Journal of Autism and Developmental Disorders (2019) 49:1352–1365
https://doi.org/10.1007/s10803-018-3837-x

ORIGINAL PAPER

Predictors of Pragmatic Communication in School-Age Siblings of Children with ASD and Low-Risk Controls

Kathryn J. Greenslade[1,2] · Elizabeth A. Utter[2] · Rebecca J. Landa[2,3]

Published online: 28 November 2018

Abstract

Little empirical evidence exists about school-age pragmatic communication or predictors in siblings at heightened familial risk for ASD (HR) and low-risk (LR) controls. The *Pragmatic Rating Scale-School-Age* (Landa unpublished) was scored for 49 HR siblings and 18 LR controls at 8–12 years. Social-communication and language measures were collected between 14 and 36 months. At 36-months, siblings were classified as ASD (HR-ASD, n = 15), broad autism phenotype (HR-BAP, n = 19), or typically developing (HR-TD, n = 15). Results revealed a pragmatic continuum with significantly better scores for HR-TD than HR-BAP or HR-ASD, and HR-BAP than HR-ASD. Per regression models including all participants, 14-month joint attention initiations predicted school-age pragmatic communication, as did 24-month social-communication and expressive language scores. Early joint attention, social-communication, and language abilities contribute to later pragmatic functioning.

Keywords Pragmatic language · High risk siblings · Broad autism phenotype · Joint attention · Social-communication · Autism

Pragmatic communication, including the social use of language and nonverbal communication behaviors, is definitively impaired in children with autism spectrum disorder (ASD) (APA 2013; Paul et al. 2009; Simmons et al. 2014). Ineffective pragmatic communication may result in communication breakdowns and adversely affect self-esteem, peer acceptance, and perceptions of social competence in everyday interactions (Prutting and Kirchner 1987; Turkstra et al. 2017). First observed in parents and later in siblings of children with ASD, subclinical social and communication differences, referred to as the broad autism phenotype (BAP), are present in some family members (Losh and Piven 2007; Landa et al. 1992). One notable manifestation of the BAP is divergence from normative pragmatic behavior. Minimal research has focused on pragmatic communication in siblings characterized as BAP (Ben-Yizhak et al. 2011; Bishop et al. 2006; Drumm and Brian 2013; Drumm et al. 2015; Miller et al. 2015) or on defining predictors of pragmatic functioning (Gillespie-Lynch et al. 2015). In the current study, the *Pragmatic Rating Scale for School-Age Children* assessed the pragmatic communication of 8- to 12-year-olds at high familial risk for ASD (younger siblings of a child with ASD) and low-risk controls, and examined early predictors of these abilities across the full sample.

Pragmatic Communication and ASD-Related Characteristics

Pragmatic communication includes linguistic and nonverbal behaviors. Pragmatic language refers to the use of spoken language to effectively convey messages across diverse social-communication contexts and types of exchanges (Roth and Spekman 1984a). It may be conceptualized as encompassing three categories of tacit rules (Grice 1975) for social language use: speech acts, discourse management, and presuppositional skills (Roth and Spekman 1984a). Speech acts relate to a linguistic message's function, including greeting, requesting information or objects, commenting, and predicting. Discourse

✉ Rebecca J. Landa
landa@kennedykrieger.org

[1] Present Address: Department of Communication Sciences and Disorders, University of New Hampshire, Durham, NH, USA

[2] Kennedy Krieger Institute, Center for Autism and Related Disorders, Baltimore, MD, USA

[3] Department of Psychiatry and Behavioral Sciences, Johns Hopkins University School of Medicine, Baltimore, MD, USA

management refers to the rules partners follow to promote the smooth flow of conversation, wherein partners take turns, and initiate, maintain, and elaborate contingently upon established topics. Presuppositional skills encompass communicative behaviors that reflect understanding of others' perspectives and sharing of appropriate information for a specific partner or situation, insuring that messages comply with Gricean Maxims (1975) (e.g., do not provide too much/too little information; do not be obscure/ambiguous). Nonverbal aspects of pragmatic communication, including: gestures; facial expressions; eye contact; vocal intonation, rate, and loudness; and management of interpersonal space may augment or detract from a speaker's meaning and the social appropriateness of the messages conveyed. While nonverbal communication behaviors are distinct from pragmatic *language*, both contribute to pragmatic *communication* and often are assessed concurrently.

Individuals with ASD demonstrate pragmatic communication impairment. Reduced range of speech acts is punctuated by diminished rates and quality of sharing interests/emotions, decreased frequency of acknowledgements and requests for information, and difficulty understanding and expressing nonliteral language and humor (APA 2013; Donaldson and Olswang 2009; Happé 1994; Lam and Yeung 2012; Ozonoff and Miller 1996; Simmons et al. 2014; Tager-Flusberg and Caronna 2007). Discourse management difficulties manifest as reduced reciprocity and turn-taking, decreased frequency of initiations, and difficulty maintaining and elaborating on others' topics (Hale and Tager-Flusberg 2005; Lam and Yeung 2012; Paul et al. 2009; Simmons et al. 2014; Tager-Flusberg and Caronna 2007). Presupposition difficulties manifest as: being overly candid, direct, or formal in articulation or vocabulary use; providing insufficient information (e.g., using ambiguous referents); providing irrelevant details; being "overly talkative" (e.g., seeming unaware that they have held the conversational floor too long); and/or formulating inefficient or disorganized messages (Lam and Yeung 2012; Paul et al. 2009; Simmons et al. 2014; Tager-Flusberg and Caronna 2007). Nonverbal pragmatic communication difficulties include: difficulty appropriately using gestures and facial expressions to augment spoken messages; poorly modulating eye contact, intonation, rate, or vocal loudness; difficulty integrating others' verbal messages with their nonverbal cues to interpret intended meaning; and poorly managing interpersonal space (APA 2013; Gessaroli et al. 2013; Paul et al. 2009; Tager-Flusberg and Caronna 2007).

The Broad Autism Phenotype and Pragmatic Communication

Twin studies (e.g., Folstein and Rutter 1977) indicated heritability of ASD. Subsequent family studies revealed subclinical features in family members of individuals with ASD, which paralleled ASD symptomatology (Landa et al. 1992; Piven et al. 1997). The subclinical characteristics (referred to as the BAP) include pragmatic language and narrative discourse differences from the norm (Landa et al. 1991, 1992; Losh and Piven 2007).

BAP characteristics were first defined using the *Pragmatic Rating Scale* (*PRS*; Landa et al. 1992) based on conversation samples. The *PRS* demonstrated strong inter-rater reliability, test–retest reliability, internal consistency, and construct validity, supported by factor analysis and significantly higher (greater variation from the norm) *PRS* total scores in parents of children with ASD than in parents of controls. Landa et al.'s (1992) finding was substantiated by self- and informant-reported poorer pragmatic functioning in parents of children with ASD on the Broad Autism Phenotype Questionnaire (Hurley et al. 2007).

Similarly, accumulated evidence revealed BAP characteristics in 18–28% of high-risk (HR) siblings of children with ASD in early childhood (Georgiades et al. 2013; Landa et al. 2007; Messinger et al. 2013; Ozonoff et al. 2014). Limited research has examined pragmatic communication in HR siblings, with most using parent-report rather than direct observation measures, due to measurement challenges. Gillespie-Lynch et al. (2015) and Bishop et al. (2006) found no significant pragmatic communication differences between school-aged HR siblings without ASD ('unaffected' siblings) and low-risk (LR) controls, on the *Children's Communication Checklist-2* (*CCC-2*; Bishop 2003), a parent-report measure. However, compared to controls, HR siblings' scores were significantly more variable, and a higher proportion of HR siblings demonstrated depressed *CCC-2* Coherence, Use of Context, and Non-Verbal Communication subscale scores (> 1 SD below the mean; Bishop et al. 2006). Drumm et al. (2015) found *CCC-2* scores and mean scores from a standardized pragmatics assessment of 8- to 11-year-old HR siblings without ASD were significantly better than expected or not significantly different from the test norms. However, 17% of participants demonstrated depressed pragmatics scores (> 1 SD below the mean). These findings highlight the need for direct assessment of HR siblings' pragmatic communication in an ecologically valid context.

