The relation between language regression and social communicative development of children with autism spectrum disorder

A relação entre regressão da linguagem e desenvolvimento sociocomunicativo de crianças com transtorno do espectro do autismo

ABSTRACT

Purpose: To investigate the relationship between language regression and the subsequent social-communicative development of preschool children with Autism Spectrum Disorder (ASD).

Methods: Thirty children with ASD participated in the study and were divided into two groups: with (n= 6) and without (n= 24) language regression. Language regression was assessed by the Autism Diagnostic Interview-Revised and the social-communicative development was measured by the Autism Diagnostic Observation Schedule.

Results: Of the 30 children who met the criteria for participation in this study, six (20%) had regression of oral language skills, with a mean age of onset of 25 months. There were no statistical significant differences in the social-communicative development between the groups with and without language regression.

Conclusion: The findings of this research do not seem to confirm the relationship between the occurrence of language regression and the subsequent impairment on social-communicative development of children with ASD.

RESUMO

Objetivo: Investigar a relação entre a ocorrência de regressão da linguagem e o desenvolvimento sociocomunicativo posterior de crianças pré-escolares com Transtorno do Espectro do Autismo (TEA).

Métodos: Participaram do estudo 30 crianças com TEA, divididas em dois grupos: com (n=6) e sem regressão da linguagem (n=24). A regressão da linguagem foi avaliada com base na Autism Diagnostic Interview-Revised e o desenvolvimento sociocomunicativo a partir do Autism Diagnostic Observation Schedule.

Resultados: Das 30 crianças que preencheram os critérios para participação neste estudo, seis (20%) apresentaram regressão das habilidades de linguagem oral, com uma média de idade de 25 meses no início da perda. No que se refere ao desenvolvimento sociocomunicativo, não foram observadas diferenças estatisticamente significativas, entre os grupos com e sem regressão da linguagem.

Conclusão: Os resultados desta pesquisa parecem não confirmar a relação entre a ocorrência de regressão da linguagem e o comprometimento posterior do desenvolvimento sociocomunicativo de crianças com TEA.

Study carried out at the Nucleus of Study and Research in Developmental Disorders (NIEPED), Universidade Federal do Rio Grande do Sul – UFRGS – Porto Alegre (RS), Brazil, with fellowship from Coordenação de Aperfeiçoamento de Pessoal de Nível Superior (CAPES) and from the Fulbright Program.

(1) Post-graduation Program in Psychology, Universidade Federal do Rio Grande do Sul – UFRGS – Porto Alegre (RS), Brazil.

Conflict of interest: nothing to declare.
INTRODUCTION

Autism Spectrum Disorder (ASD) is a neurodevelopmental condition that presents in the first years of a child’s life\(^{(1,2)}\). The term ASD has been used in publications to cover autistic disorders, Asperger’s syndrome, childhood disintegrative disorders and the pervasive developmental disorder not otherwise specified. The etiology of ASD is still unknown, but research has pointed to genetic and neurobiological correlates\(^{(3)}\). Furthermore, epidemiological data show a prevalence of one in 150 births and this has increased in recent decades. This can be explained by the expansion of the diagnostic criteria, the increase of health services related to the disorder and the change in the age of diagnosis, among other factors\(^{(4)}\).

ASD is characterized by the presence of social-communicative impairments and restricted and repetitive behavior patterns \(^{(1)}\). It is important to note that the DSM-IV-TR\(^{(5)}\) still uses three diagnostic dimensions since it does not integrates the social interaction and communication skills. These, along with repetitive and stereotyped behaviors, constitute the “diagnostic triad”. The degree of impairment in these areas varies significantly\(^{(6)}\). According to the diagnostic criteria found in the DSM-V (http://www.dsm5.org), changes in the social-communicative dimension for example, reflect difficulties in social-emotional reciprocity in verbal and nonverbal communication behaviors and in establishing and maintaining relationships. The presence of restricted and repetitive behaviors can manifest through stereotypes and repetitions of motor movements, use of objects and speech, as well as restricted interests, excessive and rigid adherence to routines and hypo- or hypersensitivity to sensory inputs\(^{(1)}\).

