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Linguistic and motor profiles in preschool and school-age children with an older sibling with autism spectrum disorder

Dunia GARRIDO^{1,*} D and Gloria CARBALLO²

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Abstract

This study examines receptive-expressive language, gross-fine motor skills, and IQ abilities in 78 children, 43 children with an older sibling with autism spectrum disorder (Sibs-ASD) and 35 children with an older sibling with typical development, ranging from 4 to 11 years of age. Depending on age, both groups were divided in preschool and school groups. The results show that more than 76% of Sibs-ASD performed at least one language and/or motor skill under 25th percentile. Significant differences were described at preschool stage in three aspects: grammatical comprehension, ball skills, and global motor skills. At school age, significant differences were found in two aspects: expressive language, and ball skills. Some differences seem to decrease over time; meanwhile others seem to increase; and others remain stable. Thus, it seems that vulnerability continues in unaffected Sibs-ASD and suggest that this population may benefit from continued screening and monitoring into the preschool and school-age stages.

Keywords: autism spectrum disorder; siblings; language skills; motor skills

Introduction

Despite the fact that traditional literature has not considered both language and motor skills to be connected, several studies have found reciprocal influence between these skills. For instance, motor development may mediate and promote the relationship between other skills, such as cognitive, social, and linguistic skills (Iverson, 2010; Leonard & Hill, 2014; Wilson, Enticott & Rinehart, 2018). According to Iverson (2010), fundamental motor skill development offers a wide range of opportunities that support and build the block of emerging communication skills. Early motor difficulties have been found to adversely impact in other areas of development, as research has suggested that children interact with people and explore the world around them through their motor development (Leonard & Hill, 2014). Thus, it seems that language and motor development are not independent but connected through comparable underlying processes in children with typical development (TD) (Iverson, 2010; Leonard & Hill, 2014).

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¹University of Granada, Faculty of Psychology Granada 18071 Spain

²University of Granada, Faculty of Psychology Granada 18071 Spain

^{*}Corresponding author: E-mail: duniagarridodelaguila@gmail.com; duniag@ugr.es

As children develop more precise gross and fine motor skills (such as walking, object manipulation and the relational development involved in games), they interact socially and linguistically with people around them. If early motor development is altered, language development can in turn be altered in a cascade effect (LeBarton & Landa, 2019). For Bornstein, Hahn and Suwalsky (2013) a "developmental cascade" defines a longitudinal relationship in which one psychological characteristic affects another psychological characteristic that might influence the developmental trajectory.

However, this interaction is not exclusive to TD children but also occurs in those with atypical development (such as children who are late to talk, Sansavini et al., 2019). Therefore, the potential relationship between differences in both motor, and speech and language skills has been addressed in several developmental disorders, such as autism spectrum disorder (ASD) (Gernsbacher et al., 2008), supporting the multisystem vision of this disorder.

ASD is characterized by challenges in social communication, and restricted and repetitive behaviours (APA, 2013). The linguistic profile in children with ASD shows wide heterogeneity among the spectrum, ranging from absence of language or a limited number of words in some children to broad vocabulary with fluent speech, although with pragmatic difficulties, in others (e.g., Hudry et al., 2010). In this sense, language scores both above and below the mean have been found in children with ASD (Gernsbacher, Morson & Grace, 2015). Some authors have pointed out disturbances and delays in both receptive and expressive language in ASD (e.g., Hellendoorn et al., 2015; Hudry et al., 2010), although others have found more difficulties in receptive vs. expressive skills (Hudry et al., 2010). Meanwhile, the meta-analysis conducted by Kwok, Brown, Smyth and Cardy (2015), which examined 74 studies about expressive and receptive language in ASD, concluded that both types of language skills are equally disrupted and that the differences between them are not clinically significant.

Although motor performance is not included as part of the diagnostic criteria for children with ASD, the severity of motor impairment is widespread in this population. In fact, several researchers have included motor skills in their studies with children with ASD and found that these skills are compromised. The literature reports that children with ASD show difficulties and a delayed motor skill development. In fact, some authors support that these differences could be detected early and considered as core symptoms of this disorder (Fournier, Hass, Naik, Lodha & Cauraugh, 2010; Sacrey et al., 2018). Some of these skills (i.e., manual dexterity, static and dynamic balance, and ball skills) are commonly reported for young children with ASD (Iverson et al., 2019; Garrido, Petrova, Watson, Garcia-Retamero & Carballo, 2017 for a review; LeBarton & Iverson, 2013) as well as school-age children and adolescents (Bhat, 2020; Fournier et al., 2010; Hilton, Zhang, Whilte, Klohr & Constantino, 2011; Leonard, et al., 2014). Unfortunately, it seems that both gross and fine motor difficulties may negatively impact language, social communication, and adaptive behaviour (Bedford, Pickles & Lord, 2016; Bhat, Landa & Galloway, 2011; Gernsbacher et al., 2008; Leonard et al., 2014).