Amongst HR siblings without ASD, pragmatic language impairments are most likely in those who also display higher ASD symptomatology (Ben-Yizhak et al. 2011; Miller et al. 2015). At age 36 months, 35% of 188 HR siblings met the parent-reported *Language Use Inventory* (O'Neill 2007) cutoff for pragmatic language impairment (< 10th percentile), a higher proportion than in the 119 LR controls (Miller et al. 2015). Pragmatic language impairment was associated with significantly higher *Autism Diagnostic Observation Schedule* (*ADOS*; Lord et al. 1999) algorithm scores, signaling higher ASD symptomatology than in children without such deficits (Miller et al. 2015). Ben-Yizhak et al. (2011) identified poorer pragmatic language abilities in 13 school-age HR siblings with elevated *ADOS* algorithm scores (≥ 4) who did not meet ASD criteria (referred to as BAP-related difficulties), as compared to 19 typically developing HR siblings and 38 controls. In that study, pragmatic communication was measured using a set of *ADOS* item-level scores, including item-level scores that contribute to *ADOS* algorithm scores, which had been used to define those with BAP-related difficulties. While the lack of independence between these measures might call the results into question, poorer pragmatics among siblings with BAP also were found using pragmatic language-related *ADOS* items that did not overlap with algorithm scores (Ben-Yizhak et al. 2011). Thus, a subset of HR siblings appears to demonstrate pragmatic language differences that may extend into the impaired range.

Predictors of School-Aged Pragmatic Communication

Predictors of school-age pragmatic communication functioning in HR siblings have received scant attention (Gillespie-Lynch et al. 2015). In the only identified study examining predictors, 12-month joint attention initiations and responses were not associated with school-age *CCC-2* pragmatic functioning in a combined group of HR siblings without ASD and LR controls; 18-month joint attention responses trended toward predicting school-age pragmatic language in this group (Gillespie-Lynch et al. 2015).

In typical development, early joint attention, broader social-communication behaviors (e.g., shared enjoyment, gesture use), and structural expressive language are the theoretical precursors of advanced social cognition, which is essential to appropriate school-age pragmatic communication (Malesa et al. 2012; Mundy et al. 2007; Landa et al. 2007; Tomasello 2001; Van Hecke et al. 2007). According to the parallel and distributed processing model (PDPM) of joint attention, young children integrate information about the object of their own visual attention with other knowledge that they have acquired while processing information about the visual attention of others. The result is a cognitive synthesis of information from these sources; this synthesis is the essence of joint attention (Mundy et al. 2009). With advances in development, the processing system (explained in the PDPM) supporting joint attention is characterized by increasing efficiency and complexity in children's processing of their own (internally derived) visual experiences, the information (externally derived) they gleaned from the visual attention of others, and the integration (parallel processing) of the two. This information processing system provides a foundation for the development of symbolic language and social learning, and continues to function throughout the lifespan, supporting social-cognition/presupposition. All of these competencies facilitate topic initiation and maintenance within real-time exchanges (Mundy et al. 2009; Turkstra et al. 2017). Although Tomasello et al.'s (2005) social-cognitive model proposes a different evolution of joint attention (i.e., "true" joint attention relying on an understanding of others' intentions, and symbolic language "replacing" joint attention as a means of covertly coordinating mental attention), this model likewise theorizes that joint attention supports the development of symbolic language, social cognition, and behavior aligned with social norms (Mundy et al. 2009; Tomasello et al. 2005). Consistent with these models, 9- to 18-month-olds' response to joint attention and initiation of joint attention predict 24-month (Mundy et al. 2007) and 5-year (Malesa et al. 2012) structural language as well as 30-month social competence (based on parent-report of compliance, empathy, pretend play, prosocial peer interactions, etc.) (Van Hecke et al. 2007). Typically, expressive language undergoes rapid expansion of productive vocabulary between 18- and 24-months; according to the social-pragmatic theory, this acquisition of symbolic communication is supported by children's use of social cognition to decode adults' intended referents in social interactions (Tomasello 2001). Such social cognitive skills as well as the ability to combine appropriate vocabulary and nonverbal communication behaviors in efficient, well-formed expressive messages are critical to later effective pragmatic communication (Tursktra et al. 2017). By age 24 months, HR siblings with early and late manifestations of ASD show social-communication delays on the , as do many children who demonstrate BAP characteristics (Landa et al. 2007). Shared enjoyment, use of ge*ADOS*stures, and related social-communication behaviors at 24 months are the nascent forms of later reciprocal pragmatic communication. The question remains whether school-age pragmatic functioning, at 8–12 years of age, is predicted by early social-communication behaviors, such as 14-month joint attention and/or 24-month expressive language and *ADOS*-tested social-communication behaviors. Early strengths in joint attention and expressive language, in the absence of early ASD symptoms, could have a cascading effect that contributes to later pragmatic communication competency.

The current study examined pragmatic communication functioning in school-age siblings of children with ASD (HR) and low-risk (LR) controls, as well as early predictors of this functioning in the full sample. The following research hypotheses were addressed. First, between-group differences in pragmatic communication functioning at 8–12 years were examined, comparing three subgroups of HR siblings (based on 36-month diagnostic classifications) and LR controls. More specifically, 36-month HR siblings with typical development (HR-TD) were hypothesized to demonstrate significantly better school-age pragmatic communication functioning than HR siblings with 36-month BAP characteristics (HR-BAP), who are predicted to exhibit significantly better school-age pragmatic functioning than HR siblings with ASD (HR-ASD). No significant differences were hypothesized between HR-TD and LR groups. Second, correlations between school-age pragmatic communication, 14-month joint attention, and 24-month social-communication and expressive language were expected to be significant when conducted with the full sample, which provided a broad distribution of scores across all examined developmental domains. Finally, in regression models, 14-month frequency of joint attention initiation was expected to predict school-age pragmatic communication functioning in the full sample of LR and HR groups, inclusive of the three HR subgroups. Social-communication behaviors and expressive language at age 24 months were expected to predict school-age pragmatic communication functioning in the full sample. Finally, the combined set of 14- and 24-month predictors was expected to explain a significant amount of the variance in later pragmatic communication in the full sample. Note that predictor analyses were conducted with the full sample, providing a broad distribution of scores across multiple developmental domains including joint attention, social communication, and expressive language performance.

Methods

Participants

Participants were recruited for a prospective, longitudinal study of ASD. The Johns Hopkins Medicine Institutional Review Board approved this study. All families gave written informed consent for child participation; at school-age, children also provided verbal assent. Participants included 49 younger siblings of children with ASD, at high familial risk for ASD (HR) and 18 children at low familial risk for ASD (LR). Details about recruitment and proband ASD diagnosis are described in Landa et al. (2007). Exclusion criteria for HR siblings were: family's first language being other than English, birth weight < 1500 g, severe birth trauma, head injury, prenatal illicit drug or excessive alcohol exposure, and severe birth defects.

The *Pragmatic Rating Scale for School-Age Children* (*PRS-SA*; Landa, unpublished) adapted from the *PRS* (Landa et al. 1992), was scored for all participants based on behaviors observed during the *Autism Diagnostic Observation Schedule* (*ADOS*; Lord et al. 1999, 2012), administered between 8 and 12 years of age. All participants met criteria for at least an *ADOS* Module 3. Participant ages were: 39 8-year-olds, six 9-year-olds, 12 10-year-olds, seven 11-year-olds, and three 12-year-olds. All but three participants also received comprehensive developmental assessments at least twice between ages 14 and 36 months (14, 18, 24, 30, and/or 36 months), which included the *ADOS*, *Mullen Scales of Early Learning* (*MSEL*; Mullen 1995), and, through 24 months, the *Communication and Symbolic Behavior Scales Developmental Profile* (*CSBS DP;* Wetherby and Prizant 2002). Three children who joined the study at 48 months upon enrollment of an infant sibling were included to reduce between-group sex differences (two LR males, one HR female classified as typically developing—see below); none differed significantly from their group on *ADOS*-tested behaviors at enrollment. Criteria for classifying HR siblings into one of the three subgroups at age 36 months (n = 63; 48-months for n = 4) were as follows. ASD classification (HR-ASD, n = 15) required a clinical best estimate of ASD by an expert clinician; 14 of 15 children in this subgroup also met *ADOS* criteria for ASD or autism. Broad autism phenotype classification (HR-BAP, n = 19) required a clinical best estimate of no-ASD, along with one of the following: (1) elevated *ADOS* Communication + Social Interaction or Social-Affect algorithm score (within 3 points of ASD cut-off), (2) two or more *MSEL* subscale scores falling ≥ 1.5 standard deviations (SD) below the test mean, and/or (3) one or more *MSEL* subscale score(s) falling ≥ 2 SD below the test mean (following Ozonoff et al. 2014). The typically developing classification (HR-TD, n = 15) required not meeting criteria for ASD or BAP.

Of note, because some children did not complete all visits between 14 and 36 months, sample sizes for predictor analyses using data at 14 and 24 months were reduced to 51 children (LR = 9, HR-TD = 13, HR-BAP = 17, HR-ASD = 12) and 59 children (LR = 13, HR-TD = 13, HR-BAP = 18, HR-ASD = 15), respectively.