Specifically on language, delays in the acquisition and development of this ability are common in individuals with ASD and the linguistic impairments in these individuals may be present in morphology, phonology, syntax, semantics and pragmatics\(^{(7)}\). In the Brazilian context, some researches that investigated the formal aspects of oral language of individuals with ASD are highlighted. For example, one study examined the use of verbal communication skills to promote increased mean length of utterance in the context of rehabilitation. Participants were three male individuals with ASD who were 12 years old. The intervention was effective in all three cases, stressing the importance of developing strategies to improve the communicative competence of individuals with the disorder\(^{(8)}\). Another study has investigated the evolution of the functional and grammatical aspects of language in ten boys with ASD with ages between 2.7 and 11.2 years. These aspects were measured and compared at three time points: at initial assessment, after six and 12 months of rehabilitation. Although there were no statistically significant differences between variables over the period evaluated, some of these variables were correlated (e.g., verbs, interpersonal and gestural acts), which denotes the relationship between vocabulary, grammar and pragmatics\(^{(9)}\).

A more recent research has examined the attribution of mental states, vocabulary and sentence length in the speech of ten children with ASD between five and 11 years of age. This analysis was performed at three different times in order to evaluate possible changes resulting from rehabilitation. The results showed that the participants used verbs that denote mental states in speech, but increase in usage of verbs was not observed over the study period. On the other hand, there was an expansion of vocabulary and sentence length of the participants, pointing to the importance of speech and language therapy\(^{(10)}\). A different study investigated the pragmatic abilities of 14 children with ASD during interaction with their teachers in classrooms of mainstream education. Participants were aged between three and eight years. The results showed that gesture was the most used mean in the communicative act. Moreover, as to the communicative functions, a higher occurrence of less interactive ones, such as naming, was observed. The authors also highlighted the existence of a positive correlation between the verbal mode and more interactive communicative functions, as well as between the gestural mode and less interactive functions\(^{(11)}\).

Thus, language is a very relevant field regarding ASD. However, there are still aspects relating to language development in children with the disorder that are poorly understood, as is the case of regression. Therefore, the interest of researchers on regression in language abilities in some cases of ASD has recently increased.

Although there is no consensus on the operational definition of regression of language, it tends to be defined as loss of communicative use of three to five words, except for “papa” and “mama”. In addition, both the time before the regression as the duration of loss should be less than three months\(^{(12-14)}\). The etiology of regression is still unknown, but the biological correlates related to epilepsy have been identified\(^{(15)}\) as well as the existence of genetic predispositions\(^{(16)}\).

There is evidence that between 15 and 33% of children with ASD have regression of some skill, as in social, play or language\(^{(12,17)}\) and the age of onset of regression varies between 12 and 36 months\(^{(18,19)}\). Moreover, there are indications that the loss of skills is more frequent in individuals with autism specifically as compared to other spectrum disorders\(^{(12,14,20,21)}\).

Some studies also show that the oral language skill is the most commonly affected domain by the phenomenon of regression\(^{(12,17,21)}\), occurring in approximate 20% of individuals with ASD. Still, the regression of oral language is often associated with loss of social skills\(^{(13,17,21)}\).

There are recurring empirical efforts that aim to determine whether the regression of oral language presents itself as a risk factor for later development of children with ASD. In this statement, while some studies support this hypothesis\(^{(12,22,23)}\), others have found no relationship between the regression of language and greater developmental impairment\(^{(20,21,24)}\).