Younger siblings of children with ASD

Younger siblings of children with ASD (Sibs-ASD) are at elevated risk for developing some subclinical characteristics that are related to the core features of ASD such as language difficulties or delays (i.e., broader autism phenotype, BAP) (Choi, Leech, Tager-Flusberg & Nelson, 2018; Gangi, Hill, Maqbool, Young & Ozonoff, 2021). Specifically,

previous studies have described BAP being present at a rate from 21% to 30% among Sibs-ASD at preschool age (Charman et al., 2017; Messinger et al., 2013) and from 17% to 41% at school age (Ben-Yizhak et al., 2011; Shephard et al., 2017). Because these BAP might have an impact on Sibs-ASD, it would be interesting to discover what happens with children with an older sibling with ASD due to the fact that they do not have a formal diagnosis, they have special characteristics that could confer risks and challenges (Gangi et al., 2021). In this sense, studies with a focus on Sibs-ASD would support and grow the current body of research that distinguishes among Sibs-ASD with and without developmental difficulties (such as language or motor delay) (Iverson et al., 2018, 2019; Sansavini et al., 2019).

Moreover, including Sibs-ASD is important because it could provide an opportunity to characterize traits of the BAP in childhood. For instance, Sibs-ASD could show developmental delays (including motor and linguistic skills) from the first year of life (Ozonoff et al., 2014). In this study, of those Sibs-ASD who did not develop ASD themselves, 28% showed atypical profiles in language and fine motor skills.

Linguistic profile in Sibs-ASD

Evidence of disturbances and atypical behaviours in social communication and language areas has been found in Sibs-ASD (see Drumm & Brian, 2013; Gamliel, Yirmiya, Jaffe, Manor & Sigman, 2009; Iverson, 2018; Landa, Holman & Garrett-Mayer, 2007; Leonard et al., 2014; Zwaigenbaum et al., 2005). It seems that, although younger siblings of children with ASD do not match diagnostic criteria, they may be at risk of diverse difficulties, such as language delays or subclinical features of ASD if they are compared with children at low risk of having ASD (Gamliel, Yirmiya & Sigman, 2007; Messinger et al., 2013).

At preschool and school age, several studies have discussed these differences in language skills. For instance, Levy and Bar-Yuda (2011) assessed linguistic and cognitive skills in siblings of nonverbal children with ASD from 4 to 9 years old. In this study, Sibs-ASD showed a language (both expressive and receptive) performance significantly worse than Sibs-TD. However, these authors highlighted that cognitive deficits could explain this poor linguistic output.

Few studies with Sibs-ASD have found lower scores in language skills at 7 years old (Gamliel et al., 2009; Shephard et al., 2017). In particular, difficulties were found in pragmatics (Ben-Yizhak et al., 2011), structural language (Gillespie-Lynch et al., 2015), specific linguistic skills (Yirmiya & Ozonoff, 2007), and receptive and expressive language (Landa, Gross, Stuart & Faherty, 2013; Messinger et al., 2013; Miller et al., 2015; Ozonoff et al., 2014) when compared to Sibs-TD.

In school-age children, findings from several studies are heterogeneous and inconsistent (see Miller et al., 2015; Shephard et al., 2017). To illustrate, in a longitudinal study with Sibs-ASD from 4 months to 7 years old, Gamliel et al. (2009) found deficits in expressive and receptive language. Although these difficulties persisted at preschool age, most of them disappeared at 54 months of life, with the exception of expressive skills, which continued to raise concern. Following these authors, some of these linguistic difficulties appeared when children had to face tasks and school demands for the first time.

Conversely, other authors have found no differences in linguistic skills between Sibs-ASD and Sibs-TD (Drumm, Bryson, Zwaigenbaum & Brian, 2015; Gillespie-Lynch et al.,

2015; Hudry et al., 2014; Shephard et al., 2017; Warren et al., 2012). In particular, Pilowsky, Yirmiya, Shalev and Gross-Tsur (2003) found that Sibs-ASD showed even better verbal skills than siblings of children with developmental language disorders (between 6 and 15 years). However, it seems that these language difficulties can only be elicited in young children and may be no longer evident in older children.

Motor profile in Sibs-ASD

Even though there is little scientific evidence, similar to language skills, motor difficulties seem to appear at an early age in younger Sibs-ASD (LeBarton & Landa, 2019; Leonard et al., 2014; Messinger et al., 2013; Ozonoff et al., 2011). Moreover, the literature that addresses this field also shows mixed results. In this sense, some studies showed delayed skills and moderate difficulties but transitional or no differences between Sibs-ASD and other populations (Charman et al., 2017; Iverson, 2018; Messinger et al., 2013).

