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Cognitive And Language Development in Older and Younger Siblings of Children With ASD

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COGNITIVE LANGUAGE DEVELOPMENT AND BIRTH ORDER IN NON AFFECTED SIBLINGS OF CHILDREN WITH AUTISM

DESARROLLO COGNITIVO Y DE LENGUAJE EN HERMANOS DE NIÑOS CON AUTISMO

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ABSTRACT

The study aims to explore cognitive and language development in siblings of children with autism, and to compare younger and older siblings on all the measures. 36 siblings of children with autism, between the ages of 2 years, 6 months and 15 years (x=8.67; SD=3.91) were assessed using the PPVT for the receptive language abilities, the WPPSI-III or WISC-III as an estimation of IQ, the VABS as a measure of adaptive functioning, and SCQ was used to assess autism-related symptoms. 72% (n=26) of the children were older than the affected sibling, whereas 28% (n=10) were younger. PPVT scores were below normal limits. On the contrary, IQ scores for the total sample were regarded as average. In contrast, their adaptive functioning was within normal limits. Younger siblings had poorer performance on the PPVT and on the communication sub-domain of the VABS when compared to older siblings. Siblings of children with autism might exhibit abnormal language development, which could be considered as features of the broader-autism phenotype. Although it has also been suggested that birth order could affect the language development of the unaffected siblings, the relationship between these variables remains unclear. These findings offer additional support for a familial association between autism and language abnormalities.

Key Words: autism, siblings, broad autism phenotype, cognitive development, language development, IQ

RESUMEN

Este estudio busco explorar el desarrollo cognitivo y de lenguaje en hermanos de niños con autismo, y comparar hermanos menores o mayores en todas las medidas. 36 hermanos de niños con autismo, de edades comprendidas entre los 2 años 6 meses y los 15 años (x=8.67; DT=3.91) fueron evaluados con el PPVT para las habilidades de lenguaje receptivo, el WPPSI-III o WISC-III como una estimación del CI, las VABS como medida de funcionamiento adaptativo, y el SCQ para medir los síntomas relacionados con autismo. 72% (n=26) de los niños eran mayores que el hermano afectado, mientras el 28% (n=10) eran menores. Las puntuaciones del PPTV estaban por debajo de los limites esperado. Por el contrario, las puntuaciones de CI para la muestra total se consideran promedio. En contraste, su funcionamiento adaptativo estuve dentro de los límites normales. Los hermanos de niño con autismo pudieran exhibir desarrollo del lenguaje anormal, lo cual pudiera ser considerado como características del fenotipo ampliado del autismo. Aun y cuando ha sido sugerido que el orden de nacimiento pudiera afectar el desarrollo del lenguaje de los hermanos no afectados, le relación entre estas variables permanece sin clarificación. Estos hallazgos ofrecen apoyo adicional a la asociación familiar entre autismo y anormalidades en el lenguaje.

Palabras Clave: autismo, hermanos, fenotipo ampliado del autismo, desarrollo cognitivo, desarrollo del lenguaje, CI.

INTRODUCTION

A wide range of cognitive, language, social and behavioral deficits has been described in first degree in relatives of children with autism (Bailey, Palferman, Heavey & LeCouteur, 1998; Constantino, et al., 2006; Pilowsky, Yirmiya, Gross-Tsur & Shalev, 2007). The findings related to a broader range of deficits in relatives of children with autism have led the researchers suggest that the genetic risk associated with autism transcend the classic definition of the disorder; and includes the broad autism phenotype (BAP) that may be expressed by familial transmission in siblings (Bailey et al., 1995).

The BAP involves milder yet similar difficulties in the domains of communication, relationships and restricted interests and repetitive behaviors (Dawson, et al 2007; Gamliel, Yirmiya, Jaffe, Manor & Sigman, 2009; Hurley, Losh, Parlier, Reznick, & Piven, 2007). The genetic vulnerability is thought to extend beyond autism; it might also reach the subtler abnormalities which are included in the BAP. These phenotypic features may be below the diagnostic threshold; however, they may still have an effect on the general development and learning (Stone, McMahon, Yoder & Walden, 2007).

