Critical Review:
What are the language abilities of preschool siblings of children with Autism Spectrum Disorder?

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This critical review examines the language abilities of preschool siblings of children with Autism Spectrum Disorder. Studies evaluated include between and mixed between and within group nonrandomized clinical trails. Overall, the research findings suggest that preschool siblings of children with ASD have lower receptive language abilities and comparable expressive language abilities compared to children with no family history of ASD. Recommendations and clinical implications are also discussed.

Introduction

Autism Spectrum Disorder (ASD) is a neurologically based disorder resulting in problems in: communication, social interaction, and unusual patterns of behaviour, activities and interests (Autism Society of Canada, 2010). It is a growing area of service in Speech-Language Pathology. So much so, that in 2006 the American Speech-Language-Hearing Association (ASHA) developed four documents outlining the role of the Speech-Language Pathologist (SLP) in working with children with ASD. This document asserts that SLPs play a pivotal role in improving the early detection of social communication problems in children with ASD because the first evident symptom is often the lack of language or communication attempts.

Autism Spectrum Disorder is not typically diagnosed until the late preschool years, however, marked abnormities in eye contact, lack of social interaction and responses to parents bids for attention can be seen within the first year of life. Speech/Language delay is more evident in the second year of life (Zwaigenbaum et al., 2005). In the past, children with ASD have had poor prognosis, however, recent research has suggested better outcomes with early intervention (Dawson, Greenson, Meltzoff, & Toth, 2007). After receiving a diagnosis of ASD many parents wonder about the recurrence of ASD in subsequent children. Over the past 20 years researchers have examined the existence of the 'broad phenotype of autism' in relatives (parents and siblings) that is a milder impairment in one or more of the three core impairments in ASD (Yirmiya et al., 2006). Critically evaluating the literature investigating preschool siblings of children with ASD could potentially assist SLPs to identify early development of and indicators for ASD and conditions related to the broader phenotype of autism specifically in the areas of early communication and language abilities. Increased awareness of these difficulties could potentially help alert parents and prompt as well as guide early language intervention.

Objectives

The primary objective of this paper was to evaluate the existing research regarding the language abilities of preschool siblings of children with ASD. The second objective was to provide recommendations for clinicians assessing and providing treatment to siblings of children with ASD.

Methods

Search Strategy

Computerized databases, including Scopus, CINAHL, PubMed, and Psychinfo, were searched using the following search terms:
((language) OR (language abilities) AND (communication) OR (communication abilities) AND (verbal) AND (autism) OR (autism spectrum disorder) OR (autistic) OR (ASD) AND (sibling) OR (siblings))

Selection Criteria

Studies collected for inclusion in this critical review were required to examine the language abilities of siblings of children with ASD, aged zero to five years. Diagnosis of ASD in an older sibling was required.

Data Collection

The literature search yielded the following six studies consistent with the selection criteria: Three between groups nonrandomized/non randomized clinical trials and three longitudinal mixed between and within groups non randomized clinical trials. The intention of the present review was to focus on the language abilities of siblings of children with ASD. Results are organized in chronological order.

Results

Zwaigenbaum et al. (2005) initiated a longitudinal study of high-risk infants all of whom had an older sibling with ASD. The authors' original sample included 150 infant siblings of children with ASD, however, only the 65 who were followed to age 24 months were the focus of this study. These siblings were identified through families receiving services at the McMaster Children's

Hospital, the Hospital for Sick Children, and the IWK Heath Centre. A low risk comparison group of 23 infants with no 1st or 2nd degree relatives with ASD were followed to age 2 and were recruited from nurseries across the 3 aforementioned centres. This low risk comparison group was matched for gender, birth order, and age. Study measures included: early behavioural indicators of autism, visual orientating task, infant temperament, assessment of diagnostic outcome and this paper's topic of interest, language development.

