

Research Report

Coordination is key: Joint attention and vocalisation in infant siblings of children with Autism Spectrum Disorder

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Abstract

Background: Research indicates that social communicative behaviours develop atypically during the second year in Autism Spectrum Disorder (ASD). This study evaluated whether these behaviours also differed in the extent to which they were coordinated across modalities.

Aims: To measure joint attention behaviours (e.g., gaze shifts, gestures), vocalisations and their coordination among a cohort of infants with an older sibling with ASD (heightened risk—HR).

Methods & Procedures: This prospective longitudinal study examined 50 HR infants at 14, 18 and 24 months. The Early Social Communication Scales (ESCS)—a structured toy-play task that assesses infant joint attention behaviour—was administered to infants at each age point in the home. Infants' joint attention behaviours, vocalisations and instances where they overlapped were coded from videos. At 36 months, nine infants received an ASD diagnosis (HR-ASD), 15 had a significant language delay (HR-LD) and 26 were classified no diagnosis (HR-ND).

Outcomes & Results: Findings revealed that HR-ASD infants produced fewer advanced joint attention behaviours, and their vocalisations were less frequent and less advanced than HR-LD and HR-ND infants. Notably, HR-ASD infants also coordinated these behaviours together less frequently than their HR peers.

Conclusions & Implications: Differences in the coordination of early communicative behaviours may have negative cascading effects on social and language development for infants who develop ASD. Current intervention practices may be complemented by efforts to increase the coordinated quality of communicative behaviours.

Keywords: Autism Spectrum Disorder (ASD), gesture, language delay, outcome, preverbal.

What this paper adds

What is already known on the subject

Neurotypical infants frequently vocalize during bouts of joint attention—this reflects a directedness on the part of the infant. Considerable evidence indicates both early social behaviour and vocal development are atypical in ASD; yet less is known about whether their coordination also differs.

What this paper adds to existing knowledge

Results replicate prior accounts that joint attention behaviour and vocalisations are less frequent among infants who develop ASD. Importantly, they are also significantly less coordinated than both neurotypical and language-delayed peers.

What are the potential or actual clinical implications of this work?

Cross-modal coordination distinguished infants with eventual ASD from peers with language delay, which may have relevance for earlier detection efforts. Additionally, findings suggest that early intervention efforts aimed at increasing social communication may benefit from a focus on increasing the coordinated quality of target behaviours.

Introduction

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social communication and social interaction, as well as the presence of repetitive behaviours and restricted interests or activities (DSM-5; American Psychiatric Association 2013). Although caregivers and researchers report features of ASD in the first years of life, reliable diagnosis is difficult before age 2 years (e.g., Chawarska *et al.* 2014). As a result, researchers have studied a group of infants at heightened risk (HR) for an ASD diagnosis (namely, the younger siblings of older children with ASD); for HR infants, the ASD recurrence rate is estimated at 18.7% (Ozonoff *et al.* 2011). Observing this population prospectively from infancy allows researchers to assess early development in infants who later develop ASD and identify potential early markers of the disorder (Goldberg *et al.* 2005; Yirmiya *et al.* 2006).

Delays in early communication are well documented in HR infants who are eventually diagnosed with ASD (see Jones *et al.* 2014 for a review). Additionally, in comparison to infants with no family history of ASD (low risk—LR), even HR infants who do not go on to receive an ASD diagnosis have an elevated likelihood of language delays (Cassel *et al.* 2007, Goldberg *et al.* 2005, Patten *et al.* 2014, Yirmiya *et al.* 2006). The present study, therefore, was designed to investigate two areas of communicative development—joint attention and vocalisation—in a sample of HR infants. These behaviours were measured longitudinally at 14, 18 and 24 months in three groups of HR infants: infants later diagnosed with ASD (HR-ASD), infants with eventual language delays (HR-LD) and infants without a diagnosis or language delay (no diagnosis; HR-ND). Joint attention and vocalisation are especially important as they constitute raw materials for social communication during infancy (e.g., Bakeman and Adamson 1984, Bruner 1977, Gros-Louis *et al.* 2006). Indeed, joint attention and vocal development both predict later language ability (Charman *et al.* 2000, Morales *et al.* 2000, Murray *et al.* 2008) and serve as foundational targets for early intervention (e.g., Mundy and Crowson 1997).

To date, the literature on HR infants has typically examined these early communicative behaviours in isolation. Yet, these skills not only develop simultaneously during infancy: they are also frequently used in combination when infants interact with caregivers and peers

(Harding and Golinkoff 1979, Bakeman and Adamson 1984). For example, an infant may look at a desired but out-of-reach object, shift gaze to a caregiver, and vocalize to attract the attention of the caregiver while simultaneously reaching to request the object. It has been suggested that this type of coordination makes these behaviours more salient to the caregiver, and therefore potentially more successful in eliciting social responses (e.g., Parladé and Iverson 2015, Iverson and Wozniak 2016). Given the dearth of research examining joint attention and vocalisation production simultaneously, the present study focused on the development and coordination of these behaviours in HR infants across the second year.

Joint attention and vocalisation in typical development

Joint attention—the shared, triadic attention of two social partners with an object—is a critical component of early communicative growth in neurotypical infants and plays a role in early language development (Bakeman and Adamson 1984, Morales *et al.* 2000) and cognition (Morales *et al.* 2000, Mundy *et al.* 2007). By 6 months, infants follow the gaze of a caregiver to a new location (Murray *et al.* 2008). By 12 months they behave in a variety of ways to establish joint attention; for instance, they alternate gaze between a caregiver and objects, they reach to request objects and they hold up attractive objects to show others (e.g., Carpenter *et al.* 1998). These behaviours typically serve two functions: (1) they establish shared attention to an object or event with a social partner to indicate interest (e.g., making eye contact with an adult while holding a toy); and/or (2) they request the help of a social partner to achieve a desired goal (e.g., handing a closed container to an adult to open it).