Few published studies have reported on the development profiles of non autistic siblings of children with autism. Cognitive, adaptive, social imitation, play and language abilities were examined in 42 non-autistic siblings and 20 children with no family history of autism. Siblings of children with autism were below in expressive language and social communication skills, and used fewer words, gestures than comparison children (Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). In a different study, language delay, articulation disorder, reading retardation and spelling difficulties were identified in half of the non autistic identical twins

and in 10% of the fraternal twins (Bailey et al., 1995). The reports on the infant siblings of autistic children have described abnormalities in language development and cognitive deficits (Gamliel, Yirmiya, & Sigman, 2007; Stone et al, 2007). However, another group of studies has failed to find impairment in the non affected siblings of children with autism. On that account, Pilowsky, Yirmiya, Shalev, and Gross-Tsur (2003), reported that language abilities of siblings of autistic children did not differ from those of siblings of children with mental retardation. In a different study, Pilowsky et al., (2007) found that neuropsychological functioning in siblings of children with autism emerged as similar to that of siblings of children with mental retardation; and better than the siblings of language delayed children. Thus, there is ambiguity in the reported information about the development of non-autistic siblings.

There has been evidence of a trend for decreasing nonverbal IQ scores with increasing birth order in autism multiplex families (Lord, 1992). In a sample of multiple affected siblings, Spiker et al., (2001), found that second born siblings were more likely to be classified as nonverbal and thus were more language impaired than first born siblings. From this group, less than half of the firstborn children had nonverbal IQs in the mental retardation range, while 70% of the second born siblings fell into that category (Spiker et al., 2001). In a different study, Reichenberg, Smith, Schmeidler, and Silverman (2007) also demonstrated that first born siblings had significantly better useful phrase speech than their younger siblings. Nevertheless, most of the recent studies with siblings of autistic children are being performed with infant siblings, and this fact excludes the possibility to explore the effects of birth order in non-affected siblings.

The review of the literature rends not enough data to conclude about language and cognitive development of the non affected siblings of children with autism, and to identify the effect of birth order in these domains. These studies have been carried out almost exclusively with Caucasian families in the western countries (Orsmond & Seltzer, 2007), with absence of information about the clinical and cognitive performance of siblings of individuals with autism in Hispanic samples. The aims of this study were to explore the cognitive and language development in siblings of children with ASD in a Hispanic sample, and also to compare younger and older siblings on all the measures.

METHOD

Participants

The participants of this study were drawn from a larger sample of single affected families who were participating in an epidemiology study of autism in Maracaibo County. At the moment of the study, 43 non verbal children with autism were part the study. 8 of those children did not have siblings. From the remaining children, there were a total of 52 siblings. However, 14 were outside the age range of the study; and for two children, the parents declined their participation in the research. We had a participation rate of 94.74%.

The final sample included 36 non autistic children, aged between 2 years, 6 months and 15 years. (x=8.67; SD=3.91). 72% (n=26) were older than the affected sibling, whereas 28% (n=10) were younger. Males constituted 55.55% (n=20) of the sample. All the children in the epidemiology study (affected sibling) met ADOS criteria for autism, and had language skills below 30 months (single words speech).

Procedures

The parents of the children participating in the epidemiology of autism project were contacted by phone and asked to participate in the siblings study. Children who were recruited underwent an assessment procedure that included language, cognitive, adaptive skills and autism symptoms measures. All assessments and interviews were conducted by experienced licensed psychologists. Parents and guardians filled out an Informed consent with the help of the psychologist or social worker.

Instruments

The parents were interviewed using the Vineland Behavior Adaptive Scales (VABS) (Sparrow, Balla & Cicchetti, 1984), which is a structured interview with the parent/guardian about adaptive behavior (observed communication, social behavior, and daily living skills at home and in the community). The parent/guardian is questioned closely about specific behaviors. To receive full credit for a skill, the child must engage in the behavior usually and consistently; and not for things a child only does when prompted.

The Social–Communication Questionnaire (SCQ)- Spanish version was used to assess autism-related symptoms (Rutter, Bailey & Lord, 2003). This instrument helps evaluate communication skills and social functioning in children who may have autism or autism spectrum disorders. It is completed by a parent or other primary caregiver. We used the recommended cutoff score of 15 to differentiate controls from possible cases of ASD.

