



Exploring receptive and expressive language components at the age of 36 months in siblings at risk for autism spectrum disorder



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ABSTRACT

Background: Language difficulties are highly prevalent in children with autism spectrum disorder (ASD) as well as in their younger siblings (high-risk (HR) sibs). Children with ASD show substantial heterogeneity in difficulties with different language components, but it remains unknown whether this variability is also present in HR-sibs.

Method: Receptive (RL) and expressive language (EL) were evaluated in siblings of typically developing children (low-risk (LR) sibs, $N = 33$) and HR-sibs ($N = 30$) at 36 months, using the Mullen Scales of Early Learning (MSEL), the Dutch version of the Reynell Developmental Language Scales – 2nd edition (RDLS-2) and spontaneous language analysis (SL). Next, composite scores for receptive and expressive phonology, grammar, semantics and pragmatics were formed. Group comparisons were performed and delays in the different language components were explored.

Results: HR-sibs scored significantly lower than LR-sibs on all standardized measures of RL. For EL, significantly lower scores were only found using the MSEL and not using the RDLS-2 nor using SL. HR-sibs scored significantly lower than LR-sibs for receptive and expressive semantics and receptive grammar. HR-sibs with characteristics of ASD presented with less language difficulties than HR-sibs without characteristics of ASD. The majority of HR-sibs showed a delay in one or more language components but these were not consistently detected by the different measures.

Conclusions: Language delays are highly prevalent in HR-sibs and their representation resembles the language profile of children with ASD. Evaluation of language at the level of phonology, grammar, semantics and pragmatics will detect considerably more children experiencing delayed language than holistic approaches.

1. Introduction

Autism spectrum disorder (ASD) is a lifelong neurodevelopmental disorder which is defined by qualitative impairments in social

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interaction and communication, and repetitive and stereotyped patterns of behaviour, interests and activities (American Psychiatric Association, 2013). In addition to these core features, language difficulties are also highly prevalent in ASD (e.g., Levy et al., 2010; Loucas et al., 2008). Language difficulties are not required for a diagnosis of ASD but a comorbid diagnosis of a language disorder is often seen (American Psychiatric Association, 2013).

1.1. Language development in ASD

Individuals with ASD show substantial variability in their mean receptive and expressive language scores with the majority scoring significantly lower than typically developing individuals (Brignell et al., 2018; Kjelgaard & Tager-Flusberg, 2001; Kwok, Brown, Smyth, & Cardy, 2015). Nevertheless, receptive language can sometimes be more affected than expressive language (Boucher, 2012; Kwok et al., 2015). Furthermore, individuals with ASD show considerable heterogeneity in problems with different language components, such as phonology, grammar (morphology and syntax), semantics and/or pragmatics (Boucher, 2012; Eigsti, de Marchena, Schuh, & Kelley, 2011; Naigles & Tek, 2017; Tager-Flusberg, 2006; Tek, Mesite, Fein, & Naigles, 2014). Despite this variability, a certain profile of language difficulties can be detected that might change with age (Boucher, 2012; Rapin & Dunn, 2003). Preschool children with ASD seem to show marked delays in language development with difficulties in receptive and expressive semantics (Boucher, 2012). These difficulties tend to persist at school age and are generally seen as most reliably and severely affected in children with ASD (Boucher, 2012; Naigles & Tek, 2017). A substantial subgroup of preschool children with ASD also shows difficulties in expressive phonology and grammar (Boucher, 2012; Ellawadi & Weismer, 2015; Rapin & Dunn, 2003; Wittke, Mastergeorge, Ozonoff, Rogers, & Naigles, 2017). At school age, the majority of the children with ASD however shows intact expressive phonology and difficulties with regard to expressive syntax tend to resolve (Boucher, 2012; Eigsti et al., 2011; Kjelgaard & Tager-Flusberg, 2001; Rapin, Dunn, Allen, Stevens, & Fein, 2009; Williams, Botting, & Boucher, 2008). Difficulties in pragmatics appear to be present in most individuals with ASD and they also seem to persist throughout the life span (Eigsti et al., 2011; Ellawadi & Weismer, 2015; Naigles & Tek, 2017).

1.2. Language development in younger siblings of children with ASD

Prospective studies of younger siblings of children with ASD (high-risk siblings; HR-sibs) highlight that, because of the high heritability of the disorder, 10–20% of these siblings are likely to develop ASD (Szatmari et al., 2016). Moreover, a substantial subgroup of siblings who do not fully develop ASD (28%), show a mild expression of the disorder, called the “broader autism phenotype” (BAP; Losh, Childress, Lam, & Piven, 2008; Ozonoff et al., 2014) and/or other developmental problems such as delayed language development (Charman et al., 2017; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). This indicates that only approximately 50–60% of HR-sibs show typical development. Consequently, a prospective longitudinal study following HR-sibs may prove to be a strong research design to uncover language development even before an ASD diagnosis becomes apparent (Drumm & Brian, 2013).

HR-sibs with and without ASD often score significantly lower than siblings of typically developing children (low-risk siblings; LR-sibs) on receptive and expressive language measures between the ages of 6–36 months (e.g., Charman et al., 2017; Garrido, Petrova, Watson, Garcia-Retamero, & Carballo, 2017; Hudry et al., 2014; Landa & Garrett-Mayer, 2006; Messinger et al., 2013; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011). Additionally, their language development during the first three years of life seems to be characterized by a high variability (Brian et al., 2014; Landa, Gross, Stuart, & Bauman, 2012; Longard et al., 2017). At the age of 24 months, HR-sibs without ASD were three to four times more likely to exhibit language delays than LR-sibs but by the age of 36 months they did not seem to show significantly more language delays than LR-sibs (Charman et al., 2017; Marrus et al., 2018). Beyond three years of age, HR-sibs with BAP showed significantly lower scores for receptive and expressive language than LR-sibs but typically developing HR-sibs did not (Ben-Yizhak et al., 2011; Drumm, Bryson, Zwaigenbaum, & Brian, 2015; Gamliel, Yirmiya, Jaffe, Manor, & Sigman, 2009; Miller et al., 2016). In conclusion, both receptive and expressive language of HR-sibs with and without ASD can be characterized by delays but very little is known about these delays at the level of different language components.