Measures

Administered at all visits between 14 and 36 months and at the school-age visit, the *Autism Diagnostic Observation Schedule* (*ADOS;* Lord et al. 1999, 2012) is a semi-structured, clinician-administered, play-based assessment that evaluates social-communication and repetitive and stereotyped behaviors and interests characteristic of ASD. The *ADOS* provides algorithm scores (e.g., *ADOS-Generic*: Communication + Social Interaction; *ADOS-2*: Social Affect), which are used to determine whether children meet the measure's thresholds for ASD or autism. These algorithm scores contributed to children's 36-month diagnostic classification. Children's 24-month *ADOS* Communication + Social Interaction algorithm scores served as a dependent variable.

The *Mullen Scales of Early Learning* (*MSEL*; Mullen 1995), a standardized developmental assessment, was administered at all visits between 14 and 36 months. *MSEL* Gross Motor, Fine Motor, Visual Reception, Receptive

Language, and Expressive Language subscale items generate subscale T scores ($M=50$; $SD=10$). Expressive Language T scores served as a dependent variable.

The *Communication and Symbolic Behavior Scales Developmental Profile* (*CSBS DP*) Behavior Sample (Wetherby and Prizant 2002) presents standardized communicative temptations followed by a brief play sample to assess children's social, communication, and symbolic skills. The number of times children initiated joint attention during the *CSBS DP* was a dependent variable. Initiating joint attention was defined as the coordinated use of communicative signals (≥ 2 of the following 3 behaviors: gaze, vocalizations, or gestures, such as pointing/showing) to comment on an object or action during the *CSBS DP* Behavior Sample. Inter-rater reliability for the frequency of joint attention initiation, calculated for 20% of the sample, was 80%.

The *Pragmatic Rating Scale* (Landa et al. 1992) was modified to create the *PRS-SA*, a measure of pragmatic communication in school-age children who use at least phrase-level language (see Klusek, Martin and Losh 2014 for *PRS-SA* data on school-aged children with ASD and Fragile X). The *PRS-SA*'s 22 items measured three categories of pragmatic communication: discourse management (e.g., topic maintenance, effective language formulation), presupposition (e.g., sufficiency of information, adequacy of clarifications), and nonverbal communication (e.g., gesture use, eye contact, intonation).

Behaviors represented by *PRS-SA* items were rated on a 4-point scale (0 = appropriate, 1 = mild variation, 2 = problematic/pervasive, 3 = significant problem). Like *ADOS* scoring, *PRS-SA* ratings of 2 and 3 were combined for reliability; 3's converted to 2's for scoring purposes. Of note, *PRS-SA* ratings were based on discourse samples from the *ADOS*; however, these ratings were distinct from *ADOS* codes, with separate operational definitions and rating schemes for each behavior. Also, *PRS-SA* raters were blinded to children's *ADOS* scores. *PRS-SA* total score was calculated by summing ratings for the 22 rated behaviors, and was used as the dependent variable representing school-age pragmatic communication functioning. Higher scores indicated greater pragmatic difficulties.

The *PRS-SA* was rated by consensus for 12 children (16%), to calibrate the raters. Interrater reliability of *PRS-SA* total scores (point of analysis for the current study), calculated for 13 of the remaining children (24%), was $ICC(2,1)=.977$. Intraclass correlations also were obtained for each of the 13 children by comparing each *PRS-SA* item-level rating (#1–22) made by two independent raters; *ICC*'s for each child then were averaged across all 13 children, yielding a mean $ICC(2,1)$ of .814. Reliability greater than or equal to .750 is considered excellent (Cicchetti 1994). Reliability fell below .750 for compared item-level ratings for four children: two were considered good (.60–.74); two were considered fair (.40–.59) (Cicchetti 1994). Samples with fair reliability generally had very limited rating variability, so discrepancies were "more unexpected." Consensus ratings were conducted for these four cases for use in data analyses.

Data Analysis

To account for non-normal distributions and differences in variability (Levene's test p's < .001 for comparisons of HR-TD, HR-BAP, and HR-ASD), nonparametric analyses were employed. Run in SPSS, Kruskal–Wallis and Mann–Whitney tests assessed group differences in descriptive variables and school-age pragmatic communication functioning. Effect size $r=z/\sqrt{n}$ is reported for Mann–Whitney tests, with .100, .300, and .500 representing small, medium, and large effects, respectively. Note: tests were run with and without the three participants who had not received developmental assessments until 48 months; results did not differ. Therefore, reported results are for the full sample.

Also run in SPSS, Spearman's rho examined correlations in the full sample. Each variable was checked for outliers; analyses re-run without outliers (*CSBS*: 1 LR, 1 HR-BAP; *MSEL*: 3 HR-ASD; *PRS-SA, ADOS*: none) yielded the same conclusions. Reported results include all participants.

Lastly, three nonparametric regression models run in STATA examined predictors of *PRS-SA* scores. These nonparametric models mimic ordinary least squares linear regression analyses, but do not assume a linear functional form. Note that STATA only provides z-values for nonparametric regressions to 2 decimal places. Model 1 examined the unique contribution of 14-month joint attention initiations in predicting later pragmatic communication, independent of 24-month social-communication and expressive language, the development of which are theoretically supported by joint attention (Mundy et al. 2009). Model 2 examined the unique contributions of 24-month variables, specifically *ADOS* social-communication behaviors and *MSEL* expressive language. Model 3 ascertained the combined predictive power of all three variables. Prior to conducting this third nonparametric regression model, a variable inflation factor (VIF) analysis was conducted within a parametric regression analysis to assess multicollinearity with the three variables; note that no nonparametric version of this analysis exists. VIF values were all < 1.6, and thus less than the two cut-off values (4 and 10) commonly used as indicators of problematic collinearity (Lavery et al. 2017). All regression analyses were conducted using the full sample to provide a broader distribution of scores, and avoid limited distributions that might be encountered in the small subgroup samples or the non-ASD sample. This larger distribution was expected to be more representative of the full performance continuum of joint attention, social-communication, expressive language, and pragmatic communication; thus, conclusions from analyses cannot be said to represent relations within the LR group or HR subgroups.

Results

Descriptive Statistics

Demographic variables and standardized test scores for the LR group and HR diagnostic classification subgroups are presented in Table 1. No significant between-group differences were detected in age, race, ethnicity, or socio-economic status (Hollingshead 1975). There was a significant sex difference across groups ($X^2 = 12.210$, $p = .007$) and in *PRS-SA* scores ($Z = -3.179$, $p = .001$, $r = .388$). Follow-up tests revealed no significant sex-based differences in *PRS-SA* scores in the LR controls ($Z = -.817$, $p = .425$, $r = .193$) or HR-TD ($Z = -.355$, $p = .776$, $r = .092$). However, *PRS-SA* scores were significantly higher (more atypical) for HR-BAP males ($M = 21.000$, $SD = 6.074$) than females ($M = 12.222$, $SD = 6.723$; $Z = -2.412$, $p = .013$, $r = .553$). There was only one female in the HR-ASD subgroup; her *PRS-SA* score of 34 approximated the male mean of 32.571. Note: we did not control for sex in subsequent analyses due to nonequivalent sex ratios across groups/diagnostic classifications.

Group Differences in Pragmatic Language

Table 1 summarizes means and standard deviations for *PRS-SA* scores in each group/diagnostic classification. Figure 1 illustrates between-group differences in *PRS-SA* scores. Significant between-group differences were found in *PRS-SA* scores, $H(3) = 38.005$, $p < .001$. Based on visual inspection, distributions of school-age pragmatic communication functioning were similar for LR and HR-TD groups, but were not similar for HR-TD, HR-BAP, or HR-ASD subgroups. Follow-up tests revealed no significant difference in *PRS-SA* scores of LR controls and HR-TD ($U = 112.500$, $Z = -.815$, $p = .421$, $r = .142$). *PRS-SA* scores were significantly lower (better) in the HR-TD than HR-BAP ($U = 59.000$,

Table 1 Demographics and standardized test results

	Groups				Significance
	LR controls n = 18	HR-TD n = 15	HR-BAP n = 19	HR-ASD n = 15	
Demographics					
Sex (M:F)	7:11	6:9	10:9	14:1	$X^2(3) = 12.210$, $p = .007$
Race (Cauc:oth)	18:0	13:2	18:1	13:2	$X^2(3) = 3.082$, $p = .379$
Ethnicity (H:N:UK)	2:9:7	0:8:7	0:15:4	2:4:9	$X^2(6) = 11.880$, $p = .065$
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Significance
Hollingshead	58.028 (9.097)	57.467 (9.224)	55.947 (9.932)	57.167 (7.729)	$F(3,63) = .173$, $p = .915$
School-age visit					
Age in months	114.024 (18.115)	118.332 (19.506)	104.955 (13.237)	115.762 (13.572)	$F(3,63) = 2.255$, $p = .091$
ADOS CSS	1.467 (0.743)	2.083 (1.443)	2.778 (2.157)	8.067 (2.052)	LR,HR-TD,HR-BAP < HR-ASD
PRS-SA	8.889 (7.435)	9.533 (4.533)	16.842 (7.669)	32.667 (7.118)	LR/HR-TD, ns HR-TD < HR-BAP < HR-ASD
14-month visit	n = 9 Mean (SD)	n = 13 Mean (SD)	n = 17 Mean (SD)	n = 12 Mean (SD)	Significance
CSBS DP IJA	5.444 (3.941)	4.077 (2.629)	4.294 (3.077)	1.750 (2.379)	LR > HR-ASD
24-month visit	n = 13 Mean (SD)	n = 13 Mean (SD)	n = 18 Mean (SD)	n = 15 Mean (SD)	Significance
ADOS CoSo	4.846 (3.648)	3.385 (2.434)	5.444 (4.076)	10.267 (4.713)	LR,HR-TD,HR-BAP < HR-ASD
MSEL EL*	62.500 (9.155)	54.077 (8.976)	51.222 (8.674)	36.467 (14.861)	LR,HR-TD,HR-BAP > HR-ASD LR > HR-BAP