Several studies have found that although most of Sibs-ASD do not receive a formal diagnosis of ASD, they are at higher risk of showing delays in gesture communication and motor development than children without a family history of ASD. Furthermore, a longitudinal study conducted by Leonard et al. (2014), where they specified motor performance from the ninth month of life, detected that 31.6% of Sibs-ASD at preschool age (i.e., ranging from 5 to 7 years old) showed motor difficulties (i.e., below the 15th percentile in standardized assessments). Thus, this study supports the thesis that motor difficulties persist beyond infancy.

Sibs-ASD show differences in both gross motor skills (which include large muscle movements such as sitting up, crawling, walking, or running) (Gonzalez, Alvarez & Nelson, 2019) and fine motor skills (which include manual movements, handling objects, or drawing) that are involved in ASD (Bhat et al., 2011; Iverson et al., 2019; Landa & Garrett-Mayer, 2006; LeBarton & Iverson, 2013). Unfortunately, limited research has been conducted related to both gross and fine motor skills in Sibs-ASD older than 4 years (see Garrido et al., 2017 for a review).

Similarly, some studies have found that these differences persist in school-age Sibs-ASD (Leonard & Hill, 2014). However, other studies have found that the motor performance of Sibs-ASD is essentially correct (Hilton et al., 2011).

Thus, given the heterogeneity that exists in language and motor development among this at-risk population and the limited previous work comparing preschool and schoolaged children in these skills, our primary aim was to assess whether language and/or motor skills differed between the Sibs-ASD and Sibs-TD groups using standardized assessment and considering two age groups: the preschool and school stages. Specifically, our research questions were as follows:

- Are there significant differences in language and motor skills between Sibs-ASD and Sibs-TD at preschool age?
- 2. Are similar findings observed in preschool vs. school-aged children?

Following previous findings that stated that first-degree relatives of people with ASD often show differences compared to relatives of people with TD across several domains (Leonard et al., 2014; Levy & Bar-Yuda, 2011), we hypothesized that Sibs-ASD would show several differences compared to Sibs-TD at preschool age across language and motor skills. Moreover, we hypothesized that at school, these differences (if any) would

disappear (i.e., both groups of children would show similar patterns of language and motor skills) as other authors found previously (Hilton et al., 2011; Pilowsky et al., 2003).

Methods

Participants

A total of 78 children ranging from 4 to 11 years old (mean age = 7;5 years, SD = 2.46) and their parents (mean age = 36.77, SD = 6.33) were enrolled in this study. From the whole sample of participants, 55% (n = 43) were male, and 45% (n = 35) were female. Specifically, two cohorts of children were included.

The first consisted of 43 children with an older full biological sibling with a formal diagnosis of ASD (Sibs-ASD) under DSM-IV-TR criteria (APA, 2000), and DSM-5 criteria (APA, 2013) that it was confirmed via administration of the ADOS-G (Lord, Rutter, DiLavore & Risi, 2002) or the ADI-R (Le Couteur, Lord & Rutter, 2003). An additional inclusion criterion was not having a previous diagnosis of ASD, significant motor, language, learning disabilities, visual impairment, or a genetic syndrome. In this sense, the Gilliam Autism Rating Scale (GARS; Gilliam, 2004) was administered to exclude a diagnosis of ASD. The second cohort included 35 children with an older sibling with typical development (Sibs-TD) with no previous history of ASD diagnosis in first-degree relatives. As in the group of Sibs-ASD, parents of Sibs-TD completed the GARS scale to exclude a potential diagnosis of ASD. All participants were recruited through local agencies serving families of children with ASD and several schools located in Granada (Spain), were from monolingual Spanish-speaking households, and did not undergo speech or language therapy.

Because we intend to analyse potential differences depending on age, we described two groups according to two stages: preschool (i.e., children from 4;00 to 6;8 years) and school (i.e., children from 7;6 to 11;11 years) periods, which mainly cover these educational stages in Spain. The demographics of the two cohorts were similar (see Table 1). There were no significant differences in children's gender, age, or parent education. However, parents of Sibs-ASD were significantly older than those of Sibs-TD. This is concordant with prior research of parents of children with ASD. Parents provided written informed consent forms prior to participation. Ethical approval was obtained from the Ethics Committee of the University of Granada (Spain).

Measures

This study used a number of tests and questionnaires to measure language skills (i.e., receptive and expressive language skills), motor skills, intelligence, and severity of ASD.