Language development was assessed using *The Mullen Scales of Early Learning-AGS Edition* and the *MacArthur Communicative Development Inventories-Words and Gestures (CDI-WG).*

At 36 months of age all participants were seen by an experienced clinician for a diagnostic evaluation using the DSM-IV criteria for ASD. The clinician was blinded to prospective study data and each child's sibling grouping, adding experimental rigor. The use of a blinded clinician controlled for any potential expectations or biases in their assessment of ASD in the study participants. At this point infant siblings were divided into groups based on this evaluation. Groups included: those classified as having ASD (receiving a clinical diagnosis), those siblings who exceeded the threshold for ASD, and non-ASD siblings.

The study provided level 2b experimental evidence as it contained a non-randomized clinical trial and between group comparisons. An analysis of variance (ANOVA) was conducted to compare the expressive and receptive language of the 4 groups: siblings with ASD, high risk group, non-ASD siblings, and controls. Results, using *Mullen* data, revealed that at 12 months of age those who went on to have an ASD diagnoses had lower mean scores on expressive language compared to both other sibling groups and controls, however, this only approached statistical significance. A significant difference ($p=0.007$) was found for receptive language with siblings with ASD having lower scores at 12 months, indicating an overall group effect and a significant difference between subgroups on post-hoc testing using Tukey LSD. On the *CDI-WG* those siblings who were later diagnosed with ASD had fewer gestures and fewer understood phrases. An overall group effect and significant difference between subgroups on post-hoc testing using Tukey LSD were found.

The longitudinal nature of this study was a principle strength as this allowed for direct observation of early behavioural manifestations of the ASD and its developmental sequence. Also, siblings in this study could be followed from a very young age allowing inspection of early neurodevelopmental systems. This has an advantage over retrospective studies as they are subject to recall biases and are likely to include inaccurate parental reports.

Some language measures were incomplete due to some measures being initiated after the study was begun and incomplete parent questionnaire data. Therefore, although the authors stated that the study contained 65 infant siblings and 23 controls, in fact the language abilities of only 40 (*The Mullens*) and 35 (*CDI-WG*) infant siblings, and 12 (*The Mullens*) and 15 (*CDI-WG*) controls were examined. Due to the small sample size and unequal groups, additional follow up and evaluation of other high risk samples is needed to better assess the sensitivity and specificity of these findings, a limitation stated by the authors.

Yirmiya et al. (2006) conducted a longitudinal non-randomized clinical trial to examine whether the development of siblings of children with autism (SIBS-A) was delayed or deviant as compared to siblings of children with typical development (SIBS-TD). Siblings were assessed at ages 4 and 14 months, which was the focus of this paper. It should be noted that these siblings were also assessed again at 24 months, 36 months, and 54 months, using various other procedures, which were published in journal articles to be discussed further in this critical review.

At 4 months of age the SIBS-A group consisted of 21 dyads of mothers and at 14 months was comprised of 30 toddlers. The comparison group also contained 21 dyads at 4 months and 31 dyads at 14 months. These SIBS-TD were recruited from maternity wards of the Hadassa Hospital in Jerusalem. To be included in the comparison group the older siblings must have exhibited typical development. At both ages, groups were matched as closely as possible for age, gender, birth order, number of children in the family, and sex of sibling.

At 4 and 14 months the *Bayley Scales of Infant Developmental – 2nd edition (BSID-II)* was used to assess general development and language. At this point in time this study was the only one known to the authors to assess the development of SIBS-A at 4 months of age. It was predicted that at 14 months of age SIBS-A would display deficits in communication and general cognitive abilities as assessment by the *BSID-II*.

Preliminary t-tests and chi-squares revealed no significant sex differences on any independent or dependent variable, therefore, analysis were completed independent of sex. At 14 months significant ($p=0.025$) between group differences were seen on the *Bayley* language score, with SIBS-A having a significantly

lower developmental age then SIBS-TD. The authors then examined the individual scores of siblings in each group, a strength of the study. Among the SIBS-A group 8 had a language developmental age lower than 14 months, 6 had a language developmental age of 9 months, and 2 achieved a language developmental age of 12 and 13 months. In comparison only 3 of the SIBS-TD achieved a language developmental age lower than 14 months, all 3 being a delay of only 1 month. Given the above analysis the authors then re-grouped the SIBS-A into the 6 siblings having a delay of 5 months (SIBS-A-LD) and the 24 remaining siblings with normal language levels (SIBS-A-nonLD). An ANOVA was well selected as 3 means were compared (SIBS-A-LD, SIBS-A-nonLD, and SIBS-TD), thereby decreasing the chances of a type I error. Analyses revealed that the SIBS-A-LD had significantly lower *Bayley* language age score compared to the SIBS-A-nonLD and SIBS-TD both of which did not differ from one another. A Scheffe analyses followed ($p<.001$) to correct for multiple comparisons.