No studies thus far looked into *phonology and grammar* in HR-sibs during the first three years of life. In school-aged HR-sibs without ASD, difficulties were reported in receptive phonology but not in expressive grammar (Drumm et al., 2015; Levy & Bar-Yuda, 2011). With regard to *semantics*, contradictory results have been reported. Thus far, research also mainly focused on semantics at the level of vocabulary and not at the level of phrases and sentences. On the one hand, HR-sibs did not seem to score significantly lower than LR-sibs for receptive and expressive vocabulary from 7–38 months of age (Hudry et al., 2014) and between 4 and 6 years of age (Pisula, Ziegart-Sadowska, & Kawa, 2015). On the other hand, delays in single word/sentence comprehension and single word production were identified in HR-sibs with and without ASD by 12–18 months of age (Jones, Gliga, Bedford, Charman, & Johnson, 2014; Lazenby et al., 2016; Mitchell et al., 2006; Toth et al., 2007; Zwaigenbaum et al., 2005). Additionally, receptive and expressive vocabulary development did not only seem to be characterized by significantly slower growth during the first years of life in HR-sibs with language delay (LD) but also in HR-sibs with ASD (Iverson et al., 2018). In addition to semantics, conflicting results were also reported concerning *pragmatics*. Regardless of their diagnostic outcome and age, HR-sibs did not consistently show delays in pragmatic language when measured by standardized tests and/or parent-report (Ben-Yizhak et al., 2011; Bishop, Maybery, Wong, Maley, & Hallmayer, 2006; Drumm et al., 2015; Gillespie-Lynch et al., 2015; Warren et al., 2012). Miller et al. (2015), however, reported that at the age of 36 months HR-sibs had significantly lower parent-rated pragmatic language scores than LR-sibs, with 35% of HR-sibs and 10% of LR-sibs showing difficulties in pragmatic language. This shows that pragmatic language difficulties can already be present in about a third of HR-sibs by the age of 36 months (Miller et al., 2015).

In summary, both children with ASD and HR-sibs can encounter difficulties in receptive and expressive language, mainly with regard to semantics and pragmatics (Boucher, 2012; Eigsti et al., 2011; Jones et al., 2014; Miller et al., 2015; Naigles & Tek, 2017). Difficulties in phonology and grammar only seem to occur in a subgroup of preschool children with ASD (Boucher, 2012). This has however not been studied in young HR-sibs, up till now. The quality of language development of children with ASD before the age of six is highly predictive of adult outcome (e.g., autism symptoms, adaptive behaviour, social outcomes such as employment and friendships) (Magiati, Wei, & Howlin, 2014). This is not surprising since language is essential to understand the world around us and experiences, learn new concepts and organize thoughts, communicate needs and desires, reach academic achievement and occupational performance, and to mediate interactions between individuals in order to build social relationships (Drumm & Brian, 2013). Difficulties in different language components will influence different aspects in life for which language is necessary. Consequently, it is not only important to look into general receptive and expressive language abilities in HR-sibs but also at the level of phonology, grammar, semantics and pragmatics.

1.3. The current study

The aim of the present study was to evaluate language in LR-sibs and HR-sibs at the age of 36 months. Receptive and expressive language was evaluated in general and with regard to the different components: phonology, grammar, semantics and pragmatics.

First, we aimed to evaluate whether group differences in receptive and expressive language were present at 36 months when measured by two instruments often used for language assessment: a developmental test (MSEL; Mullen, 1995) and a developmental language test (Dutch version of the *Reynell Developmental Language Scales – 2nd edition*; Schaerlaekens, Zink, & Van Ommeslaeghe, 2003). Additionally, group differences were also explored using spontaneous language analysis (during parent-child interaction) in order to assess expressive language in a more naturalistic setting. Based on previous research we expected that HR-sibs would show significantly lower scores for both receptive and expressive language as measured by standardized tests and spontaneous language analysis (Gamliel, Yirmiya, & Sigman, 2007; Messinger et al., 2015; Miller et al., 2015; Ozonoff et al., 2014; Yirmiya, Gamliel, Shaked, & Sigman, 2007).

Second, we aimed to determine if group differences and possible delays were also present in receptive and expressive language components. Expectations were mainly based on research looking into different language components in children with ASD and to some extent in HR-sibs. We expected group differences to be more pronounced in receptive language in general and with regard to receptive and expressive semantics, as often seen in children with ASD (Boucher, 2012; Hudry et al., 2014; Jones et al., 2014; Naigles & Tek, 2017). A subgroup of HR-sibs might also show phonological and grammatical delays in expressive language at 36 months as these are also apparent in some preschool children with ASD (Boucher, 2012). Delays in pragmatic language might also be present in a subgroup of HR-sibs but they do not seem to be as often present in HR-sibs as in children with ASD (Eigsti et al., 2011; Miller et al., 2015; Naigles & Tek, 2017). Additionally, we mainly expected to see these difficulties in different language components in HR-sibs with characteristics of ASD rather than in HR-sibs without characteristics of ASD.

2. Method

2.1. Participants

Participants were 30 younger siblings (or in 1 case, half-sibling) of children with ASD (HR-sibs) and 33 younger siblings of typically developing children (LR-sibs). The current study was part of a longitudinal prospective study of HR-sibs and LR-sibs during the first three years of life, with assessments at the ages of 5, 10, 14, 24 and 36 months. The protocol was approved by the ethical board of the Faculty of Psychology and Educational Sciences of Ghent University. Written informed consent was given by one of the parents at every assessment. Inclusion criteria for LR-sibs were full-term birth and no ASD within first-degree relatives. HR-sibs and their older sibling with ASD had no known genetic condition linked to ASD. Both LR-sibs and HR-sibs had at least one parent who was fluent in speaking and writing Dutch. HR-sibs were recruited through centres for developmental disorders, rehabilitation centres, home guidance centres and parent support groups. LR-sibs were recruited through well-baby clinics and day-care centres. In addition, both HR-sibs and LR-sibs were recruited through Facebook and the website of the aforementioned longitudinal study.

2.2. Procedure

For the current study, receptive and expressive language was assessed at the age of 36 months. These assessments included on one hand, the administration of two standardized tests often used for language assessment in ASD and in HR-sibs: the *Mullen Scales of Early Learning* (MSEL; Mullen, 1995) and the *Dutch version of the Reynell Developmental Language Scales – 2nd edition* (RDLS-2; Schaerlaekens et al., 2003). On the other hand, expressive language was also evaluated using *spontaneous language analysis* during parent-child interaction.

Following the assessment at 36 months, the diagnostic status of HR-sibs was determined using the Autism Diagnostic Observation Schedule – 2nd edition (ADOS-2; Lord et al., 2012) administered at 14, 24 and 36 months, the Autism Diagnostic Interview - Revised (Lord, Rutter, & Le Couteur, 1994) administered at 36 months and all other available information from all visits, combined with expert clinical judgement. Five children qualified for the diagnostic criteria of ASD according to DSM-5 (APA, 2013) at the age of 36 months, two of whom were also developmentally delayed (DD). Eleven children were categorized as BAP. One of these children with BAP also had LD. Children categorized as BAP presented with subclinical characteristics of ASD which was defined as presenting with

impairments on at least two elements of the first two diagnostic criteria for ASD according to the DSM-5 (APA, 2013; social communication and interaction, A domain; repetitive and restricted behaviour, B domain). In the case of five children, a categorization of atypical development was made when there was for example only LD or DD without difficulties related to the diagnostic criteria of ASD. Finally, eight children were evaluated as typically developing. In LR-sibs, all children showed typical development with the exception of one child that presented with high scores on the ADOS-2 at different ages (LR 15, see Table 7). This LR-sib was excluded from further analyses when characteristics of ASD were taken into account.