Receptive language

The Peabody Picture Vocabulary Test (PPVT-III; Dunn, Dunn & Arribas, 2006)

This provides an estimation of receptive vocabulary in children older than two years old. This test shows good internal consistency, with Cronbach's alpha scores ranging from 0.80 to 0.99, and a test-retest reliability of 0.94 (Dunn et al., 2006).

Table 1. Demographics of Sibs-ASD and Sibs-TD samples

		Preschool stage				School stage				
	Sibs-ASD (N = 25)		0.20	Sibs-TD (N = 18)		Sibs-ASD (N = 18)		ASD 17)		
	Mean	DT	Mean	DT	Mean	DT	Mean	DT		
Chronological age	5.55	.85	5.24	.98	9.67	1.27	9.96	1.35		
Gender										
Female (%)	12	48%	10	56%	7	39%	6	35%		
Male (%)	13	52%	8	44%	11	61%	11	65%		
Cognitive abilities										
WPPSI/WISC-IV	107.36	9.90	103.44	8.89	108.50	17.00	110.94	9.41		
Severity of ASD										
GARS	57.52	33.02	38.06	27.94	43.45	13.60	33.71	20.44		
Mean age for parents	37.50	6.61	34.29	3.67	39.28	7.59	34.75	5.47		
Parent education										
High school (%)	9	36%	5	28%	8	44%	6	35%		
College (%)	4	16%	5	28%	4	22%	4	24%		
Graduate (%)	12	48%	8	44%	5	28%	7	41%		

The comprehension test of grammatical structures

In Spanish, Test de comprensión de estructuras gramaticales (CEG) (Mendoza, Carballo, Muñoz & Fresneda, 2005), this includes a total of 20 Spanish grammatical structures, which provides qualitative and quantitative evaluation of children from 4 to 12 years. This test shows good internal consistency, with a Cronbach's alpha score of 0.91 (Muñoz, Fresneda, Mendoza & Carballo, 2008).

Expressive language

The Clinical Evaluation of Language Fundamental (CELF-4; Semel, Wiig & Secord, 2006) This evaluates whether a child from 5 to 21 years has a language delay or disorder. In this study, we included the subscale of expressive language (normative mean of 100 and SD of 15), which comprises the following tasks: formulated sentences, recalling sentences, and word classes-expressive. These three domains (i.e., formulated sentences, recalling sentences, and word classes-expressive) show good reliability (Cronbach's alpha scores of 0.88, 0.96, and 0.88, and a test-retest reliability of 0.71, 0.89, and 0.91 respectively) (Semel et al., 2006). For those children under 5, we used the Clinical Evaluation of Language Fundamental-Preschool version (CELF-P-2; Wiig, Secord & Semel, 2004). For the CELF-4, we included the subscale of expressive language that includes the following tasks: word structure, recalling sentences, and expressive vocabulary. Similar to the CELF-4, these three domains (i.e., word structure, recalling sentences, and expressive vocabulary) show good reliability (Cronbach's alpha scores of 0.84, 0.91, and 0.80, and a test-retest reliability of 0.89, 0.96, and 0.76 respectively) (Wiig et al., 2004).

Motor skills

The Movement Assessment Battery for Children-Second Edition (MABC-2; Henderson, Sugden & Barnett, 2007)

This evaluates three modules (i.e., manual dexterity, ball skills, and static and dynamic balance) in children from 3 to 16 years. All modules were used for both age groups. Additionally, this test provides an overall score of total motor skills that shows good reliability (Pearson's correlation coefficient of 0.80). Moreover, the three modules (i.e., manual dexterity, ball skills, and static and dynamic balance) also show good reliability (Pearson's correlation coefficients of 0.77, 0.84, and 0.73, respectively). Additionally, it shows good test-retest reliability, ranging from 0.62 to 0.92 (Henderson et al., 2007).

Intelligence

The Wechsler Intelligence Scale for Children (WISC-IV; Wechsler, 2012)

This test provides a general intelligence quotient (IQ) for children from 6 to 16 years. This scale provides an overall good reliability (internal consistency coefficient of 0.97 and a test-retest reliability of 0.93). For those children under 6, we used the preschool version (the Wechsler Preschool and Primary Scale of Intelligence -Third Edition, WPPSI-III; Wechsler, 2009). This scale also shows good internal consistency (split half reliability: 0.94) and test-retest reliability of 0.92.

Severity of ASD

Moreover, because we were interested in evaluating children without an ASD diagnosis, parents completed the Gilliam Autism Rating Scale (GARS; Gilliam, 2004). This questionnaire is a norm-referenced screening instrument that helps to identify ASD and identifies three aspects related to ASD (i.e., stereotyped behaviours, communication, social interaction, and developmental disturbances) and an overall autism quotient. This autism quotient is frequently used as the severity of ASD traits. A cut-off of 130 and higher indicates that the child is very likely to have autism. This scale shows good internal consistency, with Cronbach's alpha scores ranging from 0.70 to 0.90, and a global test-retest reliability of .88 (Gilliam, 2004).