While the literature on joint attention has focused on nonverbal behaviours such as eye gaze and gesture, infants also vocalize during social interactions. At the same time that joint attention behaviours emerge and develop there are simultaneous changes in vocal development. Early vocalisations consist of vowel-only sounds (e.g., [ah]), but by 6 months infants begin to produce syllabic vocalisations, which contain consonant–vowel units (e.g., [bababa]; Oller *et al.* 1999, Paul *et al.* 2011). Throughout the first year these increase in frequency and complexity (e.g., Oller *et al.* 1999). These

vocalisations are an important milestone as they serve as a prerequisite for later word production (e.g., Oller *et al.* 1999).

As infants develop, they begin to combine multiple communicative behaviours, such as pairing vocalisations with gestures (Bates *et al.* 1979, Bruner 1977). Pairing vocalisations with other nonverbal communicative behaviours may be particularly important for communication development because vocalisations are influential in directing caregivers' attention and responses. For example, a caregiver who is across the room is more likely to respond to an infant's reach/request gesture paired with a vocalisation than to the same gesture alone (Goldstein *et al.* 2003, Gros-Louis *et al.* 2006, Harding and Golinkoff 1979, Hsu, Fogel, and Messinger 2001). Studies suggest that caregivers frequently respond contingently to their infants' vocalisations (e.g., Gros-Louis *et al.* 2006, but see Fagan and Doveikis 2017) and provide richer responses to syllabic vocalisations (e.g., by imitating or expanding on the infants' vocalisation) relative to less advanced vowel-only vocalisations (Gros-Louis *et al.* 2006). Considering that extensive research has documented a robust relation between joint attention behaviours and later language ability (e.g., Charman *et al.* 2000, Morales *et al.* 2000, Murray *et al.* 2008), it is surprising that there has been relatively little work describing the role of vocalisations in enhancing infants' joint attention behaviour (Winder *et al.* 2013). Understanding the coordination of these behaviours during social interactions may be useful in understanding communicative development in both typical and atypical development.

Joint attention and vocalisation in HR infants and toddlers with ASD

Infants with ASD demonstrate differences in the frequency and content of their joint attention behaviours compared with neurotypical infants (Charman 1998, Goldberg *et al.* 2005, Shumway and Wetherby 2009, Toth *et al.* 2006). In addition, there is evidence that HR infants as a group—including those who do not receive an ASD diagnosis—show differences in joint attention behaviours relative to LR samples. On average, HR infants without ASD produce joint attention behaviours at rates more similar to those of children with ASD than to those of LR peers (Cassel *et al.* 2007, Goldberg *et al.* 2005, Yirmiya *et al.* 2006). For example, Goldberg *et al.* (2005) used the Early Social Communication Scales (ESCS; Mundy *et al.* 2003)—a standardized assessment aimed at eliciting joint attention behaviours—to compare three groups of children: LR toddlers (mean age = 15.3 months), HR toddlers without ASD (mean age = 17.1 months) and young children with ASD

(mean age = 29.9 months). There were no significant differences in joint attention behaviours between children with ASD and the HR toddlers without ASD. However, both groups produced significantly fewer joint attention behaviours than the LR toddlers.

Differences in pre-speech vocalisations and vocal development have also been described in studies of HR infants, and especially HR-ASD infants. For example, Paul *et al.* (2011) examined vocalisations from 6 to 12 months in HR and LR infants. Although HR infants did not differ from LR infants in overall frequency of vocalisations, there were notable differences in the quality and types of vocalisations observed. Specifically, HR infants produced more non-speech (e.g., growl, yell, squeal) and fewer syllabic vocalisations than their LR peers (see also Plumb and Wetherby 2013, Schoen *et al.* 2011). While the Paul *et al.* (2011) study did not have diagnostic outcomes for their sample, other research has demonstrated that infants with a later ASD diagnosis exhibit delays in the onset of babbling (i.e., syllabic vocalisations with repeated consonant–vowel units; e.g., [bababa], [mamama]) and lower rates of overall and syllabic vocalisation compared with neurotypical peers (Patten *et al.* 2014). In addition, the social function of vocalisations appears to differ in ASD: HR-ASD infants produce fewer vocalisations directed at social partners compared with their LR peers (Ozonoff *et al.* 2010). Although the current research has established differences between LR and HR infants as a group, and between ASD infants and neurotypical infants, none of these studies was able to compare vocal development among HR infants with and without language delays or ASD. Thus, the specificity of delays observed in vocal development among HR infants remains unclear.

Given the apparent differences in joint attention and vocalisations reported in studies that have considered these behaviours separately, it is unsurprising that HR infants with a later ASD diagnosis also coordinate them less frequently (Parladé and Iverson 2015, Winder *et al.* 2013). In a longitudinal study comparing LR, HR-ASD, HR-LD and HR-ND infants, Parladé and Iverson (2015) examined the development of gesture + vocalisation coordinations (i.e., infant produces gesture while simultaneously vocalizing) and found that while both HR-LD and HR-ASD infants exhibited initial delays in production of these coordinations, only HR-ASD infants showed significantly slowed growth over time compared with all other groups. Indeed, the slope of growth for the HR-ASD group was close to zero, indicating that although their HR peers were increasingly coordinating gestures with vocalisations, HR-ASD infants were not.

This pattern is striking, and further research is needed to determine whether it is robust. Differences in isolated joint attention behaviours and isolated vocalisations appear to be characteristic of HR infants as

a group. Differences in their coordination, however, appear to be specific to infants with ASD. Identifying qualities of communicative development that are unique to ASD has valuable theoretical and clinical implications—particularly in light of the substantial variability in communication that characterizes HR infants as a group. The present study set out to extend this work by assessing the coordination of joint attention and vocalisations longitudinally in three groups of HR infants who varied in developmental outcome at age 3 years.

Present study

The aim of this research was to examine the development of: (1) the frequency and quality of joint attention behaviours; (2) the frequency and quality of vocalisations; and (3) the coordination of joint attention behaviours with vocalisations in HR-ASD, HR-LD and HR-ND infants. These behaviours were assessed longitudinally at 14, 18 and 24 months of age during administration of the ESCS. By focusing on HR infants, we can examine individual variability in the behaviours under consideration in a population with a variety of developmental outcomes during a time period before many children receive a diagnosis.