The Peabody Picture Vocabulary Test (PPVT)-Spanish adaptation (Dunn, Padilla, Lugo & Dunn, 1986) was administered to all participants in order to have a measure of receptive language. The PPVT is comprised by 125 cards, and the results allow for a quantitative and qualitative evaluation of the child´s language development.

The IQ was estimated based on the full administration of the WPPSI-III (Wechsler, 1989) for those children aged 2 years 6 months to 6 years 6 months; and WISC-III (Wechsler, 1991) for the children older than 6 years 6 months.

Statistical Analysis

Results were analyzed using SPSS software (version 17). Group comparisons (older vs. younger) were conducted using the Mann-Whitney U, and a significance level of 0.01 was used to reduce the possibility of type I error.

Results

The first step of the analysis was aimed to identify whether any of the children presented symptoms of autism by using the SCQ. From the total sample, only one child scored 14, which is considered as a borderline score for ASD, and represents 2.78% of the sample. Autism

symptoms were virtually absent in the siblings, since the mean score or the SCQ was 2.86 (SD 2.71).
On the subject of language development, PPVT scores were below normal limits (x=73.96; SD=38.49); which indicates that their receptive language skills were within the borderline range. 14% (n=5) obtained scores within the borderline range, while 31% (n=11) scored within the deficient range (<70) (Table 1). These results indicate that 45% of the children exhibited receptive language impairment.

On the contrary, the mean scores for cognitive functioning corresponded to the average classification. All the cognitive indicators, FSIQ (x=91.52, SD=15.98), VIQ (x=94.08, SD=20.81), and PIQ (x=92.06, SD=12.73) were within normal limits(Table 1). Nevertheless, a more detailed analysis showed that for the Full IQ, 25% (n=9) of the children scored within the deficient range, while 3% (n=1) scored within the borderline range. For the VIQ, 14% (n=5) were in the deficient range and 11% (n=) in the borderline. Similar proportions were found for the PIQ, 11% (n=4) of the children obtained deficient scores, and 6% (n=2) borderline scores. These findings indicate that around 28% of the siblings showed signs of cognitive impairment, and VIQ was more affected (25%) than PIQ (17%).
Their adaptive functioning was also within normal limits for all the sub-domains. Communication (x=112.28; SD=20.97) and daily living skills (x=90.42; SD=12.15) were within average range; while socialization domain was above average range (x=125.56; SD=14.97). Only 15% of the sample (n=5) obtained scores below average in the daily living skills domain, 3% (n=1) in the socialization domain, and 11% (n=4) in the communication domain(Table 1).

Table 1

Scores in the different domains

DOMAINS	MIN	MAX	MEAN	SD
AUTISM SYMPTOMATOLOGY (SCQ)	0	14	2.86	2.71
RECEPTIVE LANGUAGE (PEABODY)	55	116	73.96	38.49
VERBAL IQ (WECHSLER)	57	149	94.08	20.81
PERFORMANCE IQ (WECHSLER)	66	114	92.06	12.73
FULL SCALE IQ (WECHSLER)	63	132	91.52	15.98
COMMUNICATION (VABS)	72	173	112.28	20.97
DAILY LIVING SKILLS (VABS)	71	123	90.42	12.15
SOCIALIZATION (VABS)	84	160	125.56	14.97

When compared, younger siblings exhibited poorer performance on the PPVT (x=.57.7; SD=18.54) than older siblings (x=95.46SD=14.26) (U=0; p<0.001). All the children from the younger group (n=10) obtained scores within the deficient range in the PPVT whereas only 1

of the older group obtained similar results. Also, for the VABS communication domain, the younger siblings group scored lower (x=97.04, SD=18.77) than the older group (x=97.04, SD=19.77) (U=58.5; p<0.01). The remaining measures did not show significant differences between the two age groups. However, there was a trend for the daily living skills to be better developed in the younger group (U=79.5; p=0.07) (Table 2)

Table 2

Comparison of younger and older siblings on the different domains

BIRTH ORDER	SCQ		PEABODY		FULL IQ		VERBAL IQ	
	mean	SD	mean	SD	mean	SD	mean	SD
OLDER (N=26)	2.5	1.84	95.46	14.26	92.65	15.45	93.85	19.6
YOUNGER (N=10)	3.8	4.21	57.7	18.54	90	18.02	94.7	24.83
MANN-WHITNEY U	118.5		.000		110.5		116	
SIGNIFICANCE	.689		.000		.497		.639	