Analytic approach

All statistical analyses were performed using SPSS statistics software version 25.0. To address our research question regarding the comparison between language and motor skills, we transformed all raw scores into percentiles. Descriptive and Mann–Whitney U tests were run to compare linguistic and motor performance between Sibs-ASD and Sibs-TD matched according to chronological ages. Nonparametric analyses were selected due to the small sample size (i.e., samples contained fewer than 20 participants). Effect size was calculated with Eta Squared (considering .004-.039, .039-.110, and >.110 as small, medium, and large effect sizes respectively, Cohen, 1988). Moreover, in order to adjust the analyses for the severity of ASD, we used ordinal logistic regression, including those variables that showed significant differences between groups. To standardize criteria across all measures (i.e., expressive, and receptive language skills and gross and fine motor

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skills), we defined severe difficulty (scores below the 25^{th} percentile) and profound difficulty (scores below the 15^{th} percentile).

Results

Descriptive statistics (including the mean, standard deviation, and range) for all variables of interest are shown in Table 2 for preschool and school stages. From the whole sample, only 15.38% (N = 12) showed at least one linguistic skill (i.e., receptive and/or receptive language) under the 25^{th} percentile. Of them, 66.67% (N = 8) belonged to the Sibs-ASD group. Even more importantly, from the sample of Sibs-ASD at the preschool stage, 44% (N = 11) and 32% (N = 8) showed scores under the 15^{th} percentile in receptive and expressive language, respectively. At school age, however, only three (16.67%) and 2 (11.11%) Sibs-ASD showed scores under the 15^{th} percentile in receptive and expressive language, respectively.

Regarding motor skills, 43.59% (N = 34) were under the 25^{th} percentile in at least one motor skill (i.e., manual dexterity, ball skills, balance, and/or global score). From those, the majority (73.53%, N = 25) belonged to the Sibs-ASD group. Thus, from the Sibs-ASD sample, our results show that 76.74% (N = 33) of children were under the 25^{th} percentile in at least one of the motor areas. In the same vein, as was previously the case with language skills, from the sample of Sibs-ASD at preschool stage, 52% (N = 13) and 8% (N = 2) showed scores under the 15^{th} percentile in gross and fine motor skills respectively. At school age, however, only five (27.78%) Sibs-ASD showed scores under the 15^{th} percentile in gross motor skills.

Analyses related to group differences in age, gender, and cognitive abilities did not show significant differences between groups at either preschool stage (all with ps>.05), or school age (all with ps>.05).

Preschool stage

The results are shown in Table 3. Specifically, analyses showed significant differences in three of all evaluated aspects: grammatical comprehension, ball skills, and global motor skill (all with ps<.05, and medium and large effect sizes). Ordinal logistic regression analyses showed that autism traits did not account for any of these differences (all with p>.05). In contrast, results did not show differences between groups in expressive language, receptive vocabulary, dexterity or balance. A graphical representation of the results obtained in language and motor skills is shown in Figures 1 and 2.

School stage

The results are shown in Table 4. Specifically, analyses showed significant differences on two of all evaluated aspects: expressive language (i.e., recalling sentences and global scores), and ball skills (all with ps<.05, and large effect sizes). Ordinal logistic regression analyses showed that autism traits did not account for any of these differences (all with p>.05). On the other hand, the results did not show differences between groups in receptive language (neither vocabulary nor grammatical structures), dexterity, balance, or global motor skills. A graphical representation of the results obtained in language and motor skills is shown in Figures 3 and 4.