LR low-risk, *HR-TD* high-risk typical development, *HR-BAP* high-risk broad autism phenotype, *HR-ASD* high-risk autism spectrum disorder; *M:F* male:female; *Cauc:Oth* Caucasian:other; *H:N:UK* Hispanic:Non-Hispanic:unknown; *ADOS* Autism Diagnostic Observation Schedule, *CSS* Calibrated Severity Score; *PRS-SA* Pragmatic Rating Scale, School-Age total score; *CSBS DP IJA* Communication and Symbolic Behavior Scales Developmental Profile Initiation of Joint Attention frequency; *CoSo ADOS* Communication + Social Interaction algorithm score; *MSEL EL* Mullen Scales of Early Learning Expressive Language T score; *ns* not significant

*LR:n = 14

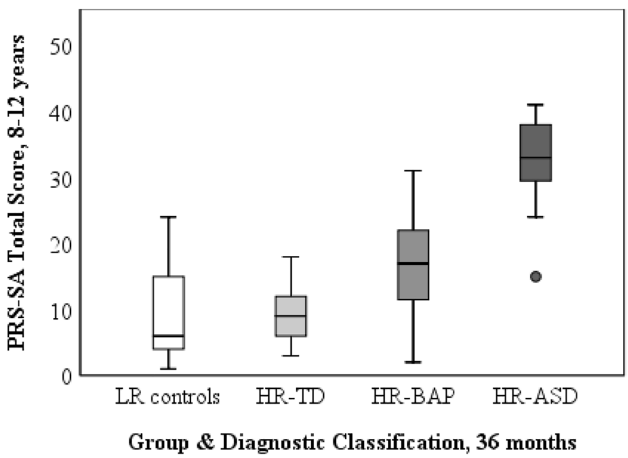

Fig. 1 Boxplot of *PRS-SA* total scores at age 8 to 12 years for group and diagnostic classifications made at 36 months

$Z = -2.899$, $p = .003$, $r = .497$) and HR-ASD subgroups ($U = 2.000$, $Z = -4.588$, $p < .001$, $r = .838$). *PRS-SA* scores were significantly lower for the HR-BAP than HR-ASD subgroup ($U = 18.500$, $Z = -4.305$, $p < .001$, $r = .738$), meaning that the HR-ASD group displayed more atypical pragmatic behavior than the HR-BAP group.

Predictors of Pragmatic Language

Table 2 summarizes the correlations between *PRS-SA* scores and predictors across the full sample (all four groups/diagnostic classifications; top) as well as post-hoc correlations in the non-ASD sample (LR, HR-TD, and HR-BAP; bottom). In the full sample, *PRS-SA* scores were strongly correlated with frequency of joint attention initiation at age 14 months ($r = -.420$, $p = .002$, $r^2 = .177$) and 24-month *MSEL* Expressive Language T scores ($r = -.506$, $p < .001$, $r^2 = .257$), and moderately correlated with 24-month *ADOS* Communication + Social Interaction algorithm scores ($r = .379$, $p = .003$, $r^2 = .144$) (see Fig. 2). To determine whether current data replicated correlations from Gillespie-Lynch et al. (2015), post-hoc analyses examined Spearman's rho correlations in the non-ASD sample (combined LR, HR-TD, HR-BAP groups); this secondary analysis was considered preliminary due to the reduced variability and sample size resulting from removal of the ASD group. In the non-ASD sample, *PRS-SA* scores were not correlated with frequency of joint attention initiation at age 14 months ($r = -.197$, $p = .228$, $r^2 = .039$) or 24-month *MSEL* Expressive Language T scores ($r = -.276$, $p = .067$, $r^2 = .076$) or *ADOS* Communication + Social Interaction algorithm scores ($r = .018$, $p = .908$, $r^2 < .001$). Note, when one outlier was removed, the correlation with *MSEL* Expressive Language T scores in the non-ASD sample no longer trended toward significance ($r = -.213$, $p = .164$, $r^2 = .046$).

Table 3 summarizes the three non-parametric regression models examining 14-month and 24-month predictors of school-age pragmatic communication in the full sample. Regression models in the non-ASD sample were not warranted, as correlations were nonsignificant. In Model 1, frequency of joint attention initiation during the *CSBS DP* at 14 months accounted for a significant amount of the variance in school-age pragmatic functioning in the full sample ($R^2 = .239$). Frequency of joint attention initiation had

Table 2 Correlation matrix for PRS-SA scores, 14-month joint attention initiations, and 24-month social-communication and expressive language scores in the full and no-ASD samples

Full sample (LR, HR-TD, HR-BAP, & HR-ASD)	*CSBS DP* joint attention initiations, 14 m		*ADOS* CoSo, 24 m		*MSEL* EL, 24 m	
	Spearman's rho	*p*	Spearman's rho	*p*	Spearman's rho	*p*
1. *PRS-SA*, 8-12y	−.420	.002	.379	.003	−.506	<.001
2. *CSBS DP* joint attention initiations, 14 m			−.374	.009	.447	.001
3. *ADOS* CoSo, 24 m					−.360	.005
Non-ASD sample (LR, HR-TD, & HR-BAP)	**Spearman's rho**	***p***	**Spearman's rho**	***p***	**Spearman's rho**	***p***
1. *PRS-SA*, 8-12y	−.197	.228	.018	.908	−.276	.067[a]
2. *CSBS DP* joint attention initiations, 14 m			−.112	.517	.163	.336
3. *ADOS* CoSo, 24 m					.008	.958

PRS-SA Pragmatic Rating Scale, School-Age total score; *CSBS DP* Communication and Symbolic Behavior Scales Developmental Profile; *ADOS CoSo* Autism Diagnostic Observation Schedule Communication + Social Interaction algorithm score; *MSEL EL* Mullen Scales of Early Learning Expressive Language T score; *y* years; *m* months