For example, a study developed in the United States investigated the phenomenon of regression of social-communicative skill and its relation to aspects related to the overall development of children. Participants were 125 children with and without ASD, whose average age was 8.05 years. The participants were divided into three groups: (1) with ASD and regression, (2) with ASD and no regression, and (3) with typical development. The relationship between regression and symptoms of ASD, comorbid psychopathology, challenging behaviors and
social skills was analyzed. The loss of social-communicative skill proved to be characteristic of ASD, and the group with regression showed greater impairment in all areas investigated as compared to those without regression. Among these impairments, greater severity of symptoms in all areas of the triad and larger deficits in social skills, especially in relation to the appropriateness of behavior to the social context, were reported.

A different study conducted in the UK investigated the correlation between regression of language, autistic symptoms and course of development of 255 children with and without ASD, with an average age of 10.3 years. The average age at which regression occurred was 25 months — moment at which the children produced their first simple phrases. The loss of oral language skills proved to be characteristic of the group with the disorder. The results also revealed that the group of children with ASD and historical regression showed more severe autistic symptoms compared to those with the disorder and without regression.

Another study conducted in the United States specifically investigated the regression of language emphasizing the associated behaviors and the consequences of this phenomenon. Participants were 196 children with loss of language skills, whose average age was 50.6 months at the time of the study. It was observed that regression of language was usually accompanied by loss of social skills and play. Regarding the language level before the regression, most of the children were in the period of acquisition of phrases, with 78% of the children acquiring the first words before 18 months. It was also observed that 73% of participants had cognitive skills below those expected for their age. The researchers concluded that regression of language skills in childhood seems to be a phenomenon that adversely affects the child’s overall development, pointing to the investigative importance of the topic.

In contrast to the previously presented studies, a study conducted in the United States found no relationship between the presence of regression of language and severity of autistic symptoms. The clinical characteristics associated with regression of language skills in 114 children with ASD with a mean age of 41.4 months were investigated. As examined in other studies, the loss of language skills occurred in 29.8% of participants, at an average age of 19.5 months. However, no significant differences were observed between participants with and without regression with regard to the severity of symptoms in the three diagnostic areas (social interaction, communication and repetitive and stereotyped behaviors).

In the same line of investigation, another study conducted in the United States examined the possible relationship between regression of social-communication skill and behavioral characteristics (e.g., symptoms, adaptive behavior) at three and four years. The study included 72 children with ASD, with a mean age of 25.2 months. The group with regression showed greater severity of symptoms only in the dimension of social interaction assessed by Autism Diagnostic Interview-Revised (ADI-R). There were no significant differences between the groups regarding the severity of symptoms in the areas of communication and

METHODS

Study Design and Participants

A retrospective study with contrasting groups was carried out. One group was composed of children diagnosed with autistic disorder with a history of regression of oral language skills (Group 1) and the other of children with the same diagnosis but without a history of regression (Group 2). The groups were matched by gender, race and language level. Thus, the sampled children were preschool, caucasians, diagnosed with autistic disorder and had complete data concerning the measures investigated. The exclusion criteria consisted on the presence of associated physical and sensory disabilities as well as other disorders in the spectrum.

This study is part of a project (CAPES/Fulbright, 2009-10) that investigates predictors of social development in children with ASD, using a database composed of 150 children in preschool age, patients from the Cincinnatti Children’s Medical Center (CCHMC). It consists on a clipping from the general database of CCHMC which was especially authorized for the realization of this project.

This study was financed by Capes/Fulbright and developed at the Graduate Program in Psychology, Universidade Federal do Rio Grande do Sul in partnership with The Kelly O’Leary Center for Autism Spectrum DisordersDivision of Developmental Disabilities Cincinnati Children’s Hospital Medical Center, Ohio, USA.

Instruments

The ADI-R was used to evaluate the regression of oral language. It consists on a standardized semi-structured
interview administered to parents or caregivers of children with suspected ASD. It has 93 items that investigate social and communicative impairments and the presence of repetitive and stereotyped behaviors, both in relation to the first five years of a child’s life, as the current behavior, referring to the 12 months preceding the interview. In this study, items 11 to 28 of the ADI-R were used to specifically investigate the occurrence of regression.