In summary, at 14 months, SIBS-A achieved lower language scores on the *Bayley Scales* and initiated fewer nonverbal gestures. Of the 31 SIBS-A 6 had a language delay of 5 months and were responsible for the differences between SIBS-A and SIBS-LD.

The study was relatively well designed with every effort possible employed to insure homogeneity of sibling groups from a very young age. However, sample sizes were relatively small, and the study lacked an additional comparison group to account for a sibling with special needs. Such a variable could account for growing up with a sibling requiring disproportionally more of parents' attention thereby decreasing opportunities for language development.

Yirmiya et al. (2007) continued their investigations of the cognitive and linguistic abilities of the same SIBS-A (n-30) and SIBS-TD (n=30) at 24 and 36 months of age. These two points in time were felt to be important as they inform understanding about typical acquisition of language skills and early diagnosis of ASD targets children around age 24 months. They hypothesized that more SIBS-A would display language and cognitive difficulties.

At age 24 months the *Reynell Developmental Language Scales (RDLS)* was used to provide assessments of expressive and receptive language. At 36 months of age the *Clinical Evaluation of Language Fundamentals-Preschool (CELF-Preschool)* was used as it provides scaled scores for receptive and expressive language and a total scaled score for each subtest.

Families were contacted 1 week before their child reached 24 and 36 months. An examiner tested the children using the *CELF-Preschool*, however, no mention was made of any blinding procedure, a possible source of experimenter bias.

T-tests were completed to compare differences between groups at age 24 and 36 months. No significant group differences were found on the *RDLS* subscales for receptive or expressive language. However, when examining the individual trajectories for siblings in each group, significantly more SIBS-A received a scaled score below 85 on the receptive scale. A significant difference was also found between the number of siblings in each group who scored below 70 on the receptive scale with 6 SIBS-A falling in this range compared to 0 SIBS-TD. Significant differences emerged for the total *RDLS* score (receptive and expressive) with more SIBS-A falling 1 or 2 standard deviations (SD) below the mean. At 36 months significant group differences were seen on the *CELF-Preschool* on the receptive scale ($p<.025$). Furthermore, SIBS-A score significantly lower compared to SIBS-TD on all 3 receptive subtests but not on any of the 3 expressive subtests. In fact, a significant difference occurred with more SIBS-A scoring 1 or 2 SDs below the mean on the receptive scale of the *CELF-Preschool*, this difference was found using Fisher's exact test of analysis. The use of Fishers exact test of analysis was appropriate in this case as sample sizes were small.

The authors further sub-grouped 7 SIBS-A into an at-risk group based on previous and current assessment results. When analyses were repeated with these 7 SIBS-A removed no significant differences ($p>.05$) were observed on any language measure. These finding indicate that at ages 24 and 36 months specific language vulnerabilities exist and can be observed by SLPs.

Once again, this study was relatively well designed with every effort made to insure homogeneity of sibling groups from a very young age. Attrition remained very low with only 1 less participant in the SIBS-TD group, an occurrence not explained by the authors. However, sample sizes were relatively small, *CELF-Preschool* examiners were not blinded to sibling status, and the study lacked an additional comparison group to account for a sibling with special needs.

Based on the methodological issues stated above the results of the study and their ability to affect clinical decisions are considered suggestive.

Gamliel, Yirmiya, and Sigman (2007) continued their examination on the development of cognition and language in the same SIBS-A and SIB-TD samples from

4 to 4.5 years. The SIBS-A and SIBS-TD groups both contained 39 children. Any SIBS-A added to the study after 4 months of age was matched as closely as possible to SIBS-TD according to parents age, ethnicity, income, and level of education.