2.3. Measures

2.3.1. Mullen scales of early learning (MSEL)

The MSEL (Mullen, 1995) is a standardized developmental test measuring gross and fine motor skills, visual perception and receptive and expressive language in infants and preschool children (0–68 months). Overall cognitive ability is presented by the Early Learning Composite (ELC), a standard score derived from the *T* scores of the aforementioned cognitive scales, with the exception of gross motor skills. The MSEL has demonstrated good internal consistency and test-retest stability (Mullen, 1995). The scores for receptive and expressive language at 36 months have been used in the current study.

2.3.2. Reynell developmental language scales – second edition (RDLS-2)

The Dutch version of the RDLS-2 (Schaerlaekens et al., 2003) is a standardized language test measuring receptive and expressive language in children (2–5 years). Different levels of receptive language are tested by giving instructions of rising verbal complexity which the child must follow with the use of different tools and toys. Expressive language is assessed with regard to vocabulary, language content and language structure using objects, pictures of objects and situational images. The RDLS-2 showed good psychometric quality in Dutch (Zink, Van Ommeslaeghe, Stroobants, Janssen, & Schaerlaekens, 1993).

2.3.3. Spontaneous language analysis during parent-child interaction (SL-PCI)

Parent and child were observed during 15 min of free play and 2 min of reading in an interactive children's book. In case of multilingual families, the parent who was fluent in speaking and writing Dutch was requested to play with the child. Parents (56 mothers, 2 fathers) were asked to interact with their child as they would normally do at home. Two native Dutch-speaking coders, blind for group status and research hypothesis, watched the parent-child interactions in order to transcribe and evaluate all spontaneous language used by the child. A subsample (20%) was transcribed by both coders in order to determine interrater reliability using intra-class correlation coefficients. Reliability ranged from fair to excellent for the different scales (phonology: ICC = .51; vocabulary: ICC = .48; grammar: ICC = .88; pragmatics: ICC = .74) (Cicchetti, 1994). The evaluation of different language domains was made according to the spoken language benchmarks as described by Tager-Flusberg et al. (2009). Phonology was assessed by determining the percentage of intelligibility. This is the ratio of intelligible utterances over all utterances used during the interaction. The type token ratio (TTR), which is the ratio of the number of different words (types) over the total amount of words (tokens), was calculated to map vocabulary. Grammatical development was assessed by determining the mean length of utterance (MLU) in morphemes for the entire language sample. This consisted of the ratio of the amount of morphemes over the amount of sentences. Lastly, pragmatic language was evaluated by an inventory of the communicative functions (e.g., requesting, commenting, ...) used by the child and the presence of a narrative and/or conversational turn-taking during parent-child interaction (PCI). Parents were, however, not instructed to elicit a narrative or conversational turn-taking, leading to very few children showing this behaviour spontaneously. Therefore, only the number of different communicative functions used by the child during PCI was considered in further analysis of pragmatic language.

2.3.4. Language components

Using the abovementioned measures receptive and expressive language were divided into different components: phonology, grammar (morphology and syntax), semantics and pragmatics. Items measuring more than one language component were added to the main language component the item evaluated. No items were represented in multiple language components. All receptive and expressive language components showed good internal consistency. The associated measurement items and the internal consistency of the different language components are represented in Table 1.

With regard to the MSEL, items measuring precursors of language or preverbal language were not included as the main focus of this paper was on verbal language. This was supported by the fact that all children presented with the maximum score on these items indicating that they achieved all precursors of language and all preverbal language at the age of 36 months. Consequently, items measuring comprehension and production of first words (item 8 for receptive language and item 11 for expressive language) were selected as the starting items for inclusion. Additionally, item 33 of receptive language was excluded since this was the only item measuring receptive phonology. This item showed no variance since all children scored zero. Furthermore, item 21 of expressive language, the only item measuring working memory, was excluded. All items and sections of receptive and expressive language of the RDLS-2 were included and distributed based on the manual indicating the main language component measured by the section (Schaerlaekens et al., 2003). With regard to spontaneous language analysis, measures of phonology and grammar were considered separately. Vocabulary, more specifically TTR, was added to the level of semantics. Lastly, pragmatic language measured during PCI could not be added to the concept of pragmatics as it seemed to measure a different aspect of pragmatic language than the items on the MSEL and RDLS-2.

Table 1
Item distribution and internal consistency (Cronbach's alpha) of receptive and expressive phonology, grammar, semantics and pragmatics.

		MSEL	RDLs-2	SL-PCI	α	
					LR	HR
<i>Receptive language</i>	Phonology	(33)	/	/	/	/
	Grammar	20, 22, 25, 31	7, 10	/	.69	.82
	Semantics	8 - 19, 21, 23, 24, 26 - 30, 32	1, 2, 3, 4, 5, 6, 8, 9	/	.69	.96
	Pragmatics	/	/	/	/	/
<i>Expressive language</i>	Phonology	/	/	% intelligibility	/	/
	Grammar	17, 22, 25, 28	Spontaneous Language	MLU in morphemes	.77	.91
	Semantics	11, 14 - 16, 18 - 20, 23, 24, 26, 27	Vocabulary: objects, pictures and words	TTR	.69	.91
	Pragmatics	12, 13	Language content	Communicative functions ^a	.71	.81

Note. MSEL = Mullen Scales of Early Learning (represents item numbers), RDLs-2 = Dutch version of the Reynell Developmental Language Scales – 2nd edition (represents section number for receptive language), SL-PCI = spontaneous language analysis during parent-child interactions, α = Cronbach's alpha, LR = low-risk siblings, HR = high-risk siblings, TTR = type token ratio, MLU = mean length utterance, ^a = eliminated from final language domain due to insufficient internal consistency.

2.4. Data-analysis

Preliminary analyses revealed a limited amount of outliers in the data (i.e., values higher/lower than the mean \pm 3 times the standard deviation (*SD*)). Since outliers were not considered to be random but characteristic of our sample, outliers were replaced by the highest/lowest value allowed (mean \pm 3SD) rather than deleted. Further, the limited number of missing data in this study (7.9%) was caused by fussiness or crying of the child or because the appointment was cancelled and could not be rescheduled in time (illness of the child, parents too busy). As expected missingness turned out to be completely at random (MCAR; Schafer & Graham, 2002) given Little's test of MCAR versus missingness at random (MAR; Little, 1988) was not significant ($\chi^2(4) = 1.35, p = .85$). Therefore missing data was imputed using Expectation Maximization (EM) and all cases ($N = 63$) were used for all analyses.