Materials and methods

Participants

A total of 50 (30 male; 20 female) full biological, later born infant siblings of children with a confirmed diagnosis of ASD participated in the present study. Participants were recruited through a university Autism Research Program and through flyers, professional referrals and word of mouth. Before enrolment, older siblings were administered the Autism Diagnostic Observation Schedule—Generic (ADOS-G; Lord *et al.* 2000) by a trained clinician. Infants were eligible to participate in the study only if their older sibling scored above the Autism cut-off on the ADOS-G and met DSM-IV criteria for Autistic Disorder. Parents signed informed consents before participation; the research protocol was approved by the University of Pittsburgh Institutional Review Board. All infants were from full-term, uncomplicated pregnancies and from monolingual English-speaking families. A total of 43 infants were Caucasian, four were Hispanic, two were Asian American and one was African American.

Outcome classification

At 36 months, all infants were administered the ADOS-G by an experienced clinician. The ADOS-G reliably differentiates children with ASD from children who

might have other developmental delays or no ASD diagnosis (Lord *et al.* 2000). Diagnoses were made based on administration of the ADOS, DSM-IV-TR criteria and clinical best estimate. Clinicians were formally trained on the administration of the ADOS and met official research reliability criteria. They were naïve to all prior study data. Based on these criteria, nine infants (six males, 3 females) were classified as HR-ASD.

HR infants were classified as language delayed (HR-LD) if they did not receive a diagnosis of ASD and either of the following criteria were met (Iverson *et al.* 2018, Parladé and Iverson 2015, West *et al.* 2018): (1) standardized scores on the MacArthur–Bates Communicative Development Inventory, Words and Sentences (CDI-II; Fenson *et al.* 1993) or CDI-III at or below the 10th percentile at more than one time point between 18 and 36 months (e.g., Ellis Weismer and Evans 2002, Heilmann *et al.* 2005); or (2) standardized score on the CDI-III at or below the 10th percentile and a standardized score on the Receptive and/or Expressive Language subscales of Mullen Scale of Early Learning (MSEL; Mullen 1995) equal to or greater than 1.5 SD (standard deviation) below the mean at 36 months (e.g., Landa and Garrett-Mayer 2006, Ozonoff *et al.* 2010). Based on these criteria, 15 infants (nine males, 6 females) were classified as HR-LD. The remaining 26 infants (15 males, 11 females) did not meet the criteria for ASD or LD and were classified as having no diagnosis (HR-ND).

Demographic information for infants in each outcome group are presented in table 1. All demographic information was collected at the first visit. As can be seen, the majority of families were Caucasian and held college or graduate degrees. Mann–Whitney *U*-tests revealed that mothers' years of education did not differ statistically between the outcome groups, nor did mean ages of mothers or fathers. Although information on family income was unavailable, parental occupations were identified in order to provide a general index of socioeconomic status. Over half the mothers in the study (56%) did not work outside the home, so paternal occupations were used to calculate Nakao–Treas occupational prestige scores, a measure of occupational social standing that correlates highly with socioeconomic factors (Nakao and Treas 1994). For one LD family it was impossible to identify the father's occupation with enough precision to assign a prestige score. Mann–Whitney *U*-tests with occupational prestige data from the remaining families indicated no statistically significant difference between groups.

Procedure

Infants were visited at home on a monthly basis between 5 and 14 months, with follow-up visits at 18, 24 and 36 months. At the 14-, 18- and 24-month visits, infants

Table 1. Demographic information for the three outcome groups

	HR-ND	HR-LD	HR-ASD
<i>Gender</i>			
Male	11 (42.3%)	6 (40.0%)	6 (66%)
Female	15 (57.7%)	9 (60.0%)	3 (33%)
Racial/ethnic minority	0 (0%)	4 (26.7%)	3 (33%)
Speech intervention ^a	1 (3.8%)	8 (53.3%)	6 (66%)
<i>Mean age (years)</i>			
Mother	33.65 (3.14)	34.73 (3.36)	31.00 (4.50)
Father	36.57 (5.40)	37.87 (3.91)	34.78 (4.82)
<i>Maternal education</i>			
Postgraduate	12 (46.2%)	6 (40.0%)	2 (22.2%)
College	13 (50.0%)	8 (53.3%)	3 (33.0%)
High school	1 (3.8%)	1 (6.7%)	4 (44.4%)
<i>Paternal education</i>			
Postgraduate	6 (23.1%)	6 (40.0%)	4 (44.4%)
College	19 (73.1%)	8 (53.3%)	3 (33.0%)
High school	1 (22.2%)	1 (22.2%)	2 (22.2%)
Mean paternal occupational prestige	53.80 (14.95)	52.02 (14.52)	53.38 (17.32)

Note: ^aData reflect the numbers of infants who were receiving speech-language services in each outcome group.

were administered the ESCS (Mundy *et al.* 2003). The ESCS is a 15–20-min experimenter-administered semi-structured toy-play interaction widely used to assess nonverbal joint attention behaviours at these ages. Administration occurred at a table with the child sitting across from the experimenter. The same toys were presented at each visit and included wind-up toys, a puppet, a book, posters and social toys (e.g., sunglasses, a bowl and spoon, a comb and a hat). Trained examiners administered the ESCS in accordance with standard procedures outlined in the abridged ESCS manual (Mundy *et al.* 2003). In addition, because the goal of this research was to investigate the coordination of nonverbal joint attention behaviours as elicited in the ESCS and infant vocalisation, infants wore a wireless microphone housed in a vest worn over their clothing, allowing for high-quality recording of vocalisations.

Coding

Coding for this study was completed by members of the research team naive to group membership. Two different teams completed the ESCS coding and the vocalisation coding.

ESCS

The ESCS coding followed standard procedures described in the ESCS manual (Mundy *et al.* 2003). The ESCS is a reliable and valid measure of early social communication development (e.g., Mundy *et al.* 1988, 1994, 1995) that focuses on two types of spontaneous joint attention behaviours: initiating behavioural requests (IBR) and initiating joint attention (IJA). IBR

behaviours (e.g., extending an arm towards a toy) are those that request a social partner to do something (e.g., hand the toy to the child). IJA behaviours are those that function to establish shared visual attention with the experimenter to communicate interest in an object or activity; in either mutual gaze or shared gaze towards an object (e.g., holding up a toy to show the experimenter).