For analysis the authors grouped siblings who received scores at least 2 SDs below the mean on language measures. At 14 months 5 siblings were identified as showing signs of the broad phenotype, known as SIBS-A-BP14, and 6 were identified at 24 months known as SIBS-A-BP24. The remaining SIBS-A who did not reveal language delays was termed SIBS-A-nonBP (n=27), and the SIBS-TD (n=37).

Developmental trajectories were compared using analysis of variance for the 4 groups at 14, 24, 36, and 54 months. Due to small groups, effect sizes were also examined. An effect size of .80 was employed, meaning anything at .80 or larger was considered to be a meaningful group difference. This effect size is considered large as it indicates a non-overlap of almost 50% in the two distributions. Post-hoc LSD analyses were utilized correctly as they were used only when significant differences were found.

At each of the four ages, for both receptive and expressive language, significant differences emerged among the four groups on the language assessments used, with the exception of receptive language at 54 months. No significant differences were found between the groups on the *CELF-P* receptive score.

Results for receptive language at 14 months: SIBS-A-BP14 revealed a significantly lower language score than did the SIBS-A-BP24, SIBS-A-nonBP, and SIBS-TD. Examination of effect sizes showed large group differences between SIBS-A-BP24 and SIBS-A-nonBP, and between SIBS-A-BP24 and SIBS-TD, with lower language scores for SIBS-A-BP24.

Results for receptive language at 24 months: The authors used post-hoc LSD analysis, and as a group SIBS-A-BP14 showed significantly lower receptive scores than SIBS-nonBP and SIBS-TD. This trend was also found for SIBS-A-BP24 as they also had significantly lower receptive scores than SIBS-A-nonBP and SIBS-TD. A large effect size was found when comparing SIBS-A-BP14 and SIBS-A-BP24. SIBS-A-BP24 had lower receptive language scores compared to SIBS-A-BP14.

Results for receptive language at 36 months: Post-hoc LSD tests found SIBS-A-BP14 to have a significantly lower receptive score than SIBS-A-nonBP and SIBS-TD. Again this trend was found for SIBS-A-BP24 as they also had a significantly lower receptive scores than SIBS-A-nonBP and SIBS-TD.

Results for receptive language at 54 months: Effect sizes demonstrated SIBS-A-BP14, SIBS-A-nonBP, and SIBS-TD to have higher language scores than SIBS-A-BP24 at 54 months.

Results for expressive language at 14 months: Expressive language results at 14 months are the same as those for receptive language. This is due to the use of the BSID-II, as it assesses global language as apposed to differentiating receptive verses expressive abilities

Results for expressive language at 24 months: Significant differences and effect sizes regarding expressive language abilities among the four groups were the same as those found at 24 months for receptive language.

Results for expressive language at 36 months: The post-hoc LSD test demonstrated SIBS-A-BP14 to have significantly higher expressive scores on the *CELF-P* than SIBS-A-BP24, and significantly lower scores than SIBS-A-nonBP and SIBS-TD. SIBS-A-BP24 had significantly lower expressive scores than all three groups.

Results for expressive language at 54 months: The post-hoc LSD test demonstrated only the SIBS-A-BP24 group to have significantly lower expressive scores than SIBS-A-nonBP and SIBS-TD. Inspection of effect sizes indicated that SIBS-A-BP14 had higher expressive language scores than SIBS-A-BP at 54 months.

Overall results indicate that some language difficulties were still present at 54 months. Although many of the siblings identified as having difficulties at 14 and 24 months were still experiencing difficulties at 54 months of age, most SIBS-AnonBP did not differ from SIBS-TD in language abilities.

A limitation of the study remains its small sample size. It is still yet to be seen if other, larger studies, will reproduce these results. Throughout these three articles no mention was made as to how SIBS-A were recruited, thereby, introducing a possible extraneous variable. A strength of this study is that it adds to the literature on the development of siblings of children with ASD, identifying earlier markers for and conditions related to the broad phenotype of ASD. Based on the above strengths, methodological issues, and results of Yirmiya et al. and Gamliel et al. studies and their ability to affect clinical decisions are considered suggestive.