Table 2. Study variables of both preschool and school stages

			Preschool stage				School stage						
		Sibs-ASD			Sibs-TI)	Sibs-ASD		Sibs-TD				
		Mean	SD	Min-Max	Mean	SD	Min-Max	Mean	SD	Min-Max	Mean	SD	Min-Max
Receptive language	Vocabulary	78.68	25.91	12 - 100	82.17	30.40	12 - 100	73.67	8.03	60 – 85	82.47	17.65	39 - 100
	Grammatical structures	29.96	23.58	1- 70	49.06	34.66	1 – 97	49.83	28.86	5 – 97	45.88	26.53	10 - 90
Expressivelanguage	Word structure	56.58	26.54	4 -100	68.83	21.65	40 - 98	44.44	14.44	25 – 80	51.35	17.63	26 – 80
	Recalling sentences	64.96	23.92	5 –100	73.61	22.79	42 – 100	51.67	15.05	10 - 80	63.65	20.44	25 – 90
	Expressive vocabulary	56.63	29.06	1 -100	72.06	21.20	44 – 99	49.72	18.27	5 – 85	58.65	23.89	10 - 95
	Global	59.42	25.22	5 - 100	71.67	21.90	42 – 99	48.61	12.34	25 – 75	70.39	20.81	26 – 99
Motor skills	Manual dexterity	40.84	29.06	2 – 75	50.72	20.56	10 - 84	66.12	25.72	25 – 91	64.71	21.71	25 – 84
	Ball skills	23.91	23.33	1 - 84	48.11	28.94	5 – 84	33.39	18.72	9 - 84	60.29	17.56	37 – 91
	Balance	51.04	26.33	16 - 91	61.33	19.79	16 – 91	65.89	27.20	5 – 98	64.06	30.84	5 – 95
	Global	34.56	23.30	5 – 75	49.94	22.40	16 – 84	52.61	31.49	5- 98	64.06	30.84	20 – 95

Note: For receptive language, expressive language, and motor skills, the percentile rank is included

Table 3. Analyses of differences between Sibs-ASD and Sibs-TD at the preschool stage.

			Analyses		
		U	р	η²	
Receptive language	Vocabulary	192.50	.408	.002	
	Grammatical structures	144.50*	.046	.091	
Expressive language	Word structure	156.50	.130	.066	
	Recalling senteces	178.00	.333	.031	
	Expressive vocabulary	167.00	.212	.047	
	Global	161.50	.162	.057	
Motor skills	Manual dexterity	181.00	.268	.027	
	Ball skills	116.00*	.006	.167	
	Balance	171.00	.179	.041	
	Global	145.50*	.048	.089	

^{*=} p < .05

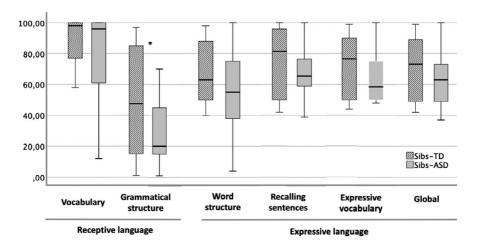


Figure 1. Graphical representation of language skills for both the Sibs-ASD and Sibs-TD groups at the preschool stage. Note: *p<.05.

Discussion

This study extended prior works related to language and motor skills in Sibs-ASD. The primary aim of this work was to assess whether performance in language and motor abilities differed for the Sibs-ASD and Sibs-TD groups in two stages: preschool and school ages. Specifically, in this study, we have analysed those skill deficits that the literature has shown in younger Sibs-ASD (i.e., language and motor skills) in a sample of older children without language delay. Our results support the wide heterogeneity in developmental abilities in Sibs-ASD. Specifically, our results show that those differences in language and motor development not only appear in young children, but also in Sibs-ASD older

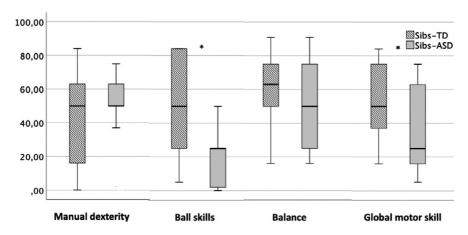


Figure 2. Graphical representation of motor skills for both the Sibs-ASD and Sibs-TD groups at the preschool stage. Note: *p<.05.

Table 4. Analyses of differences between Sibs-ASD and Sibs-TD at the school stage.

			Analyses		
		U	р	η²	
Receptive language	Vocabulary	95.50	.053	.103	
	Grammatical structures	139.00	.643	.006	
Expressive language	Formulated sentences	109.00	.143	.060	
	Recalling sentences	87.50*	.029	.134	
	Word clases-expressive	106.50	.124	.067	
	Global	75.00*	.009	.189	
Motor skills	Manual dexterity	142.50	.727	.003	
	Ball skills	77.00*	.011	.180	
	Balance	122.00	.296	.030	
	Global	150.00	.920	.000	

^{*=} p<.05

children, showing that Sibs-ASD are at increased risk for ASD-related characteristics (Messinger et al., 2013). In fact, more than 76% of Sibs-ASD were under the 25th percentile in at least one language and/or motor skill.