Group comparisons were performed with regard to participant characteristics. When the assumption of normality was not met, a Mann-Whitney U test was performed, otherwise a one-way ANOVA was performed. Spearman correlations between the participant characteristics and different language components were explored for LR-sibs and HR-sibs separately.

Parametric group comparisons were not possible with regard to *receptive and expressive language development* due to a lack of normal distribution in our data. Consequently, all group comparisons were performed using the Mann-Whitney U test. First, group comparisons were performed for receptive and expressive language as measured by standardized tests (MSEL and RDLs-2) and expressive language as evaluated during PCI using spontaneous language analysis. Spearman correlations between the different measures were also explored. Second, group comparisons were repeated for the constructed receptive and expressive language components: phonology, grammar, semantics and pragmatics. Additionally, Spearman correlations between the language components and the original language measures were explored. Third, group comparisons were performed taking into account the presence of ASD characteristics in HR-sibs. Consequently, HR-sibs were divided into two groups. HR-sibs with characteristics of ASD (HR-A sibs) consisted of children who classified as having ASD or BAP. HR-sibs without characteristics of ASD (HR-NA sibs) consisted of children who showed (a)typical development but no difficulties related to ASD. Among LR-sibs, one child (LR 15, see Table 7) was excluded when ASD characteristics were taken into account due to high scores on the ADOS-2 (Lord et al., 2012). The Kruskal-Wallis test was performed and post-hoc pairwise analyses were explored when the latter was significant. Fourth, it was explored if delays in receptive and/or expressive phonology, grammar, semantics and pragmatics were present in LR-sibs and HR-sibs. Delays were defined by scoring 1.5 standard deviations below the mean of LR-sibs, or below or on the equivalent of percentile 7. The current study used the LR-sibs as a norm group in measures for which no norms (such as percentile scores) were available.

3. Results

3.1. Participant characteristics

HR-sibs scored significantly lower than LR-sibs with regard to nonverbal abilities ($U = 335.00, p < .05$), which is the sum of the raw scores for visual perception and fine motor skills on the MSEL, and with regard to the ELC of the MSEL ($U = 200.50, p < .001$). Nonverbal abilities were significantly correlated with semantics for both receptive ($r_s = .38, p < .05$) and expressive language ($r_s = .39, p < .05$), yet only in HR-sibs. In addition, only in HR-sibs, nonverbal abilities were also significantly correlated with expressive grammar ($r_s = .42, p < .05$) and pragmatics ($r_s = .63, p < .05$). Nonverbal abilities were not significantly correlated with the different language components in LR-sibs. Correlations between the ELC and the different language components were not conducted as items of the MSEL are represented in both measures. HR-sibs also showed significantly more characteristics of ASD (ADOS-2 social affect (SA): $U = 700.50, p < .01$; ADOS-2 restrictive and repetitive behaviour (RRB): $U = 817.50, p < .001$). ASD characteristics were, however, not significantly correlated with the different components of receptive and expressive language in HR-sibs. In LR-sibs, on the other hand, RRB was negatively correlated with expressive phonology ($r_s = -.49, p < .01$). Lastly, the educational level of the mother was significantly lower in HR-sibs than in LR-sibs ($U = 347.50, p < .05$). The educational level of the

Table 2
Participant characteristics.

	LR-sibs	HR-sibs	
Sex ratio (Male:Female)	19:14	13:17	$\chi^2(1) = 1.28$
	<i>M(SD)</i>		
Family SES	51.11 (7.86)	47.18 (11.52)	$U = 414.50$
Educational level mother	6.33 (.78)	5.83 (.99)	$U = 347.50^*$
Chronological Age			
MSEL	36.70 (.72)	37.50 (2.25)	$F(1, 62) = 3.77$
RDLS-2	36.79 (.80)	37.51 (2.07)	$F(1, 62) = 3.40$
PCI	36.84 (1.57)	36.58 (1.30)	$F(1, 57) = .46$
Nonverbal intelligence	75.94 (5.44)	70.70 (10.55)	$U = 335.00^*$
ELC	120.27 (10.42)	101.43 (21.17)	$U = 200.50^{***}$
ADOS-2			
SA	2.39 (1.32)	3.93 (2.20)	$U = 700.50^{**}$
RRB	3.64 (2.42)	6.73 (2.32)	$U = 817.50^{***}$

Note. LR-sibs = low-risk siblings, HR-sibs = high-risk siblings, M = mean, SD = standard deviation, SES = socio-economic status (Hollingshead, 1975), chronological age is indicated in months, MSEL = Mullen Scales of Early Learning, RDLS-2 = Dutch version of the Reynell Developmental Language Scales – 2nd edition, PCI = parent-child interaction, Nonverbal intelligence = the sum of the scores for visual perception and fine motor skills on the MSEL, ELC = Early Learning Composite of the MSEL, ADOS-2 = Autism Diagnostic Observation Schedule – 2nd edition, SA = Social Affect, RRB = repetitive and restrictive behaviours, * $p < .05$, ** $p < .01$, *** $p < .001$.

mother was not significantly correlated with the different components of receptive and expressive language in LR-sibs and HR-sibs. The other participant characteristics did not differ between both groups. Participant characteristics are presented in Table 2.

3.2. Receptive and expressive language measured with different instruments

With regard to *receptive language*, HR-sibs scored significantly lower than LR-sibs both on the MSEL ($U = 291.50, p < .01, \Delta = 1.47$) and the RDLS-2 ($U = 293.00, p < .01, \Delta = .54$). When looking into *expressive language*, however, mixed results were found. HR-sibs scored significantly lower than LR-sibs on the MSEL ($U = 169.00, p < .001, \Delta = 1.80$) but not on the RDLS-2 ($U = 396.50, p = .18, \Delta = .14$). No significant differences were found between LR-sibs and HR-sibs for phonology ($U = 398.00, p = .18, \Delta = .57$), grammar ($U = 358.00, p = .06, \Delta = .52$), semantics ($U = 564.00, p = .34, \Delta = .44$) and pragmatics ($U = 513.00, p = .80, \Delta = .06$) using spontaneous language analysis during PCI. Correlational analyses showed that receptive and expressive language of both standardized measures (MSEL and RDLS-2) were significantly correlated. Semantics and pragmatics measured using spontaneous language analysis were however not consistently correlated with expressive language on the MSEL and RDLS-2. Descriptive statistics are presented in Table 3 for receptive and expressive language measured by the MSEL and RDLS-2 and using spontaneous language analysis. Spearman correlation coefficients between the different measures are presented in Table 4.