Toth et al. (2007) examined the cognitive, social communication, social emotional functioning, imitation, functional symbolic play, and language skills of 18 to 27 month old siblings of children with ASD (n=42) and children with no family history of ASD (n=20). The 2 groups differed by 2 months of age on average; therefore chronological age was entered as a covariate in all analysis. Similar to Zwaigenbaum et al. (2005), the *Mullens Scales of Early Learning* was used to assess language skills. *The Communication and Symbolic Behaviour Scale-Developmental Profile (CSBS-DP)* was used to assess the following areas of interest: sounds, words, and language comprehension.

Analysis of variance was used to examine receptive and expressive language scores on the *Mullens*. A significant group difference (p=.033) was found for receptive language with siblings of children with ASD having lower scores than the comparison group. These groups did not differ on expressive abilities. Multivariate analysis of covariance (MANCOVA) on *CSBS-DP* data revealed significant group differences (p=.02). Siblings obtained lower cluster scores in overall social use of gestures, words, and language comprehension.

A strength of this study was its sample size as it contained the largest group of siblings of children with ASD of all studies reviewed. Additionally, like Zwaigenbaum et al. (2005), they have chosen to evaluate language abilities prospectively as opposed to retrospectively, thereby eliminating potential errors in parents' ability to recall their child's language abilities across ages. Nine of the participants with no family history of ASD did not have an older sibling. Therefore, environmental influence older sibling interactions (ASD or non-ASD) could not be controlled for. Additionally, the clinicians administering the *CSBS-DP* were not blinded to group membership, a possible source of error as experimenter biases may have occurred.

Chuthapisith et al. (2007) also examined the language development of siblings of children with ASD and typically developing children during the preschool period. Twenty-nine siblings of children with ASD were age matched with twenty-eight typically developing children who had no siblings with ASD. No significant differences were found in terms of birth order, family size, maternal educational level or family income.

Pattern analysis, copying, quantitative, and bead memory subtests of the *Stanford-Binet IV Intelligence Scale, Fourth Edition (SB:IV)* were used to yield non-verbal IQ. Verbal reasoning area scores were used to estimate verbal IQ. The *SB:IV* was translated into Thai and adapted for use in this study.

The authors state that due to the normal distribution of each group, ANOVA were used to compare IQ scores between groups, followed by post-hoc analysis. No significant differences were found between the two groups on non-verbal IQ scores and mean verbal IQ scores were similar. Inspection of participant's individual data revealed that 7 of the 10 children having the highest test composite scores, greater than in the average range, were in the siblings of ASD group. Six of these siblings were diagnosed with Asperger syndrome.

Due to the narrow demographics of the sample, care should be taken with interpretation and generalization of the findings from this study. The study participants were recruited from a single institution, which tends to service individuals from more advantaged upper class socio-economic groups. Secondly, there exists no standardized instrument in Thailand to confirm ASD diagnoses. Third, the *SB:IV* was translated for use and there are no available norms for Thai children, therefore scores should be compared cautiously to Western research. It should also be argued that the *SB:IV* does not offer a comprehensive assessment of a child's language skills, lacking criterion validity, as compared to well-standardized assessments specific to language such as the *CELF-Preschool*. It can also be assumed that the *SB:IV* may have been utilized with a non-standardized method of administration. Due to the large number of study limitations, these findings must be interpreted as equivocal.

Discussion

Studies reviewed provided a moderate level of evidence. Thus, the critical appraisal of appropriate research material indicates that these studies offer suggestive evidence.

Zaigenbaum et al. (2005) found that those who went on to have an ASD diagnoses had significantly lower receptive language abilities and fewer gestures than siblings of typically developing children. Yirmiya and colleagues (2006) longitudinal study of siblings at ages 4, 14, 24, 36, and 54 months found siblings of children with ASD: achieved lower language scores on the *Bayley Scales* and initiated fewer nonverbal gestures at 14 months; significantly more siblings of children with ASD received a scaled score below 85 on the receptive scale at 24 and 36 months and significant group differences were seen on the *CELF-Preschool* on the receptive scale. Although many of the siblings identified as having difficulties at 14 and 24 months were still experiencing difficulties at 54 months of age, most siblings of children with ASD did not differ from siblings of typically developing children in language abilities. Toth et al. (2007) found a significant group difference for receptive language on *The Mullens* with

siblings of children with ASD having lower scores. However, groups did not differ on expressive abilities. Chuthapisith et al. (2007) found that 7 of the 10 children having the highest test verbal IQ scores were in the siblings of ASD group. However, these results are regarded as equivocal due to multiple shortcomings of the study.