This is especially important, given that the results from this study seem to indicate that even Sibs-ASD without language delay may still present a different profile from Sibs-TD. Furthermore, since both developmental domains are interlinked (Piaget, 1952), so that a minimum disruption to one of them, such as motor skills, could have an impact on other domains (Iverson, 2010; LeBarton & Landa, 2019), including those that apparently are not directly related to, such as language. Thus, this work represents a unique contribution to

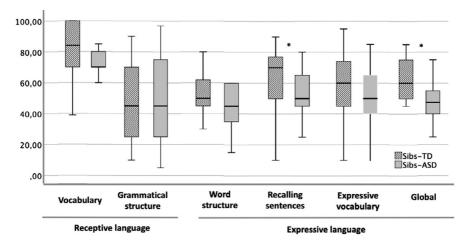


Figure 3. Graphical representation of language skills for both the Sibs-ASD and Sibs-TD groups at the school stage. Note: *p<.05.

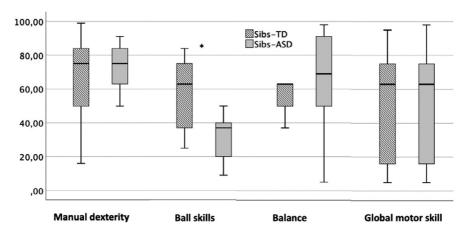


Figure 4. Graphical representation of motor skills for both the Sibs-ASD and Sibs-TD groups at the school stage. Note: *p<.05.

the literature in terms of extending the age and scope of measurement of a previously evaluated high-risk sample.

Preschool stage

Regarding our first research question, we hypothesized that Sibs-ASD would show a range of differences compared to Sibs-TD at preschool age across formal measures of language and motor skills, given that first-degree relatives of people with ASD often show a broad range of differences across several domains. However, the results of our study only partially confirmed our initial hypothesis, since statistical differences were only found in three domains: grammatical comprehension, ball skills, and global motor skill.

The literature shows that children who have an older sibling with an ASD diagnosis show delayed development in language and difficulties in motor skills before 4 years. For example, several longitudinal studies of infant Sibs-ASD have revealed that approximately 28% of high-risk siblings who do not meet the diagnostic criteria for ASD exhibit atypical behavioural patterns in toddlerhood (Ozonoff et al., 2014), are "nontypically developing" at age 3 and before (Girault et al., 2020; see Garrido et al., 2017 for a review), or show features of emergent general developmental delay or speech and language difficulties (Shephard et al., 2017).

Language Profile in Sibs-ASD vs. Sibs-TD

The overall findings of our study indicate that Sibs-ASD showed significant differences in one linguistic skill (i.e., grammatical comprehension) compared to Sibs-TD at the preschool stage (from 4 to 7 years). These results support other studies that have described difficulties in language skills in this population. For instance, it has been found that unaffected Sibs-ASD have an elevated prevalence of language and communication delays compared to low-risk children (Messinger et al., 2013). Some studies have also found lower language skills in Sibs-ASD relative to Sibs-TD. In this line, our results are partially consistent with those from Gamliel et al. (2007) that described differences in language skills (i.e., receptive and expressive skills) in Sibs-ASD by age of 54 months.

In addition, our results partially support results from other studies that did not find difficulties in Sibs-ASD as a group. For instance, Pisula and Ziegart-Sadowska (2015) found that Sibs-ASD (between 4 and 6 years) did not display difficulties in communication and language skills (both receptive and expressive vocabulary). Moreover, Warren et al. (2012) described that global measures of language abilities in Sibs-ASD at 5 years were not different from those in Sibs-TD.

Motor profile in Sibs-ASD vs. Sibs-TD

Regarding the motor profile, the overall findings of our study indicate that Sibs-ASD children showed significant differences in two principal motor aspects (i.e., gross motor, and global motor skills) compared to Sibs-TD children at the preschool stage (from 4 to 7 years). These results are in line with those described by Leonard et al. (2014). These authors assessed Sibs-ASD between 5 and 7 years old with the M-ABC-2 (Henderson et al., 2007), and they detected motor difficulties (i.e., scoring below the 15th percentile on the MABC-2) in almost 32% of children, despite having similar IQ levels to the Sibs-TD group. In a similar vein, our results described that 24% of Sibs-ASD scored below the 15th percentile in global motor skills. However, our results are in contrast with other studies that did not find difficulties in these areas. For instance, Hilton et al. (2011) found that motor proficiency was not impaired in unaffected Sibs-ASD (i.e., only 6% of these children showed motor difficulties). However, as Leonard et al. (2014) stated, it is difficult to compare our results to previous studies when using different tests.

School stage

Regarding our second research question, we hypothesized that both groups of children (i.e., Sibs-ASD and Sibs-TD) would show similar patterns at school age when compared with preschool age in language and motor skills. However, as in our first hypothesis, the

results of our study only partially confirmed it (i.e., similar scores were found between groups, except for expressive language and ball skills). Unfortunately, limited research has been conducted related to motor skills with Sibs-ASD older than 4 years to be comparable to our study.