Table 3
Descriptive statistics of the raw scores and group comparisons for receptive and expressive language using standardized tests and spontaneous language analysis during parent-child interaction.

	Receptive language				Expressive language				
		Mean rank (range)	<i>M(SD)</i>	<i>U</i>	Δ	Mean rank (range)	<i>M(SD)</i>	<i>U</i>	Δ
MSEL	LR	38.17 (31.00 – 41.00)	34.61 (2.21)	291.50**	1.47	41.88 (31.00 – 43.00)	37.12 (2.86)	169.00***	1.80
	HR	25.22 (14.00 – 41.00)	31.37 (5.87)			21.13 (14.19 – 42.00)	31.97 (5.78)		
RDLS-2	LR	38.12 (37.00 – 61.00)	49.42 (6.74)	293.00**	.54	34.98 (16.00 – 63.00)	41.51 (12.85)	396.50	.14
	HR	25.27 (2.00 – 54.00)	45.75 (10.22)			28.72 (.00 – 68.00)	39.73 (16.39)		
SL – PCI	Phonology	LR				34.94 (82.81 – 99.55)	91.95 (4.36)	398.00	.57
		HR				28.77 (66.67 – 99.38)	89.46 (7.07)		
	Grammar	LR				36.15 (2.11 – 4.90)	3.49 (.65)	358.00	.52
		HR				27.43 (1.27 – 5.65)	3.15 (.98)		
	Semantics	LR				29.91 (.21 – .67)	.34 (.09)	564.00	.44
		HR				34.30 (.21 – .90)	.38 (.15)		
	Pragmatics	LR				31.45 (4.00 – 9.00)	6.45 (1.17)	513.00	.06
		HR				32.60 (4.00 – 9.00)	6.52 (1.06)		

Note. MSEL = Mullen Scales of Early Learning, RDLS-2 = Dutch version of the Reynell Developmental Language Scales – 2nd edition, SL-PCI = spontaneous language analysis during parent-child interactions, LR- sibs = low-risk siblings, HR-sibs = high-risk siblings, *M(SD)* = mean (standard deviation), Δ = Glass' Δ , ** $p < .01$, *** $p < .001$.

Table 4

Spearman correlations for receptive and expressive language using standardized tests and spontaneous language analysis during parent-child interaction.

	MSEL	RDLS-2	SL-PCI			
			Phonology	Grammar	Semantics	Pragmatics
Receptive language						
MSEL		.59**				
Grammar	.63**	.89**				
Semantics	.77**	.88**				
Expressive language						
MSEL		.52**	.26*	.45**	-.25*	.07
RDLS-2	.52**		.38**	.43**	-.17	.21
Phonology	.26*	.38**	1.00	.48*	-.26*	.28*
Grammar	.55**	.78**	.38**	.59**	-.36**	.29*
Semantics	.77**	.82**	.31*	.43**	-.18	.21
Pragmatics	.33**	.76**	.26*	.19	.03	.08

Note. MSEL = Mullen Scales of Early Learning, RDLS-2 = Dutch version of the Reynell Developmental Language Scales – 2nd edition, SL-PCI = spontaneous language analysis during parent-child interaction, * $p < .05$, ** $p < .01$.

3.3. Receptive and expressive phonology, grammar, semantics and pragmatics

3.3.1. LR-sibs versus HR-sibs

With regard to *receptive language*, HR-sibs scored significantly lower than LR-sibs for both grammar ($U = 322.50$, $p < .05$, $\Delta = .78$) and semantics ($U = 278.50$, $p < .01$, $\Delta = 1.70$). For *expressive language*, however, HR-sibs only scored significantly lower than LR-sibs with regard to semantics ($U = 318.00$, $p < .05$, $\Delta = 1.16$). No significant differences were found for phonology ($U = 398.00$, $p = .18$, $\Delta = .57$), grammar ($U = 382.00$, $p = .12$, $\Delta = .69$) and pragmatics ($U = 392.00$, $p = .16$, $\Delta = .42$). Correlational analysis showed that both standardized measures (MSEL and RDLS-2) were significantly correlated with all receptive and expressive language components. Expressive phonology and grammar were also significantly correlated with the different measures of spontaneous language analysis but expressive semantics and pragmatics were not. Descriptive statistics are presented in Table 5 for receptive and expressive phonology, grammar, semantics and pragmatics. Spearman correlation coefficients between the different language components and the MSEL, RDLS-2 and spontaneous language analysis are presented in Table 4.

3.3.2. LR-sibs versus HR-sibs with and without ASD characteristics

With regard to *receptive language*, the three groups differed significantly for semantics ($\chi^2(2) = 8.90$, $p < .05$). Post-hoc pairwise analyses revealed that HR-A sibs ($\chi^2(2) = 11.48$, $p < .05$) and HR-NA sibs ($\chi^2(2) = 15.76$, $p < .01$) showed significantly lower semantic abilities than LR-sibs. HR-A sibs and HR-NA sibs did not significantly differ from each other ($\chi^2(2) = -4.28$, $p = .52$). For grammar, only a tendency for a significant difference was seen ($\chi^2(2) = 5.81$, $p = .06$). Regarding *expressive language*, the three groups also significantly differed with regard to semantics ($\chi^2(2) = 7.70$, $p < .05$). Post-hoc pairwise analyses revealed that HR-NA sibs scored significantly lower than LR-sibs ($\chi^2(2) = 16.13$, $p < .01$). HR-A sibs, on the other hand, did not score significantly lower than both groups (LR-sibs: $\chi^2(2) = 7.50$, $p = .17$; HR-NA sibs: $\chi^2(2) = -8.63$, $p = .19$). No significant differences were reported between the three groups regarding phonology ($\chi^2(2) = 3.11$, $p = .21$) and pragmatics ($\chi^2(2) = 2.06$, $p = .36$). Yet again, a tendency for significance was seen for grammar ($\chi^2(2) = 5.01$, $p = .08$). Descriptive statistics are presented in Table 6 for receptive and

Table 5

Descriptive statistics and group comparisons for receptive and expressive phonology, grammar, semantics and pragmatics.