[a]With one outlier removed, $p = .164$

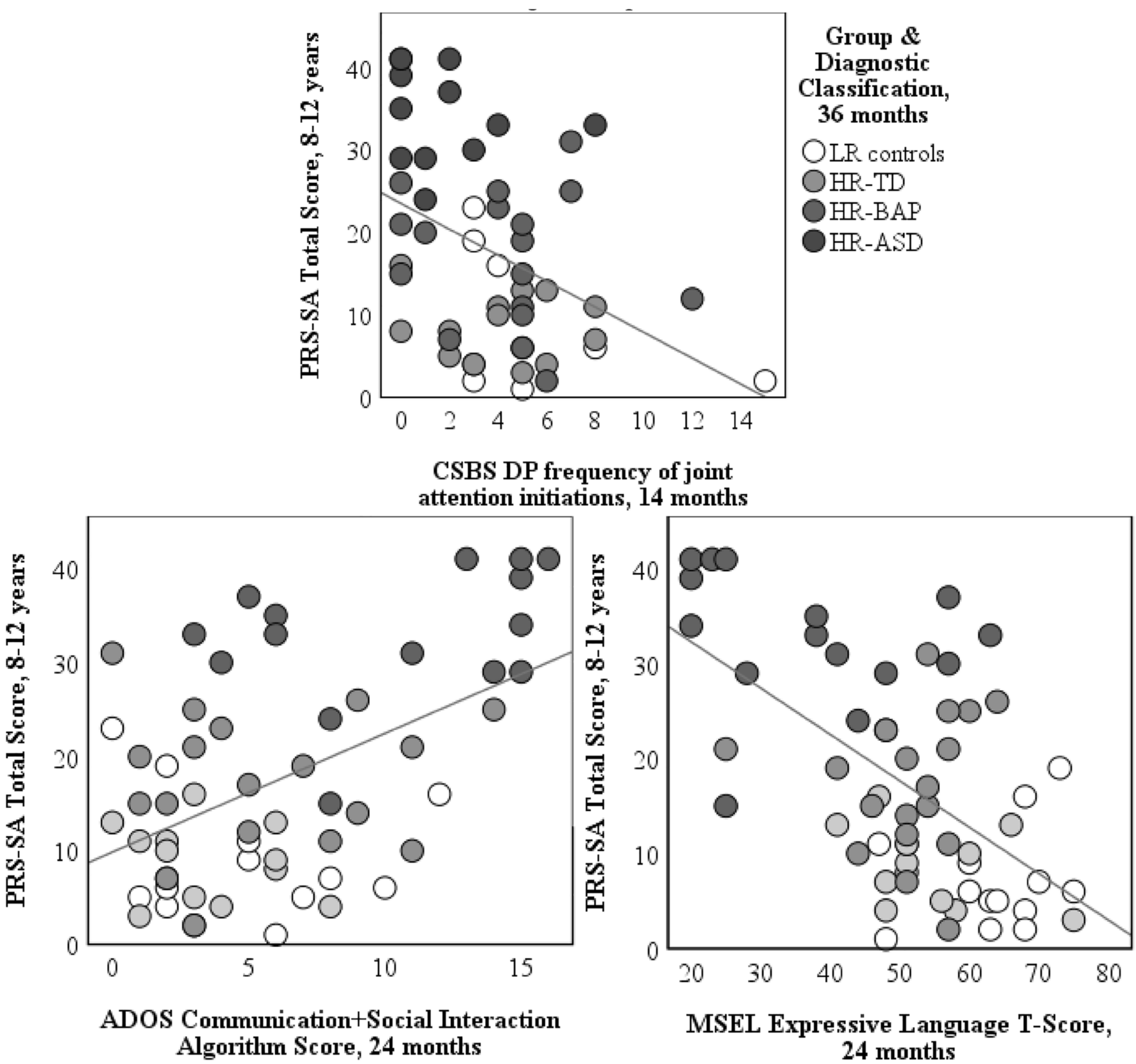

Fig. 2 Paneled scatterplots of *PRS-SA* total score at age 8 to 12 years by 14-month *CSBS DP* frequency of joint attention (top) and 24-month *ADOS* Communication + Social Interaction Algorithm score (bottom left) and *MSEL* Expressive Language T score (bottom right)

a unique inverse relation with *PRS-SA* scores ($b = -1.890$, $SE = 0.685$, $z = -2.76$, $p = .006$). That is, for each occurrence of joint attention initiation during the *CSBS DP, PRS-SA* scores decreased (became less atypical) by 1.890 points.

In Model 2, *MSEL* Expressive Language T scores and *ADOS* Communication + Social Interaction algorithm scores at 24 months accounted for a significant amount of variance in school-age pragmatic functioning in the total sample ($R^2 = .386$). *MSEL* Expressive Language T scores had a unique inverse effect for *PRS-SA* scores ($b = -.356$, $SE = .096$, $z = -3.71$, $p < .0001$); for each one-point increase in *MSEL* Expressive Language T scores, *PRS-SA* scores decreased (showed less atypicality) by .356 points. *ADOS* Communication + Social Interaction algorithm scores had a unique positive effect on *PRS-SA* scores, ($b = .636$, $SE = .259$, $z = 2.45$, $p = .014$); for each one-point increase in this predictor, *PRS-SA* scores increased .636 points.

In Model 3, 14-month frequency of joint attention initiation and 24-month *MSEL* Expressive Language T scores and *ADOS* Communication + Social Interaction algorithm scores accounted for a significant amount of variance in school-age pragmatic functioning in the total sample ($R^2 = .661$). However, none of the three variables had a unique effect on *PRS-SA* total scores (14-month *CSBS DP* joint attention initiation: $b = -.268$, $SE = 1.114$, $z - .24$, $p = .810$; 24-month *ADOS* Communication + Social Interaction algorithm scores: $b = .659$, $SE = .944$, $z = .70$, $p = .486$; 24-month *MSEL* Expressive Language T scores: $b - .300$, $SE = .303$, $z - .99$, $p = .321$).

Table 3 Nonparametric regression models, with 14-month predictors, 24-month predictors, and combined 14- and 24-month predictors of PRS-SA total scores

	Standard regression			
	R^2	*B* (SE)	*z*	*p*
Model 1: 14-month predictor				
PRS-SA	.239			
CSBS DP joint attention initiations		− 1.890 (.685)	− 2.76	.006
Model 2: 24-month predictors				
PRS-SA	.386			
ADOS CoSo		.636 (.259)	2.45	.014
MSEL EL		− .356 (.096)	− 3.71	< .0001
Model 3: 14- and 24-month predictors				
PRS-SA	.661			
CSBS DP joint attention initiations		− .268 (1.114)	− .24	.810
ADOS CoSo		.659 (.944)	.70	.486
MSEL EL		− .300 (.303)	− .99	.321

PRS-SA Pragmatic Rating Scale, School-Age total score; *CSBS DP* Communication and Symbolic Behavior Scales Developmental Profile; *ADOS CoSo* Autism Diagnostic Observation Schedule Communication + Social Interaction algorithm score; *MSEL EL* Mullen Scales of Early Learning Expressive Language T score

Discussion

The current study used a deep phenotyping approach to examine school-age pragmatic communication behavior in HR siblings of children with ASD and LR controls and very early predictors thereof. A clinician-rated behavioral observation measure was used to assess pragmatic communication functioning in school-age HR siblings and LR controls. We found that HR siblings who demonstrated ASD or BAP characteristics at 36 months exhibited deficits in pragmatic language and nonverbal communication at school-age relative to HR siblings with typical development, whose performance did not differ from LR controls. As expected, pragmatic communication was significantly correlated with 14-month joint attention and 24-month expressive language and social-communication in the full sample; post-hoc correlations in the non-ASD (LR, HR-TD, and HR-BAP) group were not significant. This result suggests a full continuum (scores ranging from neurotypical to subclinical differences to clinically significant differences) of pragmatic communication, joint attention, expressive language, and social-communication performance may be necessary to detect these relations, especially in relatively small samples. Further, in the full sample of HR siblings and LR controls, including children with ASD, frequency of joint attention initiation at 14 months predicted school-age pragmatic communication functioning, as did 24-month expressive language and autism-related social-communication behaviors in a separate, age-specific model. The full model with all three predictors explained a significant amount of the variance in school-age pragmatic communication scores; however, none of the early predictors had a unique effect on that outcome when 14- and 24-month variables were examined together. To our knowledge, this is the first report that school-age pragmatic functioning is distinguished by 36-month diagnostic classifications in HR siblings, suggesting continuity in the relative degree of typicality of social-communication functioning from early to middle childhood. This study also identified the earliest developmental predictor to date of school-age pragmatic functioning in the full sample of HR siblings and LR controls, including children with ASD: 14-month joint attention initiations.

Overall, the *PRS-SA* yielded a wide score continuum across children at low familial risk for ASD and subgroups of children at high familial risk for ASD who, together, displayed a broad continuum of ASD symptomatology ranging from no symptoms to clinically significant expression of symptoms. Thus, the *PRS-SA* showed sensitivity to a range of pragmatic communication functioning in this sample that displayed diverse pragmatic behavior. As hypothesized, no significant differences existed between the HR-TD and LR groups, and the HR-ASD subgroup demonstrated significantly higher *PRS-SA* scores, signaling more impaired pragmatic communication abilities, than other HR subgroups. Significantly higher school-age *PRS-SA* scores in the HR-BAP than HR-TD subgroup suggested poorer pragmatic communication functioning in children identified as having language or other developmental delays with or without subclinical ASD-related social-communication difficulties at 36 months. This finding is consistent with other investigators' reports of pragmatic language differences (Ben-Yizhak et al. 2011; Miller et al. 2015) and other developmental challenges (Charman et al. 2017; Georgiades et al. 2013; Landa et al. 2007; Messinger et al. 2013; Ozonoff et al. 2014) in a subset of non-ASD HR siblings. Our use of a direct observational measure of pragmatic functioning rather than a parent-report measure may explain why the results presented herein differed from prior studies showing no differences in pragmatic functioning of HR siblings compared with controls (Bishop et al. 2006; Gillespie-Lynch et al. 2015). Another possible explanation for this discrepancy in findings may relate to our separation of non-ASD HR siblings into TD and BAP subgroups based on their 36-month phenotypic presentation, while many other investigators examined non-ASD HR siblings as a single group. Our results suggest BAP characteristics exhibited as early as 36 months persist into school-age, with pragmatic functioning ranging across a score continuum that overlaps with ASD and TD subgroup scores and representing pragmatic communication difficulties that may or may not be impairing.

Another important finding of the present study was that HR siblings who met 36-month BAP criteria had differential pragmatic communication outcomes based on sex, with females' *PRS-SA* scores being significantly lower (less impaired) than males'. Although preliminary, these results suggest pragmatic communication resiliency may be greater in females, even if they meet BAP criteria early in development. This finding is consistent with Virkud et al. (2009) report of a bimodal distribution of quantitative autistic traits in female siblings within multiplex families, whereas male siblings demonstrated a unimodal distribution lacking a clear cutoff differentiating those who did or did not have ASD. In contrast, neither Klusek, Losh and Martin (2014) nor Landa et al. (1992) identified sex differences in the pragmatic functioning of parents of children with ASD. More research is needed to understand why sex differences are detectable during childhood in HR siblings but not in parents of children with ASD.