Preschool siblings of children with ASD were shown to have lower receptive language abilities and comparable expressive language abilities when compared to children with no family history of ASD. Several methodological shortcomings reduce the credibility of this conclusion. Several studies included small sample sizes and unequal groups. Additionally, due to the narrow demographics of the samples, care should be taken with interpretation and generalization of the findings. Among the studies, participants were recruited from a single institution, or as in Yirmiya and colleagues (2005) longitudinal studies, no mention was made as to how siblings were recruited, thereby, introducing a possible extraneous variable.

Recommendations and Clinical Implications

The current evidence suggests that specific language vulnerabilities exist and can be observed by SLPs as young 12 months of age. Recent research has suggested better outcomes with early intervention.

Based on the limitations of the current research discussed above, it is recommended that further research be done to investigate the language abilities of preschool siblings of children with ASD and should include the following:

1) Inclusion of a comparison group of siblings of children with other developmental disorders to determine if early language characteristics are specific to those families with a history of ASD or more broadly related to families with children with special needs.

2) More longitudinal studies should be completed to follow young siblings development and document how early language abilities change over time.

3) Demographic characteristics for future studies should more closely represent the general population with larger sample sizes.

4) Blinding procedures should be utilized for assessment of siblings to eliminate potential bias.

Future longitudinal research may provide increased understanding of the 'broad phenotype of autism', neuodevelopmental and language development in ASD thereby, leading to earlier identification and treatment. Based on the current evidence, clinicians should be mindful of the broad phenotype of ASD in their assessment and treatment of siblings of children with ASD. While some siblings with delayed expressive and receptive abilities may catch up others with difficulty in these areas others continue to struggle. Based on evaluation of the above research, clinicians may choose to closely monitor siblings of children with ASD with appropriate intervention implemented as needed.

References

American Speech-Language-Hearing Association. (2006). Principles for Speech- Language Pathologists in Diagnosis, Assessment, and Treatment of Autism Spectrum Disorders Across the Life Span.

Autism Society of Canada (n.d.). *What is Autism Spectrum Disorder?* Retrived December 10, 2010,from http://www.autismsocietycanada.ca/pdf_word/info_ASC'swhatisautisminfosheet_27_June_07_e.pdf

Chuthapisith, J., Ruangdaraganon, N., Sombuntham, T., & Roongpraiwan, R. (2007). *Language development among the siblings of children with autistic spectrum disorder. Autism, 11*(2), 149-160.

Gamliel, I., Yirmiya, N., & Sigman, M. (2007). *The development of young siblings of children with autism from 4 to 54 months. Journal of Autism and Developmental Disorders, 37*, 171–183.

Toth, K., Dawson, J., Meltzoff, A. N., Greenson, J., & Fein, D. (2007). *Early social, imitation, play, and language abilities of young non-autistic siblings of children with autism. Journal of Autism and Developmental Disorders, 37,* 145–157.

Yirmiya, N., Gamliel, I., Pilowsky, T., Feldman, R., Baron-Cohen, S., & Sigman,M. (2006). *The development of siblings of children with autism at 4 and 14 months: Social engagement, communication and cognition. Journal of Child Psychology and Psychiatry and Allied Disciplines, 47*, 511–523.

Yirmiya, N., Gamliel, I., Shaked, M., & Sigman, M. (2007). *Cognitive and verbal abilities in 24- to 36-month-old siblings of children with autism. Journal of Autism and Developmental Disorders, 37*, 218–229.

Zwaigenbaum, L., Bryson, S., Rogers, T., Roberts, W., Brian, J., Szatmari, P. (2005). Behavioral manifestations of autism in the first year of life. *International Journal of Developmental Neuroscience, 23* (2-3): 143-152.