			Mean rank (range)	M(SD)	U	Δ
RL	Grammar	LR	37.24 (7.00 – 22.00)	14.53 (3.62)	322.50*	.78
		HR	26.33 (0.00 – 22.00)	11.70 (5.09)		
	Semantics	LR	38.56 (51.00 – 71.00)	62.25 (4.79)	278.50**	1.70
		HR	24.78 (7.00 – 69.00)	54.10 (14.34)		
EL	Phonology	LR	34.94 (82.81 – 99.55)	91.95 (4.36)	398.00	.57
		HR	28.77 (66.67 – 99.38)	89.46 (7.07)		
	Grammar	LR	35.42 (10.05 – 25.90)	19.12 (3.84)	382.00	.69
		HR	28.23 (2.00 – 27.26)	16.48 (6.93)		
	Semantics	LR	37.36 (31.25 – 49.35)	40.64 (5.55)	318.00*	1.16
		HR	26.10 (2.90 – 50.36)	34.22 (11.05)		
	Pragmatics	LR	35.12 (5.00 – 29.00)	16.94 (5.13)	392.00	.42
		HR	28.57 (1.00 – 27.00)	14.77 (6.41)		

Note. RL = receptive language, EL = expressive language, LR = low-risk siblings, HR = high-risk siblings, M(SD) = mean (standard deviation), Δ = Glass' Δ , * $p < .05$, ** $p < .01$.

Table 6

Descriptive statistics and group comparisons for receptive and expressive phonology, grammar, semantics and pragmatics in LR-sibs and HR-sibs with and without characteristics of ASD.

			Mean rank (range)	M(SD)	χ^2
RL	Grammar	LR	36.80 (7.00 - 22.00)	14.58 (3.66)	5.81°
		HR-NA	24.77 (6.00 - 22.00)	12.00 (4.56)	
		HR-A	26.68 (.00 - 19.00)	11.47 (5.58)	
	Semantics	LR	37.95 (51.00 - 71.00)	62.19 (4.86)	8.90*
		HR-NA	22.19 (42.00 - 65.00)	55.38 (7.81)	
		HR-A	26.47 (7.00 - 69.00)	53.12 (18.02)	
EL	Phonology	LR	34.38 (82.81 - 99.55)	91.92 (4.42)	3.11
		HR-NA	23.92 (78.26 - 96.51)	89.03 (4.98)	
		HR-A	31.88 (66.67 - 99.38)	89.80 (8.46)	
	Grammar	LR	34.97 (10.05 - 25.90)	19.15 (3.90)	5.01°
		HR-NA	21.77 (4.27 - 22.22)	14.83 (5.45)	
		HR-A	32.41 (2.00 - 27.26)	17.75 (7.81)	
	Semantics	LR	36.94 (30.25 - 48.35)	39.68 (5.63)	7.70*
		HR-NA	20.81 (14.29 - 45.34)	31.66 (9.08)	
		HR-A	29.44 (2.90 - 49.36)	34.46 (12.33)	
	Pragmatics	LR	34.56 (5.00 - 29.00)	16.94 (5.21)	2.06
		HR-NA	26.81 (6.00 - 27.00)	14.85 (5.87)	
		HR-A	29.32 (1.00 - 25.00)	14.71 (6.98)	

Note. RL = receptive language, EL = expressive language, LR = low-risk siblings, HR-NA = high-risk siblings without characteristics of ASD, HR-A = high-risk siblings with characteristics of ASD, M(SD) = mean (standard deviation), °p < .10, *p < .05.

expressive phonology, grammar, semantics and pragmatics.

3.4. Detecting delayed language

In both groups delayed language was defined as scoring 1.5 standard deviations below the mean of LR-sibs or below or on the equivalent of percentile 7. The majority of LR-sibs ($N = 20$, ~60%) did not show a delay in different components of receptive and expressive language. In HR-sibs, however, only 37% ($N = 11$) did not show a delay in language. The remaining LR-sibs ($N = 13$, ~40%) and HR-sibs ($N = 19$, ~63%) showed a delay in at least one component of language of which three HR-sibs showed a delay on all receptive and expressive language components. Both HR-sibs with and without ASD characteristics showed delays on different receptive and expressive language components. LR-sibs and HR-sibs showing a delay in at least one language component are discussed below. Individual profiles of these children and the presence of delayed language on the standardized measures and/or spontaneous language analysis are presented in [Tables 7 and 8](#).

3.4.1. Receptive language

A delay in at least one component of receptive language was seen in five LR-sibs (~15%) and fifteen HR-sibs (~50%). When taking ASD characteristics into account, a similar number of HR-sibs with and without ASD characteristics showed delayed language in at least one component of receptive language. Both receptive grammar and semantics were delayed in six HR-sibs, of which five presented with ASD characteristics. Five LR-sibs and nine HR-sibs (of which three presented with ASD characteristics) showed a delay in either receptive grammar or semantics. The MSEL nor the RDLS-2 detected delayed receptive language in LR-sibs. In HR-sibs, three siblings showing delayed receptive language were detected using the MSEL and the RDLS-2. In one additional HR-sib, delayed receptive language was only detected using the MSEL.

3.4.2. Expressive language

A delay in at least one component of expressive language was present in eleven LR-sibs (~33%) and thirteen HR-sibs (~43%). Expressive grammar, semantics and pragmatics were delayed in one LR-sib and five HR-sibs. Three of these HR-sibs presented with ASD characteristics. One additional HR-sib (without ASD characteristics) presented with delays limited to expressive grammar and semantics. Three of these HR-sibs also showed delayed phonology. Additionally, one LR-sib and three HR-sibs (of which one presented with ASD characteristics) showed delays in expressive grammar. The remaining LR-sibs and HR-sibs showed a delay limited to expressive phonology, grammar and/or semantics. The MSEL detected delayed expressive language in four HR-sibs, but in none of the LR-sibs. One LR-sib and seven HR-sibs showed delayed language on the RDLS-2. Spontaneous language analysis during PCI detected six LR-sibs and eight HR-sibs with delayed expressive language. Three HR-sibs showing delayed expressive language were detected by all measures, the other LR-sibs and HR-sibs were detected by either none or maximum two measures.

4. Discussion

The current study was the first one to evaluate language at 36 months in LR-sibs and HR-sibs looking into receptive and expressive

Table 7
Delayed receptive and/or expressive language in LR-sibs.

	Receptive Language				Expressive Language						
	MSEL	RDLS-2	Grammar	Semantics	MSEL	RDLS-2	SL-PCI	Phonology	Grammar	Semantics	Pragmatics
LR 1											
LR 2											
LR 3									x		
LR 4											
LR 5											
LR 6			x				x				
LR 7											
LR 8											
LR 9											
LR 10										x	
LR 11											
LR 12				x			x	x		x	
LR 13							x	x			
LR 14							x			x	
LR 15											
LR 16							x				
LR 17				x		x			x	x	x
LR 18											
LR 19											
LR 20											
LR 21											
LR 22											x
LR 23			x								
LR 24											
LR 25											
LR 26											
LR 27							x	x			
LR 28											
LR 29											
LR 30			x								x
LR 31											x
LR 32											
LR 33							x	x			

Note. LR = low-risk siblings, x = score 1.5 standard deviations below the mean of low-risk siblings or below or on percentile 7, MSEL = Mullen Scales of Early Learning, RDLS-2 = Dutch version of the Reynell Developmental Language Scales – 2nd edition, SL-PCI = spontaneous language analysis during parent-child interaction.

language abilities in general and at the level of their different components (phonology, grammar, semantics and pragmatics). Additionally, this study was the first one to use multiple measures of language development simultaneously, making comparisons between the measures possible.