In the full sample, *PRS-SA* scores were strongly correlated with 14-month joint attention initiations and 24-month expressive language, and moderately correlated with 24-month ASD-related social-communication behaviors. Consistent with Gillespie-Lynch et al. (2015), the association with early joint attention initiation was nonsignificant when assessed only in the children who did not have ASD (LR, HR-TD, and HR-BAP; *CSBS*: n = 39), with similarly nonsignificant results for 24-month expressive language and social-communication behaviors (*ADOS*: n = 43, *MSEL*: n = 44). Several possible explanations might account for the difference in results between the full sample (including children with ASD) and non-ASD sample. First, the limited variability and score range observed in the non-ASD group may have been insufficient to reveal the theoretically predicted relation between early joint attention, social-communication behaviors, and expressive language and later pragmatic communication, particularly given the small sample size (see Fig. 1 to visualize variability in the non-ASD groups). Both Gillespie-Lynch et al. (2015) and the present results were found with small samples (n = 20 and n = 39, respectively), where restricted distributions and minimal variability can limit the ability to detect relations that might be apparent in larger and/or more variable samples. Second, relations in the full sample may have been driven solely by the ASD group. While possible, the data revealed a continuous distribution of *PRS-SA* scores across groups, with no clear gaps between typical development, subclinical features, and clinically significant differences in pragmatic communication, and no outliers identified in the full sample. Further, no children with ASD were considered outliers in frequency of joint attention initiation or social-communication behaviors. Three children with ASD were considered outliers in terms of 24-month expressive language; however, results did not change when these children's scores were excluded. Thus, children with ASD appeared to be part of the same distribution as children from the other three subgroups (as expected given the neurobiological literature, c.f. Constantino 2011), suggesting their data added more variability and increased the sample size to a sufficient degree to detect relations. Finally, it is possible that different mechanisms may underlie pragmatic communication development in different subgroups. Larger sample sizes in each group would be required to test this hypothesis.

In age-specific regression models using the study's full sample, frequency of joint attention initiation at 14 months uniquely predicted school-age pragmatic functioning, as did 24-month social-communication and expressive language scores. Current findings provide the first evidence of a significant relation between early frequency of joint attention initiation and school-age pragmatic communication functioning. As described previously, this result differs from Gillespie-Lynch et al. (2015), in which 12-month joint attention initiations were not associated with school-age pragmatic language. This discrepant outcome could be due to differences in age (12- vs. 14-month, with more frequent initiation of joint attention expected at age 14 months) or measurement of joint attention (e.g., Gillespie-Lynch et al.'s inclusion of low level gaze alternation vs. our requirement of higher level initiation of joint attention behavior signaled by pairing two of three communicative behaviors [gaze, vocalizations, gestures]), differences in school-age pragmatic communication measures (parent report vs. behavioral observation), or differences in grouping for analysis (separation of ASD and non-ASD vs. combined HR-TD, HR-BAP, HR-ASD, and LR controls), leading to differences in sample size and variability of predictor and outcome. As noted previously, sample sizes in both studies were quite small, with distributions and variabilities that might not adequately represent the full spectrum of pragmatic communication or joint attention initiation that exists in the general population. Thus, findings should be interpreted with caution. Also, it should be noted that all but one of the 13 children in our sample (2 HR-TD, 4 HR-BAP, 7 HR-ASD) who demonstrated one or no joint attention initiations at 14 months received a *PRS-SA* total score of 15 or higher, where 15 was the lowest total score in the HR-ASD subgroup. Thus, a slow start to the development of endogenously-motivated triadic engagement, which is well established in typical development by 14 months, may be a risk indicator for school-age pragmatic communication differences, signaling that preemptive intervention may be beneficial. Without intervention, delays in developing joint attention initiations could contribute to a cascade of developmental events and types/frequencies of social engagement opportunities that may accumulate and manifest in altered pragmatic communication behavior at school-age.

In addition, 24-month social-communication behaviors and expressive language were associated with later pragmatic communication functioning. *ADOS* social-communication scores at this age capture behaviors reflecting effective social-communication, such as directed vocalizations, gestures, pointing, showing, and joint attention responses and initiations. According to the parallel and distributed processing model, joint attention involves the integrated processing of one's own and others' visual attention (Mundy et al. 2009). Other behaviors included in the *ADOS* Communication + Social Interaction algorithm relate to children's ability to direct their attention and share their emotions with others, which similarly require the coordinated processing of their own and others' attention. Extending forward in development, integrated processing of self- and other-attention evolves into the covert coordination of mental representations required for developing symbolic language, social cognition/presupposition, and later "rapid-fire" pragmatic communication exchanges (Mundy et al. 2009). *MSEL* expressive language scores at 24 months largely reflect productive vocabulary, with contributions from combinations of gestures + words/vocalizations and use of appropriate intonation. The social-pragmatic theory of language acquisition (Tomasello 2001) suggests that both early symbolic communication and later pragmatic communication rely on social cognition. Specifically, language acquisition is considered a "by-product of social interaction" (p. 135), reflecting children's improving ability to interpret an adult partner's intentions, based on the assumption that the words communicating those intentions are relevant to the ongoing social interaction (Tomasello 2001). The strong predictive relation between 24-month expressive language and social-communication behaviors and later pragmatic functioning suggests the early social cognitive abilities that both support and are supported by language acquisition and early social-communication behaviors also may provide a foundation for more advanced presuppositional behaviors. Further, directing communication toward a partner, including the integration of appropriate gestures, intonation, and vocabulary use, during toddlerhood contributes to children's growing capacity to signal their intentions to others and infer others' intentions through differentiated and more precise communication. This clear communication of intentions is essential to successful advanced (school-age) pragmatic communication competence (Sperber and Wilson 1995; Turkstra et al. 2017).

Finally, when all three predictors (across ages 14 and 24 months) were entered into a single regression model, they combined to explain 66% of the variance in school-age pragmatic communication; yet no predictor had a unique impact on later pragmatic functioning. While variables did not meet the threshold for multicollinearity, the high correlations among predictor variables may have contributed to the lack of any variable uniquely predicting school-age pragmatic communication. Another possibility is that the sample size did not provide enough degrees of freedom to detect unique contributions in a regression model with three predictors. Overall, results of this model suggest that early joint attention initiations, social-communication behaviors, and expressive language may coalesce to form a set of early-developing risk factors that are readily observable, but that may overlap to a sufficient degree that their unique contributions cannot be detected when examined together. These aspects of development should be monitored closely in the first 2 years of life for all children, and especially in children with a family history of ASD or other ASD risk factors (for video tutorial of early ASD signs, Kennedy Krieger Institute 2013). Early disruption in the development of these skills, even in toddlers with ASD, is malleable with exposure to evidence-based intervention targeting these skills (Landa et al. 2011).

Pragmatic communication abilities associated with elevated *PRS-SA* scores may or may not be considered an impairment. Based on the World Health Organization (2011), pragmatic communication deficits would only be considered an impairment if they limit an individual's activities and participation, which are influenced by environmental factors (e.g., supports/barriers, relationships) and personal factors (e.g., culture, temperament). When pragmatic communication differences do lead to impairments, consequences can be broad-reaching, affecting a child's social interactions across everyday activities, internalizing and externalizing characteristics, and others' perception of the child's social competence and acceptability (Fujiki et al. 2001; Miller et al. 2015; Prutting and Kirchner 1987). Variation from typical pragmatic communication may not be impairing for some children at school-age, but could interfere with social acceptance and functioning later in life. More longitudinal research is needed to address this possibility.

Finally, this study's use of a dedicated clinician-rated behavioral observation tool to assess pragmatic communication functioning based on ecologically valid, semi-structured interactions represented an important contribution to the literature, offering distinct advantages over previous studies' use of standardized assessments (Drumm et al. 2015), analyses based on item-level *ADOS* codes (Ben-Yizhak et al. 2011), and parent rating scales (Bishop et al. 2006; Gillespie-Lynch et al. 2015). The *PRS-SA*'s measurement of contextualized pragmatic language and nonverbal communication is more likely to yield ecologically valid results than standardized assessments, which collect contrived communication samples that may not be representative of functional pragmatic communication use in everyday contexts (Roth and Spekman 1984b). Further, the *PRS-SA* was sensitive to pragmatic communication differences in HR siblings that previous studies using parent-report measures did not detect (Bishop et al. 2006; Gillespie-Lynch et al. 2015), possibly due to parents overestimating siblings' skills based on a skewed frame of reference (i.e., an older sibling with ASD; Drumm et al. 2015; Hess and Landa 2012).

