at greatest risk for the development of peer difficulties (Mok et al., 2014) and other studies have linked pragmatic language deficits to behavior problems (Gremillion & Martel, 2013; Ketelaars et al., 2010). Thus, addressing such difficulties early in life may result in better outcomes across multiple domains over time. Overall, close monitoring of pragmatic language development, and social communication development more broadly, in children at risk for ASD may be warranted, and early assessment of pragmatic language may benefit early detection of children at risk for later difficulties.

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Key points

- Siblings of children with ASD are at heightened risk for pragmatic language difficulties as early as 36 months of age.
- Children with pragmatic language impairment are more likely to evidence general language impairments, to
 exhibit higher levels of autism symptomatology, and to receive atypical clinical outcomes than those children
 without pragmatic language impairment.
- Close monitoring of early pragmatic language development in young children at familial risk for ASD may be warranted and may benefit early detection of children at risk for later difficulties.

Note

1. Our use of the term 'PLI' is not intended to convey a clinical diagnosis; rather, for the purposes of this study, we simply define PLI as scores below the 10th percentile on the Language Use Inventory.

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