IJA and IBR behaviours were further classified as low or high following the guidelines in the manual (Mundy *et al.* 2003). Low behaviours (e.g., alternating gaze between an object and the experimenter) are those that typically emerge earlier in development (Bakeman and Adamson 1984, Seibert *et al.* 1984, Mundy *et al.* 1994). High behaviours reflect more developmentally advanced actions (e.g., pointing to a distal object while making eye contact with the experimenter). All observable behaviours were coded from videotaped ESCS administrations with no minimum duration necessary. High and low behaviours were coded as distinct behaviours, and infants did not need to perform a low behaviour in order to get credit for a high behaviour. Further description of these categories can be found in table 2.

In this study, interrater reliability was assessed via intra-class correlation coefficients (ICCs) computed using the raw total counts between raters on 15% ($n = 22$) of the videos. Agreement was .99 for low IJA, .98 for high IJA behaviours, .96 for low IBR and 1.00 for high IBR.

Vocalisations

All vocalisations (excluding sneezing, coughing, breathing, laughing and other vegetative noises) produced by

Table 2. Description of Early Social Communication Scales (ESCS) codes

Joint attention category	Base behaviour	Description
IJA (low)	Eye contact	Child makes spontaneous eye contact with the tester while the child is holding or touching a toy
IJA (high)	Alternate gaze	Child alternates its gaze between an activated mechanical toy and the tester's eyes
	Point	Child spontaneously points with an index finger towards an activated mechanical toy, a picture in book or distal object
IBR (low)	Show	Child raises a toy upward to show the tester; does <i>not</i> give toy to the tester
	Eye contact	Child makes spontaneous eye contact with the tester when the object is out of reach
IBR (high)	Reach	Child extends an arm towards an out-of-reach toy
	Appeal	Child combines a reach and eye contact with the tester
	Give	Child extends an object towards the tester and gives up the toy
	Point	Child spontaneously points to an inactive toy

Notes: High and low behaviours can be done with or without eye contact. Children did not need to produce a low-behaviour skill to receive credit for high behaviour. IJA, initiating joint attention; IBR, initiating a behavioural request.

infants during the ESCS administration were identified and coded. Separate vocalisations were coded if there was at least a 1 s pause or an audible breath between vocalisations. All non-word vocalisations were categorized according to whether or not they contained a consonant sound. Vocalisations that did not contain a consonant sound (e.g., [aaaaa], [ooooo]) were coded as vowel only. Those that contained a consonant sound (e.g., [maba], [dadada]) were categorized as syllabic. Infants were credited with producing a word if they produced either an English word (e.g., 'ball'), or a vocalisation that was consistently used to refer to a specific object (e.g., [baba] for 'ball'). Interrater reliability was assessed via ICCs computed using raw total counts of each type of vocalisation on 20% ($n = 30$) of the videos. Reliability was .93 for vowel-only vocalisations, .91 for syllabic vocalisations and .98 for words.

Coordination

Coordination of joint attention behaviour and vocalisation was identified any time there was an overlap in time between a joint attention behaviour and a vocalisation. If an infant vocalized at any point during a joint attention behaviour, it was considered to be a coordination. Coordinations were identified separately for IJA and IBR behaviours.

Results

This study was designed to examine the development of joint attention behaviours, vocalisations and their coordination in three groups of HR infants with varying developmental outcomes. Although there were no significant differences in ESCS observation lengths as a function of outcome group, $F(2, 46) = 1.820$, $p = .173$; $M_{ND} = 16.95$, $SD_{ND} = 2.98$, $M_{LD} = 16.94$, $SD_{LD} = 2.53$, $M_{ASD} = 15.39$, $SD_{ASD} = 3.17$, frequencies for each variable were converted to rates per

10 min in order to control for slight differences in duration. Furthermore, initial examination of distributions indicated that all variables, which possessed an artificial lower bound at zero, had a positive skew; they were therefore logarithmically or square root transformed before conducting statistical analyses. Mann–Whitney U -tests did not reveal any significant gender differences in any of the variables considered. Therefore, all analyses were conducted with data collapsed across males and females.

Joint attention behaviours

Rates per 10 min of the four joint attention behaviours (low IBR, high IBR, low IJA, high IJA) were calculated for each infant; descriptive statistics are presented in table 3. In order to test for group differences in these behaviours over time, 3×3 repeated-measures analyses of variance (ANOVAs) with Age (14, 18, 24 months) as a within-subjects factor and Outcome (HR-ND, HR-LD, HR-ASD) as a between-subjects factor were computed separately for each behaviour. As can be seen in table 3, HR-ASD infants tended to produce low IBR, high IBR and low IJA behaviours less frequently than their HR-ND and HR-LD peers. However, this trend did not achieve significance: there were no main effects of Age or Outcome for these three variables. The interaction terms were also non-significant.

With regard to high IJA behaviours, however, a different pattern emerged. Results of the ANOVA revealed a significant overall effect of Outcome, $F(2,47) = 14.430$, $p < .001$, $\eta_p^2 = 0.38$. Follow-up simple effects analyses using a Sidak adjustment indicated that the differences between the HR-ASD and HR-ND and the HR-ASD and HR-LD groups were both significant, $p < .001$ and $p = .001$, indicating that the HR-ASD group produced fewer high IJA behaviours overall compared with their peers. Although there was also a tendency for the HR-LD group to produce high

Table 3. Production of joint attention behaviours by outcome group at 14, 18 and 24 months