First, group comparisons for receptive and expressive language in general were made using the MSEL, RDLS-2 and spontaneous language analysis during PCI. As expected, significantly lower scores for HR-sibs on both receptive and expressive language were found using the MSEL (Messinger et al., 2013; Miller et al., 2015; Ozonoff et al., 2014). When using a more comprehensive developmental language test, however, differences were only found for receptive language and not for expressive language. This is in line with the research of Yirmiya et al. (2007) who also did not find significant differences in expressive language at 36 months between LR-sibs and HR-sibs when using a developmental language test (Clinical Evaluation of Language Fundamentals – Preschool; Wiig, Secord, & Semel, 1992). Additionally, significant group differences in expressive language were also not found using spontaneous language analysis. Taken together, the abovementioned results could indicate that HR-sibs show relatively better expressive rather than receptive language as sometimes seen in children with ASD and in contrast to typically developing children (Boucher, 2012; Fenson et al., 1994; Marrus et al., 2018). Despite the consistency with Yirmiya et al. (2007), we did not expect that the differences in expressive language would only be visible on a general developmental test and not when using comprehensive language measures. A possible explanation can be found when looking at the correlations between the different measures and the language components (see Table 4). The MSEL was mainly correlated with semantics, while the RDLS-2 was highly correlated with all language components. This might indicate that the MSEL mainly represents how children score with regard to semantics and to a lesser extent how they score with regard to phonology, grammar and pragmatics. The fact that group differences for the different language components in

Table 8
Delayed receptive and/or expressive language in HR-sibs.

	Receptive Language					Expressive Language						
	DC	MSEL	RDLS-2	Grammar	Semantics	MSEL	RDLS-2	SL-PCI	Phonology	Grammar	Semantics	Pragmatics
HR 1	A											
HR 2	A											
HR 3	A											
HR 4	NA			x						x		
HR 5	NA				x		x	x	x	x	x	x
HR 6	A			x	x							
HR 7	A	x	x	x	x	x	x	x	x	x	x	x
HR 8	A	x	x	x	x	x	x	x	x	x	x	x
HR 9	NA											
HR 10	A											
HR 11	NA	x	x	x	x		x			x		
HR 12	A											
HR 13	A				x		x	x		x	x	x
HR 14	NA							x				
HR 15	A			x								
HR 16	NA											
HR 17	A				x			x	x	x	x	
HR 18	A											
HR 19	NA	x			x	x	x			x	x	
HR 20	NA			x								
HR 21	NA				x				x			
HR 22	NA											
HR 23	A			x	x							
HR 24	NA										x	
HR 25	A											
HR 26	A											
HR 27	NA										x	
HR 28	NA				x	x	x	x		x	x	x
HR 29	A											
HR 30	A			x	x							

Note. HR = high-risk siblings, x = score 1.5 standard deviations below the mean of low-risk siblings or below or on percentile 7, DC = diagnostic classification, NA = no characteristics of autism spectrum disorders, A = (sub) clinical characteristics of autism spectrum disorder, MSEL = Mullen Scales of Early Learning, RDLS-2 = Dutch version of the Reynell Developmental Language Scales – 2nd edition, SL-PCI = spontaneous language analysis during parent-child interaction.

expressive language were only found at the level of semantics supports this hypothesis. The lack of group differences on the RDLS-2 can then be due to similar scores for expressive phonology, grammar and pragmatics in LR-sibs and HR-sibs, possibly masking differences in expressive semantics between both groups. Spontaneous language analysis, on the other hand, does not seem to show high correlations with the different language components. This may indicate that spontaneous language analysis during a 17-min episode of PCI in a research context does not provide sufficient information to detect differences between the two groups. In conclusion, abovementioned results indicate that detecting group differences for receptive and/or expressive language between LR-sibs and HR-sibs might depend on the measurement that is used. Furthermore, children experiencing developmental problems may also encounter specific difficulties (e.g., difficulty maintaining attention, less engagement, difficulties with imitation skills, low motivation, higher frustration, poor compliance,...) during the administration of a test which may interfere with the accurate assessment of their abilities (Akshoomoff, 2006).

Second, receptive and expressive phonology, grammar, semantics and pragmatics of LR-sibs and HR-sibs (with and without characteristics of ASD) were compared using composite scores based on two standardized measures (MSEL and RDLS-2) and spontaneous language analysis. HR-sibs showed significantly lower scores than LR-sibs with regard to receptive grammar and receptive and expressive semantics. No significant differences were found for expressive phonology, grammar and pragmatics. When characteristics of ASD were taken into account, significant differences between the two groups were only reported regarding receptive and expressive semantics.

With regard to *phonology*, no significant differences were found in expressive language. It was expected that a subgroup of HR-sibs might show difficulties in expressive phonology but the amount of LR-sibs and HR-sibs showing delays in expressive phonology was similar. Together with the fact that phonology is still developing at this age, these results suggest that phonological development might be spared in HR-sibs as seen in older children diagnosed with ASD (Boucher, 2012; Eigsti et al., 2011; Schaeerlaekens, 2009).

Significant differences in the *grammatical* development were only seen for receptive language. This is in contrast to our

expectations as difficulties with grammar mainly seemed to be reported in expressive rather than receptive language in individuals with ASD (Boucher, 2012; Wittke et al., 2017). On the other hand, a subgroup of HR-sibs did show delays in expressive grammar (see Table 8) confirming our expectations that delays would not be represented in all HR-sibs as also seen in preschool children with ASD (Boucher, 2012). The development of expressive grammar is also still in an early stage at 36 months which makes it less likely that difficulties will already occur (Schaerlaekens, 2009).

With regard to receptive and expressive *semantics*, the current results suggest that early delays in both vocabulary comprehension and production might persevere beyond the first years of life in HR-sibs (Iverson et al., 2018; Jones et al., 2014; Lazenby et al., 2016; Mitchell et al., 2006; Toth et al., 2007; Zwaigenbaum et al., 2005). In line with our expectations, delays in semantics are not only among the most affected language domains in individuals with ASD but also in HR-sibs (Boucher, 2012; Naigles & Tek, 2017). However, we did not expect to see larger semantic difficulties in HR-sibs without characteristics of ASD compared to HR-sibs with characteristics of ASD. Nevertheless, the current results might suggest that, at the level of semantics, difficulties that are only present in receptive language may rather be seen in HR-sibs with characteristics of ASD while HR-sibs without characteristics of ASD may rather show difficulties in both receptive and expressive language. This could additionally confirm that children with (characteristics of) ASD might show more difficulties with receptive than expressive language (Boucher, 2012; Kwok et al., 2015). Nevertheless, it should be noted that nonverbal abilities (on which both groups differed) were significantly correlated with the semantical development of receptive and expressive language at 36 months in HR-sibs. Although the correlation is small, group differences in receptive and expressive semantics of HR-sibs can also be due to lower nonverbal abilities. As lower nonverbal abilities may be characteristic for HR-sibs, it is not possible to determine if group differences would still be present when there are no differences in nonverbal abilities (Miller & Chapman, 2001). Due to the small sample size it was not possible to match both groups on nonverbal abilities within the current study.