The utility of *PRS-SA* scores to measure a dimensional and distributional construct like pragmatic communication is impressive. Current findings of expected differences between children with typical development (HR-TD and LR), BAP, and ASD classifications at 36 months provide strong initial support for the construct validity of *PRS-SA* scores. If additional psychometric evidence replicates these robust findings, the *PRS-SA* may offer a uniquely useful means of measuring the full range of pragmatic functioning and identifying difficulties that interfere with everyday social success, even if a child does not meet ASD criteria.

Limitations and Future Directions

Small sample sizes within each group/diagnostic classification limit the generalizability of results. Specifically, HR-BAP sex differences require replication with a larger sample; HR-ASD females as well as females with ASD more broadly should be added to future samples, to determine whether *PRS-SA* scores vary based on sex in this population. Larger sample sizes also would permit examination of the association between higher levels of impairment and specific pragmatic communication deficits (e.g., discourse management vs. presupposition), and/or between specific early predictors and pragmatic communication deficits (e.g., joint attention and nonverbal communication). Furthermore, small samples prevented examination of predictors within each group/diagnostic classification, which could reveal group-specific predictors. While 14-month joint attention initiation frequency and 24-month expressive language and ASD-related social-communication behaviors predicted school-age pragmatic functioning for the combined groups/diagnostic classifications, unique predictors may account for pragmatic communication outcomes in each group and HR diagnostic classification. Color-coding of scatterplots in Fig. 2 shows different groups/diagnostic classifications, allowing readers to interpret where associations may lie for the current sample. Expansion of sample sizes would permit a clearer understanding of within-group variability and would permit independent regression analyses for each HR sibling subgroup and LR controls. Further, a larger sample size would enable testing of developmental mediation models, such as whether 14-month joint attention initiation frequency predicts school-age pragmatic communication functioning via 24-month expressive language and/or social-communication behaviors.

The current study provided preliminary support for using the *Pragmatic Rating Scale—School Age* as a reliable and valid clinician-rated direct observational measure of pragmatic communication. Additional psychometric data should be gathered in a larger sample. Specifically, a principal components analysis should test the three proposed categories of pragmatic communication behavior (discourse management, presupposition, nonverbal communication) in a sample of 150–200 participants (statistics.laerd.com 2018). If support is found for proposed categories of pragmatic communication behavior and/or reliability of isolated items (e.g., gestures), predictors for the proposed categories and/or item-level behaviors could be examined (e.g., are joint attention initiations related to later pragmatic language domains such as presupposition, or only to nonverbal communication and/or gesture use?). If additional reliability and validity evidence supports *PRS-SA* score use overall, this measure could provide an ecologically valid measure for qualifying children for speech-language intervention services, identifying strengths and weaknesses, examining functional connectivity as it relates to pragmatic language functioning, longitudinally tracking children's pragmatic functioning, and determining the stability of strengths and weaknesses over time, which may facilitate the creation of targeted interventions.

Conclusion

The present study provided evidence of pragmatic communication variations from the norm in subsets of school-age HR siblings, specifically those with 36-month ASD or BAP classifications. Further, results provide the best current evidence of specific early predictors of later pragmatic functioning across LR children and HR children classified at 36-months as TD, BAP, and ASD. If early joint attention, social-communication, and expressive language provide a foundation for school-age (and perhaps lifelong) pragmatic communication, early intervention targeting these abilities will be critical for promoting successful outcomes for HR siblings.

Acknowledgements This research was funded by Grant MH59630 from the National Institute of Mental Health (R.J.L., principal investigator). We also thank the many research clinicians who collected this study's data, and the superb staff who provided administrative support, videotaped assessment sessions, and processed data. We also thank Luther Kalb for his expert consultation in nonparametric statistical analysis. Most importantly, we thank the participants and their families, who made this study possible.

Author Contributions RJL conceived of the study and obtained the funding. KJG and RJL planned the analyses. KJG and EAU coded the PRS-SA data, with input from RJL. KJG conducted the analyses. KJG and RJL interpreted the results. KJG took the lead in writing the manuscript. RJL provided critical feedback and helped shape the manuscript. EAU read and agreed with the contents of the manuscript.

Compliance with Ethical Standards

Conflict of interest No conflicts of interest are declared.

Ethical Approval All procedures performed in this study 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. All families gave written informed consent for child participation; at school-age, children provided verbal assent.

References

American Psychiatric Association (APA). (2013). *Diagnostic and statistical manual of mental disorders* (*DSM-5*®). Washington, DC: American Psychiatric Pub.

Ben-Yizhak, N., Yirmiya, N., Seidman, I., Alon, R., Lord, C., & Sigman, M. (2011). Pragmatic language and school related linguistic abilities in siblings of children with autism. *Journal of Autism and Developmental Disorders, 41*(6), 750–760.

Bishop, D. V. M. (2003). *Children's Communication Checklist (CCC-2)*. London: Pearson.

Bishop, D. V. M., Mayberry, M., Wong, D., Maley, A., & Hallmayer, J. (2006). Characteristics of the broader phenotype in autism: A study of siblings using the Children's Communication Checklist-2. *American Journal of Medical Genetics Part B (Neuropsychiatric Genetics), 141B*, 117–122. https://doi.org/10.1002/ajmg.b.30267.

Charman, T., Young, G. S., Brian, J., Carter, A., Carver, L. J., Chawarska, K., et al. (2017). Non-ASD outcomes at 36 months in siblings at familial risk for autism spectrum disorder (ASD): A baby siblings research consortium (BSRC) study. *Autism Research, 10*(1), 169–178.

Cicchetti, D. V. (1994). Guidelines, criteria, and rules of thumb for evaluating normed and standardized assessment instruments in psychology. *Psychological Assessment, 6*(4), 284–290. https://doi.org/10.1037/1040-3590.6.4.284.

Constantino, J. (2011). The quantitative nature of autistic social impairment. *Pediatric Research, 69*(5 PT 2), 55R–62R. https://doi.org/10.1203/PDR.0b013e318212.

Donaldson, A. L., & Olswang, L. B. (2009). Investigating requests for information in children with autism spectrum disorders: Static versus dynamic assessment. *Advances in Speech-Language Pathology, 9*(4), 297–311. https://doi.org/10.1080/14417040701413720.

Drumm, E., & Brian, J. (2013). The developing language abilities and increased risks of 'unaffected' siblings of children with autism spectrum disorder. *Neuropsychiatry, 3*(5), 513–524. https://doi.org/10.2217/NPY.13.65.

Drumm, E., Bryson, S., Zwaigenbaum, L., & Brian, J. (2015). Language-related abilities in 'unaffected' school-aged siblings of children with ASD. *Research in Autism Spectrum Disorders, 18*, 83–96. https://doi.org/10.1016/j.rasd.2015.07.007.

Folstein, S., & Rutter, M. (1977). Infantile autism: A genetic study of 21 twin pairs. *Journal of Child Psychology and Psychiatry, 18*(4), 297–321. https://doi.org/10.1111/j.1469-7610.1977.tb00443.x.

Fujiki, M., Brinton, B., Isaacson, T., & Summers, C. (2001). Social behaviors of children with language impairment on the playground: A pilot study. *Language, Speech and Hearing Services in Schools, 32*, 101–113.

Georgiades, S., Szatmari, P., Zwaigenbaum, L., Bryson, S., Brian, J., Roberts, W., et al. (2013). A prospective study of autistic-like traits in unaffected siblings of probands with autism spectrum disorder. *JAMA Psychiatry, 70*(1), 42–48. https://doi.org/10.1001/2013.jamapsychiatry.1.

Gessaroli, E., Santelli, E., di Pellegrino, G., & Frassinetti, F. (2013). Personal space regulation in childhood autism spectrum disorders. *PLoS ONE, 8*(9), e74959. https://doi.org/10.1371/journal.pone.0074959.

Gillespie-Lynch, K., Khalulyan, A., del Rosario, M., McCarthy, B., Gomez, L., Sigman, M., et al. (2015). Is early joint attention associated with school-age pragmatic language? *Autism, 19*(2), 168–177.

Grice, H. P. (1975). *Logic and conversation*. Cambridge: Harvard University Press.

Hale, C. M., & Tager-Flusberg, H. (2005). Social communication in children with autism: The relationship between theory of mind and discourse development. *Autism, 9*(2), 157–178. https://doi.org/10.1177/1362361305051395.

Happé, F. G. (1994). An advanced test of theory of mind: Understanding of story characters' thought and feelings by able autistic, mentally handicapped, and normal children and adults. *Journal of Autism and Developmental Disorders, 24*(2), 129–154.

Hess, C. R., & Landa, R. J. (2012). Predictive and concurrent validity of parent concern about young children at risk for autism. *Journal of Autism and Developmental Disorders, 42*(4), 575–584.