Variable	Age (months)	HR-ND (<i>n</i> = 26)		HR-LD (<i>n</i> = 15)		HR-ASD (<i>n</i> = 9)	
		Mean (SD)	% of infants	Mean (SD)	% of infants	Mean (SD)	% of infants
Low IBR	14	6.79 (5.16)	100%	6.87 (7.41)	100%	5.79 (3.04)	100%
	18	7.83 (4.89)	100%	7.67 (4.46)	100%	5.09 (2.72)	100%
	24	6.25 (3.07)	100%	6.75 (5.15)	100%	4.64 (4.44)	100%
High IBR	14	7.30 (6.20)	100%	5.95 (3.94)	100%	8.54 (5.19)	100%
	18	8.95 (6.10)	96.2%	7.14 (6.99)	100%	6.31 (3.99)	100%
	24	9.26 (5.76)	96.2%	8.39 (6.60)	100%	6.73 (3.53)	100%
Low IJA	14	8.25 (4.23)	100%	9.54 (5.81)	100%	9.83 (8.26)	100%
	18	8.35 (6.00)	92.3%	8.65 (6.65)	93.3%	8.87 (5.38)	100%
	24	9.74 (4.88)	100%	9.94 (4.20)	100%	6.34 (5.28)	100%
High IJA	14	2.76 (2.60)	96.2%	1.66 (2.73)	73.3%	0.46 (0.64)	44.4%
	18	1.49 (1.50)	73.1%	1.35 (1.38)	80.0%	0.29 (0.15)	33.0%
	24	1.85 (1.37)	96.2%	1.61 (1.52)	66.6%	0.06 (0.23)	11.1%

Note: ND, no delay; LD, language delay; ASD, Autism Spectrum Disorder; SD, standard deviation; the number of infants = the percentage of infants who produced the behaviour.

Table 4. Rates per 10 min of vocalisation production by outcome group at 14, 18 and 24 months

Vocalisation type	Age (months)	HR-ND (<i>n</i> = 26)	HR-LD (<i>n</i> = 15)	HR-ASD (<i>n</i> = 9)
		Mean (SD)	Mean (SD)	Mean (SD)
Vowel only	14	19.08 (13.98)	14.45 (12.00)	4.47 (2.23)
	18	16.13 (11.71)	11.09 (12.32)	7.83 (7.28)
	24	5.34 (6.98)	8.56 (9.12)	9.27 (8.75)
Syllabic	14	17.18 (14.31)	16.19 (14.54)	6.34 (6.19)
	18	21.67 (26.54)	14.25 (14.88)	5.77 (7.50)
	24	15.62 (10.77)	25.62 (19.57)	11.62 (10.40)
Word	14	0.07 (0.25)	0.00 (0.00)	0.00 (0.00)
	18	4.13 (10.45)	0.21 (0.38)	0.71 (1.91)
	24	23.44 (20.17)	2.21 (9.09)	2.12 (4.94)

Note: ND, no diagnosis; LD, language delay; ASD, Autism Spectrum Disorder; SD, standard deviation.

IJA behaviours less frequently than their HR-ND peers, this difference did not achieve statistical significance, $p = .081$.

As can also be seen in table 3, the percentage of HR-ASD infants producing even a single high IJA behaviour was low at all three age points and decreased over time. This pattern was only apparent in the HR-ASD group. Two-tailed Fisher's exact tests carried out at each age indicated that at 14 months the percentage of children producing any high IJA behaviours in the HR-ASD group did not differ significantly from that for the HR-LD group, $p = .212$, but did differ from the HR-ND group, $p = .002$. However, at 18 and 24 months the percentages of HR-ASD infants producing high IJA behaviours were significantly lower than those for both the HR-ND, $p = 0.050$ and < 0.001 , and HR-LD groups, $p = 0.036$ and 0.013 .

Vocalisations

Next, we examined infant production of the three types of vocalisations (vowel only, syllabic, words)

over time by calculating rates per 10 min for each vocalisation type for individual infants. Descriptive statistics are presented in table 4 for each vocalisation type, age and outcome group; data are also plotted in figure 1. To examine differences in the types of vocalisations produced across the three age points in the three outcome groups, a 3 (Vocalisation type) \times 3 (Age) \times 3 (Outcome) repeated-measures ANOVA was carried out on the data. Results revealed significant main effects of Outcome, $F(2,47) = 6.23$, $p < 0.001$, $\eta_p^2 = 0.21$, Vocalisation type, $F(2,94) = 101.175$, $p < .001$, $\eta_p^2 = 0.68$, and Age, $F(2,94) = 9.522$, $p < .001$, $\eta_p^2 = 0.17$, as well as a significant interaction between Vocalisation type and Age, $F(4,188) = 30.781$, $p < .001$, $\eta_p^2 = 0.40$. However, these were qualified by a three-way interaction between Vocalisation type, Age and Outcome, $F(8,188) = 8.613$, $p < .001$, $\eta_p^2 = 0.27$, indicating that the relationship between infant age and vocal production varied between outcome groups.

To clarify the source of this three-way interaction, follow-up 3 (Age) \times 3 (Vocalisation type) repeated-measures ANOVAs were calculated separately for each