Language Profile in Sibs-ASD vs. Sibs-TD

In contrast to the preschool stage, in our study, Sibs-ASD showed significant differences from Sibs-TD in expressive language at school stage (from 7 to 11 years). These results are in line with some studies that have found lower expressive language skills in Sibs-ASD relative to Sibs-TD. For instance, Miller et al. (2016) found significantly lower receptive and expressive language scores in the Sibs-ASD group at school age. Additionally, Gamliel et al. (2009) indicated that 40% of Sibs-ASD aged 7 showed linguistic and cognitive difficulties.

Additionally, as we described in our results, Gamliel et al. (2009) stated that most of the group differences disappeared by the age of 4.5 years, with the exception of expressive language skills, which remained an area of difficulty. However, in our study, all children (both Sibs-ASD and Sibs-TD) showed similar cognitive skills, therefore differences in language skills should not be attributed to cognitive abilities.

Similar linguistic patterns to those described by our results (i.e., higher language abilities of Sibs-ASD within normal limits) have been found in the literature. For instance, in several studies, unaffected Sibs-ASD were not found to demonstrate deficits in social communication or language (i.e., receptive, expressive, pragmatics, and phonological processing) at ages from 6 to 16 (Drumm et al., 2015; Gillespie-Lynch et al., 2015; Pilowsky et al., 2003; Shephard et al., 2017). Ben-Yizhak et al. (2011) also reported no differences in some language abilities, finding a similar picture of linguistic skills (i.e., receptive and expressive skills) in Sibs-ASD aged 9 to 12 years.

Motor profile in Sibs-ASD vs. Sibs-TD

Regarding school age (from 7 to 11 years), our study extends the previous literature related to the description of motor development in Sibs-ASD. Similar to the preschool sample, Sibs-ASD showed significant differences from Sibs-TD in ball skills (gross motor) at the school stage. However, the percentage of Sibs-ASD that scored below the 15th percentile in gross motor skills was lower in the school vs. preschool stage (i.e., 28% vs. 52%, respectively). Unfortunately, we have not compared data from other studies, with the exception of the study of Hilton et al. (2011). In this case, they found that only 6% of unaffected Sibs-ASD showed global motor scores of at least one SD below the mean.

Limitations and future directions

Our study has several limitations, and the results should be considered in light of them. Although the current study provides a comprehensive description of linguistic and motor profiles of Sibs-ASD and Sibs-TD, one limitation to these findings stems from our wide inclusion of children between 4 and 11 years of age. Then, when we divided the participants into two younger age groups (i.e., preschool- and school- stage groups), the sample size of both groups decreased. Additionally, this study has been conducted in Spain, so conclusions cannot be drawn to different educational approaches from other

countries. Moreover, as the present study has not a longitudinal design, conclusions regarding development should be considered with caution. Thus, future work would benefit from larger and longitudinal samples of siblings in order to maximize the generalization of these results.

Another study limitation arises from our inclusion criteria. All Sibs-ASD had an older full biological sibling with a formal diagnosis of ASD. However, we did not consider the level of support (i.e., severity) in our analysis. Future studies should examine the impact of autism symptom severity on the development of linguistic and motor skills in Sibs-ASD.

Finally, our data are subjected to the sensitivity of the measures we included in this study. Following this line, future studies should consider additional measures, such as indirect information from parents and teachers. In the case of this study, although linguistic skills were measured with several tests, motor skills were tested with one measure.

Clinical implications

This study has important clinical implications. In particular, it broadens our understanding of the differences existing among those Sibs-ASD who show an apparently typical development. First, the results in language skills highlight the relevance of monitoring language, especially comprehension of grammatical structures during the preschool years, and expressive language during the school stage. Second, in a similar vein, the results in motor skills underscore the importance of including several motor skills, such as ball skills, within the evaluation of Sibs-ASD at both the preschool and school stages. Indeed, as revealed by some other studies involving Sibs-ASD (e.g., Leonard & Hill, 2014), it seems that some difficulties do not disappear over time. Thus, it is essential to monitor these children to control their development and provide their families with assistance if needed.

Conclusion

The main contribution of the current study is that it provides a wide description and comparison of linguistic and motor profiles of Sibs-ASD and Sibs-TD, considering two age ranges (i.e., the preschool and school stages) that are less commonly included in the literature. Our findings provide more comprehensive empirical evidence to previous works suggesting a heterogeneous pattern of developmental skills, where some differences seem to be smaller in the older age groups (e.g., receptive language, and global motor skills), others seem to increase (e.g., expressive language), and others seem to be similar (e.g., ball skills). Thus, our results suggest that vulnerability continues in unaffected Sibs-ASD and suggest that this population may benefit from continued screening and monitoring into the preschool and school-age stages.

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