Hollingshead, A. B. (1975). *Four factor index of social status*. New Haven: Department of Sociology, Yale University.

Hurley, R. S., Losh, M., Parlier, M., Reznick, J. S., & Piven, J. (2007). The broad autism phenotype questionnaire. *Journal of Autism and Developmental Disorders, 37*(9), 1679–1690.

Kennedy Krieger Institute. (2013). *Early signs of autism video tutorial—Kennedy Krieger Institute* [Video file]. Retrieved from https://www.youtube.com/watch?v=YtvP5A5OHpU.

Klusek, J., Losh, M., & Martin, G. E. (2014). Sex differences and within-family associations in the broad autism phenotype. *Autism, 18*(2), 106–116.

Klusek, J., Martin, G. E., & Losh, M. (2014). A comparison of pragmatic language in boys with autism and fragile X syndrome. *Journal of Speech, Language, and Hearing Research, 57*(5), 1692–1707.

Lam, Y. G., & Yeung, S. S. S. (2012). Towards a convergent account of pragmatic language deficits in high-functioning autism: Depicting the phenotype using the Pragmatic Rating Scale. *Research in Autism Spectrum Disorders, 6*(2), 792–797. https://doi.org/10.1016/j.rasd.2011.08.004.

Landa, R., Folstein, S. E., & Isaacs, C. (1991). Spontaneous narrative-discourse performance of parents of autistic individuals. *Journal of Speech, Language, and Hearing Research, 34*(6), 1339–1345. https://doi.org/10.1044/jshr.3406.1339.

Landa, R., Piven, J., Wzorek, M. M., Gayle, J. O., Chase, G. A., & Folstein, S. E. (1992). Social language use in parents of autistic individuals. *Psychological Medicine, 22*(01), 245–254.

Landa, R. J., Holman, K. C., & Garrett-Mayer, E. (2007). Social and communication development in toddlers with early and later diagnosis of autism spectrum disorders. *Archives of General Psychiatry, 64*(7), 853–864. https://doi.org/10.1001/archpsyc.64.7.853.

Landa, R. J., Holman, K. C., O'Neill, A. H., & Stuart, E. A. (2011). Intervention targeting development of socially synchronous engagement in toddlers with autism spectrum disorder: A randomized controlled trial. *Journal of Child Psychology and Psychiatry, 52*(1), 13–21. https://doi.org/10.1111/j.1469-7610.2010.02288.

Lavery, M. R., Acharya, P., Sivo, S. A., & Xu, L. (2017). Number of predictors and multicollinearity: What are their effects on error and bias in regression. *Communications in Statistics—Simulation and Computation*. https://doi.org/10.1080/03610918.2017.1371750.

Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (1999). *Autism Diagnostic Observation Schedule (ADOS)*. Los Angeles: Western Psychological Services.

Lord, C., Rutter, M., DiLavore, P. C., Risi, S., Gotham, K., & Bishop, S. (2012). *Autism Diagnostic Observation Schedule* (2nd ed.). Torrance: Western Psychological Services.

Losh, M., & Piven, J. (2007). Social-cognition and the broad autism phenotype: Identifying genetically meaningful phenotypes. *Journal of Child Psychology and Psychiatry, 48*(1), 105–112.

Malesa, E., Foss-Feig, J., Yoder, P., Warren, Z., Walden, T., & Stone, W. (2012). Predicting language and social outcomes at age 5 for later-born siblings of children with autism spectrum disorders. *Autism, 17*(5), 558–570.

Messinger, D., Young, G. S., Ozonoff, S., Dobkins, K., Carter, A., Zwaigenbaum, L., et al. (2013). Beyond autism: A baby siblings research consortium study of high-risk children at three years of age. *Journal of the American Academy of Child and Adolescent Psychiatry, 52*(3), 300–308. https://doi.org/10.1016/j.jaac.2012.12.011.

Miller, M., Young, G. S., Hutman, T., Johnson, S., Schwichtenberg, A. J., & Ozonoff, S. (2015). Early pragmatic language difficulties in siblings of children with autism: Implications for DSM-5 social communication disorder? *Journal of Child Psychology and Psychiatry, 56*(7), 774–781. https://doi.org/10.1111/jcpp.12342.

Mullen, E. M. (1995). *Mullen Scales of Early Learning*. Circle Pines: American Guidance Service.

Mundy, P., Block, J., Delgado, C., Pomares, Y., Van Hecke, A. V., & Parlade, M. V. (2007). Individual differences and the development of joint attention in infancy. *Child Development, 78*(3), 938–954.

Mundy, P., Sullivan, L., & Mastergeorge, A. (2009). A parallel and distributed processing model of joint attention, social-cognition and autism. *Autism Research, 2*(1), 2–21.

O'Neill, D. (2007). The Language Use Inventory for young children: A parent-report measure of pragmatic language development for 18- to 47-month-old children. *Journal of Speech, Language, & Hearing Research, 50*(1), 214–228.

Ozonoff, S., & Miller, J. N. (1996). An exploration of right-hemisphere contributions to the pragmatic impairments of autism. *Brain and Language, 52*(3), 411–434.

Ozonoff, S., Young, G., Belding, A., Hill, M., Hill, A., Hutman, T., et al. (2014). The broader autism phenotype in infancy: When does it emerge? *Journal of the American Academy of Child and Adolescent Psychiatry, 53*(4), 398–407. https://doi.org/10.1016/j.jaac.2013.12.020.

Paul, R., Orlovski, S. M., Marcinko, H. C., & Volkmar, F. (2009). Conversational behaviors in youth with high-functioning ASD and Asperger syndrome. *Journal of Autism and Developmental Disorders, 39*(1), 115–125. https://doi.org/10.1007/s10803-008-0607-1.

Piven, J., Palmer, P., Landa, R., Santangelo, S., Jacobi, D., & Childress, D. (1997). Personality and language characteristics in parents from multiple-incidence autism families. *American Journal of Medical Genetics (Neuropsychiatric Genetics), 74*(4), 398–411.

Prutting, C. A., & Kirchner, D. M. (1987). A clinical appraisal of the pragmatic aspects of language. *Journal of Speech and Hearing Disorders, 52*(2), 105–119.

Roth, F. P., & Spekman, N. J. (1984a). Assessing the pragmatic abilities of children: Part 1. Organizational framework and assessment parameters. *Journal of Speech and Hearing Disorders, 49*(1), 2–11.

Roth, F. P., & Spekman, N. J. (1984b). Assessing the pragmatic abilities of children: Part 2. Guidelines, considerations, and specific evaluation procedures. *Journal of Speech and Hearing Disorders, 49*(1), 12–17.

Simmons, E. S., Paul, R., & Volkmar, F. (2014). Assessing pragmatic language in autism spectrum disorder: The Yale in vivo Pragmatic Protocol. *Journal of Speech, Language, and Hearing Research, 57*(6), 2162–2173. https://doi.org/10.1044/2014_JSLHR-L-14-0040.

Sperber, D., & Wilson, D. (1995). *Relevance: Communication and cognition* (2nd ed.). Malden: Blackwell Publishing.

statistics.laerd.com (2018). *Principal components analysis (PCA): Background & requirements*. Retrieved from https://statistics.laerd.com/premium/spss/pca/pca-in-spss-3.php.

Tager-Flusberg, H., & Caronna, E. (2007). Language disorders: Autism and other pervasive developmental disorders. *Pediatric Clinics of North America, 54*, 469–481.

Tomasello, M. (2001). Perceiving intentions and learning words in the second year of life. In M. Bowerman & S. C. Levinson (Eds.), *Language acquisition and conceptual development* (pp. 132–158). New York: Cambridge University Press.

Tomasello, M., Carpenter, M., Call, J., Behne, T., & Moll, H. (2005). Understanding and sharing intentions: The origins of cultural cognition. *Behavioral and Brain Sciences, 28*, 675–690.

Turkstra, L. S., Clark, A., Burgess, S., Hengst, J. A., Wertheimer, J. C., & Paul, D. (2017). Pragmatic communication abilities in children and adults: Implications for rehabilitation professionals. *Disability and Rehabilitation, 39*(18), 1872–1885.

Van Hecke, A. V., Mundy, P. C., Acra, C. F., Block, J. J., Delgado, C. E., Parlade, M. V., et al. (2007). Infant joint attention, temperament, and social competence in preschool children. *Child Development, 78*(1), 53–69.

Virkud, Y. V., Todd, R. D., Abbacchi, A. M., Zhang, Y., & Constantino, J. N. (2009). Familial aggregation of quantitative autistic traits in multiplex versus simplex autism. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 150B*(3), 328–334. https://doi.org/10.1002/ajmg.b.30810.

Wetherby, A., & Prizant, B. (2002). *Communication and Symbolic Behavior Scales Developmental Profile*. Baltimore: Brookes.

World Health Organization. (2011). *World report on disability*. Geneva: WHO Press.