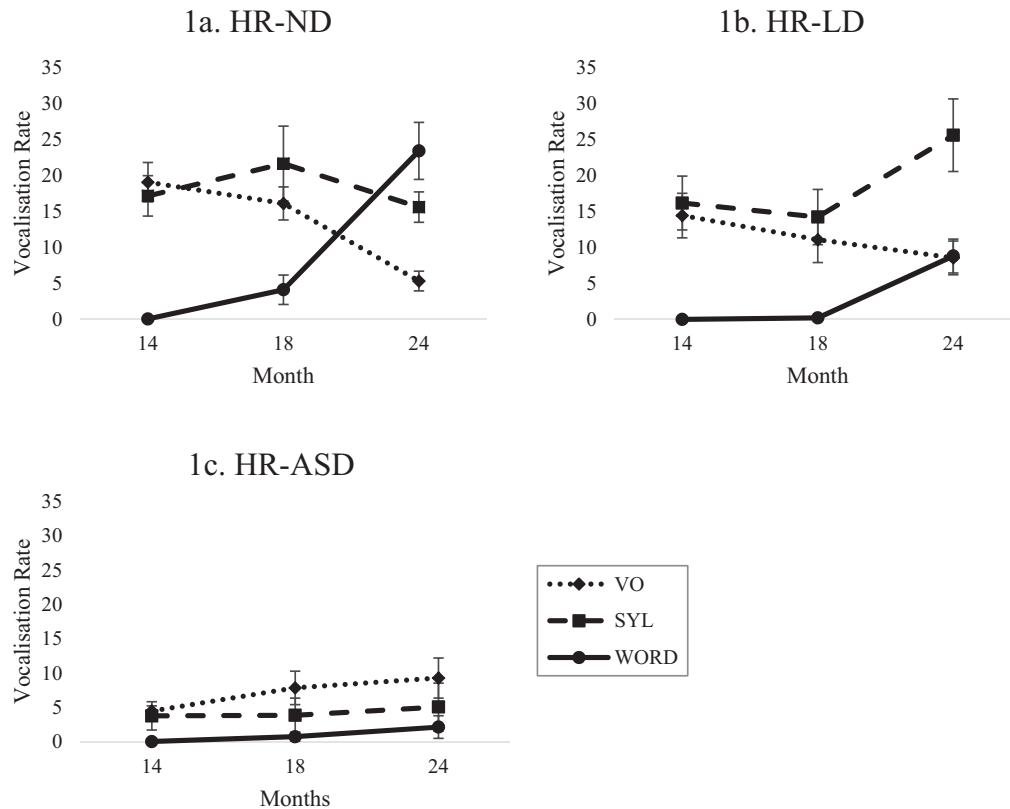


Figure 1. Rates per 10 min of vowel-only vocalisations, syllabic vocalisations and words produced at 14, 18 and 24 months by (a) HR-ND, (b) HR-LD and (c) HR-ASD infants. Error bars represent standard errors.

Outcome group. These revealed a significant Age by Vocalisation type interaction both for HR-ND, $F(4,100) = 71.71$, $p < .001$, $\eta_p^2 = 0.74$, and HR-LD infants, $F(4,56) = 10.61$, $p < .001$, $\eta_p^2 = 0.43$, but, notably, not for the HR-ASD group, $F(4,32) = 0.657$, $p = 0.63$, $\eta_p^2 = 0.08$. Thus, while ND and LD infants showed significant change with age in the frequency with which they produced different types of vocalisations, the ASD group did not. As can be seen in figure 1, among HR-ND infants, less complex vocalisations (i.e., vowel-only vocalisations) decreased with age, while words showed a sharp increase, particularly from 18 to 24 months. The HR-LD group showed a somewhat different pattern, with a smaller decrease in vowel-only vocalisations, but substantial increases in both syllabic vocalisations and word production between 18 and 24 months. In contrast, the HR-ASD group consistently produced vocalisations of all three types at low rates and with no age-related changes.

Coordination of joint attention behaviours and vocalisations

Finally, we examined how infants coordinated vocalisations with joint attention behaviours. For these

analyses, we calculated the proportions of joint attention behaviours that were paired with a vocalisation by dividing the total number of coordinations by the total number of joint attention behaviours produced. These were calculated separately for IJA and IBR behaviours. Using proportions controlled for differences in baseline frequencies of joint attention behaviours. Finally, low and high IJA behaviours were collapsed into a single category because many of the HR-ASD infants did not produce any high IJA behaviours and therefore did not have opportunities to combine them with vocalisations. Descriptive statistics for IJA + vocalisation, low IBR + vocalisation, and high IBR + vocalisation are presented in table 5. These data are also depicted in figure 2.

For IJA + vocalisations, a 3 (Age) \times 3 (Outcome) repeated-measures ANOVA yielded a significant main effect only for Outcome, $F(2,45) = 4.45$, $p = .017$, $\eta_p^2 = 0.17$. Simple effects analyses further revealed that the HR-ASD group produced a significantly lower proportion of IJA + vocalisation coordinations than the HR-ND, $p = .008$, and HR-LD groups, $p = .015$. The HR-ND group did not differ from the HR-LD group, $p = .949$.

A similar pattern was observed for low IBR + vocalisation coordinations. A 3 (Age) \times 3 (Outcome)

Table 5. Proportions of joint attention and vocalisation coordination by outcome group at 14, 18 and 24 months

	Age (months)	HR-ND (<i>n</i> = 26) Mean (SD)	HR-LD (<i>n</i> = 15) Mean (SD)	HR-ASD (<i>n</i> = 9) Mean (SD)
IJA + VOC	14	0.33 (0.26)	0.32 (0.34)	0.06 (0.08)
	18	0.58 (0.92)	0.26 (0.39)	0.15 (0.27)
	24	0.32 (0.33)	0.68 (0.84)	0.18 (0.13)
Low IBR + VOC	14	0.38 (0.26)	0.30 (0.27)	0.19 (0.14)
	18	0.45 (0.24)	0.35 (0.26)	0.24 (0.25)
	24	0.46 (0.23)	0.46 (0.17)	0.18 (0.17)
High IBR + VOC	14	0.41 (0.2)	0.29 (0.21)	0.17 (0.22)
	18	0.37 (0.29)	0.28 (0.23)	0.21 (0.24)
	24	0.49 (0.24)	0.47 (0.24)	0.22 (0.26)

Note: ND, no diagnosis; LD, language delay; ASD, Autism Spectrum Disorder; SD, standard deviation.

repeated-measures ANOVA indicated a significant main effect of Outcome only, $F(2,47) = 5.381$, $p = .008$, $\eta_p^2 = 0.19$. Again, simple effects analyses indicated that a significantly lower proportion of low IBR + vocalisation coordinations were produced by the HR-ASD group than both the HR-ND, $p = .002$, and HR-LD groups, $p = .033$. The ND and LD groups did not differ from one another, $p = .295$.

Finally, a 3 (Age) \times 3 (Outcome) repeated-measures ANOVA was calculated for high IBR + vocalisation coordinations. This revealed a significant main effect of Age, $F(2,88) = 3.428$, $p = .037$, $\eta_p^2 = 0.07$, and Outcome, $F(2,44) = 5.575$, $p = .007$, $\eta_p^2 = 0.20$. Simple effects comparisons indicated that although there was no significant change in coordinations from 14 to 18 months, $p = .878$; there was a significant increase in coordinations between 18 and 24 months, $p = .022$. With regard to the effect of outcome, differences between the HR-ND and HR-LD groups were not significant, $p = .150$, but the ASD group differed significantly from the HR-ND group, $p = .022$, and marginally from the HR-LD group, $p = .054$. Note that there were no statistical differences in the numbers of IBR behaviours produced between the three outcome groups (table 3); thus this difference emerges only in the coordination of IBR behaviours with vocalisations.