BIRTH ORDER	PERFORM. IQ		COMMUNIC.		DAILY LIVING		SOCIALIZATION	
	mean	SD	mean	SD	mean	SD	mean	SD
OLDER (N=26)	92	12.51	118	18.77	88.19	11.95	128.65	9.36
YOUNGER (N=10)	92.2	13.99	97.04	19.77	96.2	11.22	117.5	84
MANN-WHITNEY U	117.5		58.5		79.5		87	
SIGNIFICANCE	.664		.010		.074		.135	

SCQ = Social –Communication Questionnaire
PEABODY= The Peabody Picture Vocabulary Test
FULL IQ = Full Intelligence Quotient
VERBAL IQ = Verbal Intelligence Quotient
PERFORMA. IQ = Performance Intelligence Quotient
COMMUNIC. = Communication Skills
DAILY LIVING = Daily Living Skills
SOCIALIZATION = Socialization Skill

DISCUSSION

Cognitive deficits in siblings of children with autism have been a controversial area. There have been studies that offer evidence to the cognitive impairment in siblings of children with ASD, which could be considered as features of the broader-autism phenotype (Gamliel et al., 2009, Toth et al., 2007). On the other hand, some authors have agreed that the cognitive functioning of the siblings of autistic children does not differ from those of siblings of children with other developmental disabilities (Fombonne, Bolton, Prior, Jordan & Rutter, 1997; Pilowsky et al ., 2003, 2007). In our sample, 28% of the children performed below average in the cognitive measures, which could be considered a moderate evidence of impairment.

Language impairment is the most frequent finding in sibling studies (Toth, et al., 2007; Gamliel, Yirmiya, & Sigman, 2007, Rogers & Sigman, 2005, Stone, et al., 2007). This result was also corroborated in our sample, with 45% of the children exhibiting receptive language impairment, making it the most affected area. Language is one of the most severely affected domains in autism, and language difficulties might be an expression of the BAP or a variable manifestation of a genetic predisposition to autism (Bailey, et al 1995; Stone et al, 2007; Toth et al 2007).

Adaptive skills were within the above-average range. Many siblings of children with ASD might have to assume childcare roles with the affected sibling (Schuntermann, 2007). Therefore, in many ways these siblings frequently reach maturity levels above what is expected for their age, which could be related to scores in socialization and communication.

The risk for sibling recurrence is estimated at around 3% for autism and 6% for ASD, being much higher than the risk in general population (Micali, Chakrabarti & Fombonne, 2004). However, we failed to find autism symptoms in the sample, and only one case met the cutoff score; which is somewhat lower than reports of siblings of children with ASD.

It is also suggested that birth order could affect language and cognitive development of siblings. Although we did not find any difference for cognitive skills, an important aspect of this study was the difference found in receptive language and communication domains between the two age groups (older and younger). For both measures, the younger group exhibited poorer performance than the older group; results that keep with the findings of Spiker et al., (2001), and Reichenberg et al., (2007) which suggest that second-born siblings were more language impaired that first-born siblings. Nevertheless, the reasons behind these differences are still not clear; these subtler difficulties observed in siblings of children with autism could be explained as an expression of the BAP and might have implications for genetic studies in families with an autistic member.

The results of this study must be understood in the light of some limitations. We included siblings of children with severe cases of autism (non-verbal children and receiving special education), and we did not have a control group; these are both factors that limit the generalization of the results. Also comparing younger and older non-affected siblings provides more information regarding the differential expression of the BAP.

In summary, we found that as a group, non-affected siblings have impairment in receptive language but not in cognitive functioning or adaptive skills, which is consistent with most of the previous studies. When comparing younger and older children, impairment in language was mainly for the younger group. These findings offer additional support for a familial association between autism and language abnormalities. This study constitutes a good starting point not only for autism symptoms and development delay recognition in siblings of children with autism in Venezuela, but also for the design and development of adequate diagnostic and therapeutic services for these children.

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