Surprisingly, no significant differences were found between both groups for *pragmatic language*. This is in contrast to some studies showing significant differences in HR-sibs (Ben-Yizhak et al., 2011; Gillespie-Lynch et al., 2015; Miller et al., 2015). Variability in findings regarding pragmatic language may be attributed to methodological issues such as the type of measure that was used (e.g., standardized test versus parent-report), as well as diagnostic variability (e.g., ASD, BAP, LD, developmental delay) within samples of HR-sibs. Measurement limitations are one of the major barriers to research in pragmatic language since very few appropriate measurements of pragmatic language are available for young children (Drumm et al., 2015). The current study used items of standardized tests measuring mainly the ability to narrate a story based on a situational image. Narrating a story is an ability that is still developing at the age of 36 months making it less likely to find differences between both groups. Within the current sample, four LR-sibs and five HR-sibs showed a delay in pragmatic language suggesting that the ability that is being measured is equally difficult for both groups at this age. Additionally, a broad range of behaviours fall within the scope of pragmatic language (Eigsti et al., 2011; Naigles & Tek, 2017) making it more difficult to compare results of a test or parent-report when they both measure a different aspect of pragmatic behaviour. The majority of the studies in HR-sibs thus far used parent-report measuring mainly communicative skills of children (Ben-Yizhak et al., 2011; Bishop et al., 2006; Drumm et al., 2015; Gillespie-Lynch et al., 2015; Miller et al., 2015; Warren et al., 2012).

Third, delays in the different receptive and expressive language components were studied. The majority of LR-sibs did not show delays while the majority of HR-sibs did show a delay in at least one receptive or expressive language component. HR-sibs also had considerably more delays in receptive language than LR-sibs while delays in expressive language were similar between both groups. These results further confirm the hypothesis of lower receptive than expressive language in HR-sibs as seen in some children with ASD (Boucher, 2012; Fenson et al., 1994; Marrus et al., 2018). These differences were not only present in receptive language in general but also in the different components of language. Furthermore, there were more HR-sibs without characteristics of ASD (~69%) that showed a delay in at least one component of receptive and/or expressive language than HR-sibs with characteristics of ASD (~53%). In contrast to the research of Charman et al. (2017) the current results show that HR-sibs without characteristics of ASD do experience more delayed language than LR-sibs at the age of 36 months.

An important strength of the current study was the use of multiple measures for language development which made it possible to compare the different measures and to evaluate language at the level of phonology, grammar, semantics and pragmatics in LR-sibs and HR-sibs. Nevertheless, the current study also has a few limitations that need to be addressed. Due to small sample size, generalizability of the results is limited. Independent replication of the current results is needed before firm conclusions can be drawn regarding the language development of HR-sibs. The current results will only be fairly representative for LR-sibs and HR-sibs as a group. More research with larger samples of HR-sibs looking into different components of receptive and expressive language cross-sectionally and longitudinally is needed. A second limitation, due to small sample size, is that multilingual siblings (three LR-sibs and one HR-sib) were not excluded from the analysis. However, excluding them did not have an effect on the current results. Third, analyses taking into account the diagnostic outcome of HR-sibs were conducted but this led to even smaller sample sizes. Future research should explore difficulties in components of receptive and expressive language in HR-sibs developing ASD and also in HR-sibs with a non-ASD outcome, using larger sample sizes. In addition, multiple comparison groups should be considered as this may control for diverse aspects such as environmental effects (e.g., growing up with a sibling that experiences developmental difficulties) or genetic vulnerability (Pilowsky, Yirmiya, Shalev, & Gross-Tsur, 2003). Furthermore, at 36 months language development is still at an early stage for some language components. It can thus be informative to measure different language components during their emergence and, for example, evaluate if difficulties in language abilities might change throughout development in HR-sibs as they sometimes do in children with ASD (Boucher, 2012; Rapin & Dunn, 2003; Rapin et al., 2009). In addition, future research should take into account that different measures of receptive and expressive language might hold different results (e.g., developmental test versus developmental language assessment and spontaneous language analysis). Within the current study, the analysis of spontaneous

language was too limited to detect delays in expressive language. Comprehensive analyses of spontaneous language might be more promising in order to detect language delay, but it is less likely that they would be used in clinical practice. Lastly, it should be noted that LR-sibs within the current sample rarely showed delays on the standardized language measures and, on average, showed above-average ELC scores (see Table 1). This can indicate that LR-sibs in the current sample are a high-functioning group of siblings. Consequently, there may be an overrepresentation of language delays in the current sample of HR-sibs. When no norms were available, LR-sibs were used as a norm group when defining delays in the different language components. It is thus possible that scoring 1.5 standard deviations below the mean of LR-sibs is still within the typical range as opposed to defining these scores as a delay.

5. Implications

The results of the current study lead to some clinical implications, especially for those siblings experiencing delays on one or more but not all language components. Children presenting with delays in one language component seem to be missed by standardized tests and/or spontaneous language analysis (see Tables 7 and 8). This suggests that there are HR-sibs who might benefit from early intervention but are not detected by the current language measures. Consequently, it should be considered to use multiple measures when evaluating early language in LR-sibs and HR-sibs. Detection of language delays could also be improved by a comprehensive language test that provides scores for the different language components. Future research should look into the development of comprehensive and validated measures of phonology, grammar, semantics and pragmatics which can be used within diagnostic centres as they might better detect children showing difficulties in only one component. Second, we would like to address the added value of measuring different language components in light of early diagnosis and intervention. The current results showed that HR-sibs, as a group, mainly scored significantly lower than LR-sibs with regard to receptive and expressive semantics. The fact that this is also one of the first language domains that develops in children suggests that delays in language of HR-sibs might already be detected at a young age. As mentioned by Naigles and Tek (2017), HR-sibs and children with ASD seem to encounter difficulties with the content (meaning; semantics) rather than the form (phonology and grammar) of language, which implies that a different approach in early interventions is recommended. This means that HR-sibs will probably pick up on formal rules regarding grammatical language development but will need extra support in learning to understand the meaning of language.

Declaration of Competing Interest

The authors have no conflict of interest to declare.

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