Discussion

The present study examined the production of joint attention behaviours, vocalisations and their coordination in infants at HR for ASD. Consistent with previous findings, HR infants who later received an ASD diagnosis differed in production of joint attention behaviours and vocalisations compared with their HR peers (Charman 1998, Goldberg *et al.* 2005, Shumway and Wetherby 2009, Toth *et al.* 2006). Importantly, they were also less likely to coordinate these behaviours. Below we discuss these findings in turn, focusing on how these differences

may impact infants' social interactions and subsequent communicative development, and we suggest some potential clinical implications.

Differences in joint attention behaviours and vocalisation in ASD

Examination of infants' joint attention behaviour indicated that for less advanced joint attention behaviours (e.g., reaching to request a desired object), there were no significant differences between HR-ASD infants and their peers. However, HR-ASD infants displayed far fewer advanced initiating joint attention behaviours (e.g., showing objects to others) than other HR infants. This is consistent with prior work, both using the ESCS—which actively elicits these behaviours—as well as naturalistic observation (Rozga *et al.* 2011, Watson *et al.* 2013).

Regarding vocalisations, both the HR-ND and HR-LD groups increased the frequency and complexity of their vocalisations over time (although, unsurprisingly, word production was attenuated for HR-LD infants relative to HR-ND infants). In contrast, the HR-ASD group consistently produced fewer vocalisations than their peers, and production did not increase over time. In addition, the vocalisations they produced were less complex, primarily vowel-only vocalisations. This aligns with previous studies that report disruptions in vocal development in ASD (Paul *et al.* 2011, Plumb and Wetherby 2013, Schoen *et al.* 2011).

Considered together, these differences in joint attention and vocal behaviours are likely to impact infants' social environments. Caregivers are sensitive to their infants' communicative actions and are more responsive to advanced behaviours (e.g., syllabic vocalisations are more likely to receive a response than vowel-only vocalisations; Gros-Louis *et al.* 2006). A study by Leezenbaum *et al.* (2014) examined maternal responses to HR and LR infants' gestures at 13 and 18 months. There were no differences in maternal responsiveness to infant

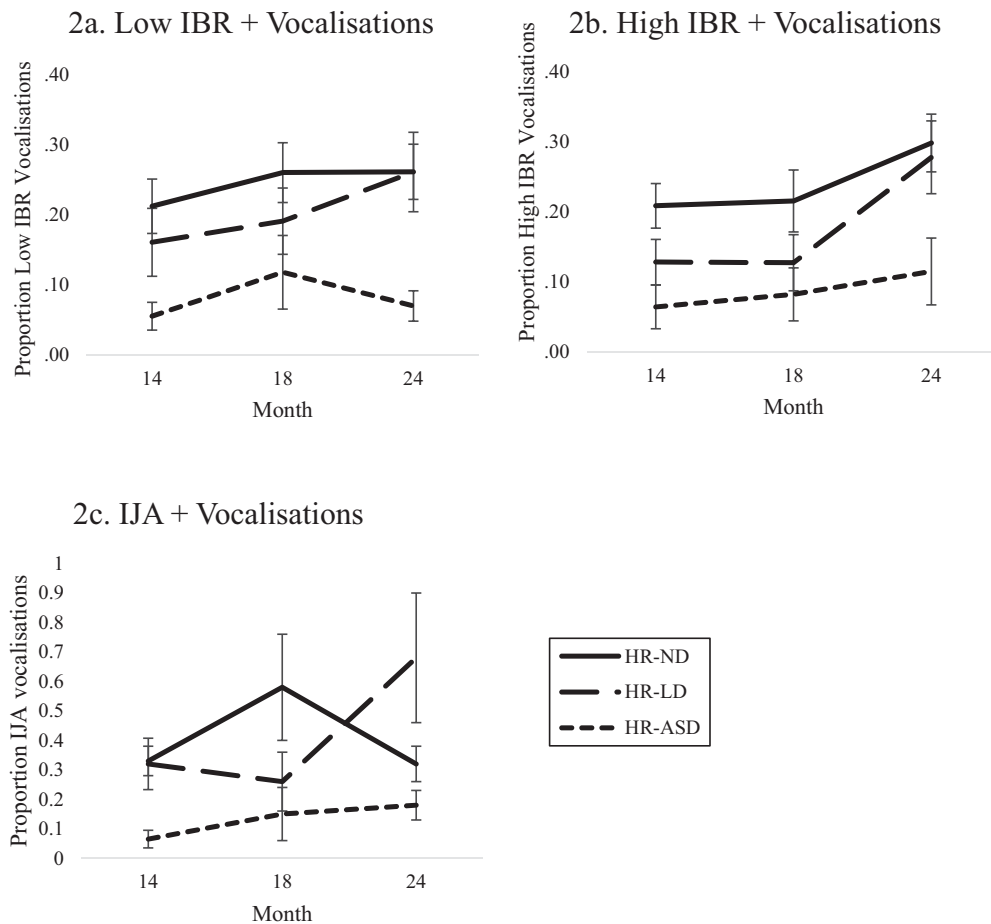


Figure 2. Proportions of (a) IJA + vocalisation, (b) low IBR + vocalisation and (c) high IBR + vocalisation coordinations produced at 14, 18 and 24 months by HR-ND, HR-LD and HR-ASD infants. Error bars represent standard errors.

gestures, but the types of gestures infants used differed between groups. Specifically, LR infants produced more point/show gestures, whereas HR infants tended to use reaches/gives. Analysis of mothers' responses to these two gesture categories indicated that both groups of mothers were significantly more likely to translate the referent (i.e., provide its label) of point/show compared with give/request gestures. Thus, because HR infants produced significantly fewer show and point gestures that were most likely to elicit a translation response, they received fewer translations, which are precisely the type of response that is effective for promoting word learning.

Although these results were obtained using an experimenter-administered assessment, they highlight an important principle: differences in *infant* communication likely engender differences in *caregiver* communication—they interact through a social feedback loop. To illustrate, consider an infant who produces syllabic vocalisations. Adults respond to these conversationally, providing valuable verbal input. In a continuous cycle of contingency, these responses reinforce

infants' more developmentally advanced vocalisations (e.g., Martinsen and Smith 1989) and assist them in acquiring new speech-like patterns (e.g., Goldstein and Schwade 2008). The present findings suggest that this feedback loop unfolds differently in ASD. Because HR-ASD infants produce less advanced and less frequent joint attention behaviours and vocalisations, and because these differences may lead to a reduction in opportunities for high quality caregiver responses, the nature of the social–communicative input they receive likely differs from that directed to HR infants without ASD (see Iverson and Wozniak 2016 for a more extended discussion). For HR-ASD infants who exhibit significant, early-appearing vulnerabilities in social–communicative development, these differences in output may influence input in ways that further constrain opportunities for learning.

Differences in coordinated communication in ASD

Although base rates of less advanced joint attention behaviours were similar across groups, differences emerged

when we analyzed their *coordination* with vocalisations, such that HR-ASD infants vocalized significantly less often during these moments than HR-LD and HR-ND peers. This result is consistent with prior work reporting that toddlers with ASD combine communicative behaviours less often than peers (Parladé and Iverson 2015, Shumway and Wetherby 2009, Wetherby *et al.* 2004) and with evidence indicating that difficulties with coordination of communicative behaviours is characteristic of communication in ASD (Shumway and Wetherby 2009, Stone *et al.* 1997).

Coordinating behaviour across modalities is a fundamental part of mature social communication, which involves a complex interplay between vocalisation, gesture, eye gaze and facial affect. From relatively early in life, neurotypical infants coordinate these behaviours at above-chance levels (Bates 1976, Crais *et al.* 2009, Stone *et al.* 1997). In infancy, these coordinations may serve an especially important function by increasing the salience and success of bids for joint attention (Goldin-Meadow *et al.* 2007, Martinsen and Smith 1989). Vocalisations are particularly effective in directing the attention of others (e.g., Gros-Louis *et al.* 2006). Communicative behaviours such as gaze shifts, smiles and gestures require that the social partner visually monitor the infant to respond contingently, and thus they may go unnoticed. Vocalisations add an auditory cue that may enhance the likelihood of infants' success in gaining or redirecting adult attention (e.g., Martinsen and Smith 1989). Maybe even more importantly, adding a vocalisation to another behaviour represents a high degree of persistence and directedness on the part of the child. This directed, persistent quality may signal a readiness to learn to social partners. Future work should investigate the extent to which these coordinations contribute to infants' social input from caregivers above and beyond base rates of individual communicative behaviours.

Limitations and future directions

While this study has numerous methodological strengths, including a longitudinal and prospective design, it is important to note some limitations and directions for future work. First, we were unable to include an LR comparison group in this study. As described above, previous research has found differences between LR infants and HR infants, including those who do not go on to develop ASD, in both joint attention and vocalisation production (Cassel *et al.* 2007, Goldberg *et al.* 2005, Yirmiya *et al.* 2006). By virtue of having an older sibling with ASD, HR infants differ from LR infants both genetically and with regard to their home environments. However, the specific impact of having another child with ASD in the home on HR infants' developmental experience is not yet well understood. While

understanding the specificity of development among HR infants with and without language delays and ASD is a significant contribution to the literature, evaluating how HR infants, particularly those without an ASD diagnosis, compare with infants at low risk for ASD will be important to examine in future studies. Second, the sample of infants in this study was predominantly Caucasian and with highly educated parents, and thus external validity may be limited. A replication of this study with a more diverse population would help generalize the results. Finally, given the low production of some behaviours, even at 24 months in the ASD and LD groups, an extension of this study to later time points would clarify whether developmental trajectories in these groups plateau or ultimately follow a similar but delayed course compared with neurotypical peers.

Conclusions and clinical implications

Due to core social communication deficits in ASD (American Psychiatric Association 2013), a central aim of research on infant development in ASD has been to uncover how communicative development differs from neurotypical samples. This effort has revealed differences in the onset and frequency of early communicative behaviours (e.g., Jones *et al.* 2014). The present study extends this work, finding that these behaviours also differ along another dimension: their coordination across modalities. Our findings have two general clinical implications. First, features of communicative behaviours—in particular, the extent to which they were coordinated with one another—distinguished HR infants with ASD from those with non-ASD language delay. This information could bolster efforts to enhance the differential diagnosis of ASD versus language delay. Parents of infants eventually diagnosed with ASD often report that delayed language was the earliest indicator that their infant was developing atypically (e.g., Sacrey *et al.* 2015). Clinically, however, it is difficult to distinguish between ASD versus language delay in very young children (Camarata 2014), yet the ability to make this distinction has important consequences for decisions about the provision and direction of early intervention services. The findings reported above suggest that inclusion of opportunities to observe production of communicative behaviours in coordination in early screening and evaluation efforts is warranted.

Second, many early intervention approaches for very young children with or at risk for ASD focus on increasing infants' communicative behaviours, with a particular emphasis on increasing bouts of joint attention via social gaze (see Bradshaw *et al.* 2015 for a review). While these interventions have demonstrated some success in achieving this goal, it may also be useful to promote the coordination of *multiple* communicative behaviours.

For example, encouraging production of vocalisations together with other communicative behaviours (e.g., eye gaze, gestures) may provide infants with more effective means of conveying communicative messages. Vocalisations combined with other social cues may be more salient to caregivers, enhancing the likelihood of an adult response, and they may also provide additional clues about the focus of the child's attention (e.g., Goldstein *et al.* 2010). In addition, coaching parents to recognize and respond to less salient or less developmentally advanced forms of communication (e.g., give/request gestures) with rich verbal input linked to infants' current focus of attention may provide infants with increased opportunities for communicative exchange. Such an approach could enrich the everyday communicative environment in ways that encourage positive growth in very young children with communication delays with or without ASD. In particular, incorporating these techniques in naturalistic developmental behavioural interventions, which leverage parent coaching within naturalistic settings (e.g., during a bath-time routine) to enhance early learning for children with ASD (Schreibman *et al.* 2015) may be beneficial. These models often incorporate imitation of joint attention behaviours; however, additional emphasis should focus on encouraging parents to imitate and expand on their child's less salient gestures and vocalisations (e.g., vowel-only vocalisations). Exposing young children to more advanced variations of their own gestures and vocalisations could challenge them to try new and diverse communicative behaviours while enhancing caregiver–child interaction.

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