The Autism Diagnostic Observation Schedule (ADOS) (27) is a standardized observational and semi-structured instrument that investigates the social interaction skills, communication, play and repetitive and stereotyped behaviors. In this study, only the dimensions that assess the development of social-communicative skills were used — i.e. those related to social interaction and communication. The ADOS was selected for being an independent measure of the ADI-R and because the observation is made from standardized situations. The behaviors are encoded in scores from 0 to 2 or 0 to 3 depending on the item, being 0 a less committed behavior and 2 or 3 more unusual behaviors. The ADOS consists of four modules for the different levels of expressive language. These modules relate to, respectively, (1) pre-verbal behaviors and isolated words and simple sentences, (2) non-fluent production of sentences, and (3 and 4) fluent speech. In this study, the final scores of the algorithm in the dimensions of social interaction and communication in Modules 1 and 2 and 3 were used since the fourth module concerns adolescents and adults.

The ADI-R and ADOS instruments are considered the gold standard for having been validated in several countries while maintaining its sensitivity and diagnostic accuracy. Thus, the interviewer must undergo prior training that enables the management and coding of the results.

Procedures and Ethical Considerations

The author of the main project, of which this study is part, obtained formal authorization from CCHMC for the use of the database preserving the anonymity of the participants, who were identified by codes.

RESULTS

Of the 30 children who met the criteria for participation in this study, six (20%) had regression of oral language skills. The average age of onset of loss was 25 months (SD=6.19), with a mean duration of 16.3 months (SD=4.24). Regarding the level of language before the regression, five children (83.3%) were using at least five words spontaneously and frequently, and one (16.7%) was using words spontaneously only on an occasional basis. It was reported that four children (66.7%) who had language regression also lost skills of social interaction, as for example, interest and social involvement. The other two children (33.3%) showed no loss in other areas beyond language.

Thus, six children were assigned to regression group (Group 1) and 24 to the no regression group (Group 2). At the time of assessment, the average age of Group 1 was 4.83 (SD=0.33) and of Group 2 3.91 (SD=0.15). The Mann-Whitney U test showed that children with regression of oral language were older than those without such a history, and this difference was statistically significant (U=134, p=0.004).

Given that ADOS is divided into modules according to the language level of the child, the distribution of the different modules was carried out in the two groups (Table 1). The results showed that participants in both groups tended to concentrate in Modules 1 and 2, that is, they were children whose language level ranged from pre-verbal behaviors to sentence production. This distribution was made because the items of each module are formulated according to the language level of the child with different cutoff points. It is therefore pertinent to inform that the cutoffs for Autistic Disorder in Modules 1 and 2 of the ADOS in the dimensions of communication, social interaction and the combined total of both areas are, respectively, 4 and 5; 7 and 6; 12 and 12.

Based on this distribution, the average of the final scores were then obtained in the dimensions investigated, separated by module and group, as shown in Table 2. From the data analysis relating to the final scores, the averages were above the cutoff point for autistic disorder in all dimensions investigated. Thus, this finding shows that both groups (with and without regression) have significant social and communicative impairment.

| Table 1. Distribution of groups with (G1) and without (G2) regression in ADOS modules* per item in the ADOS modules |
|---|---|---|
| Group | Module 1 | Module 2 | Total |
| | n (%) | n (%) | n (%) |
| 1 | 2 (33.3) | 4 (66.7) | 6 (100) |
| 2 | 12 (50) | 12 (50) | 24 (100) |

*Pre-verbal behaviors and isolated words; **sentence production

| Table 2. Mean final scores in the investigated domains according to group |
|---|---|---|
| Domains | Module 1 M (SD) | Module 2 M (SD) |
| COM | Group 1 (with regression) 6.00 (1.00) | 6.00 (1.22) |
| | Group 2 (without regression) 5.91 (0.60) | 6.00 (0.39) |
| INT | Group 1 (with regression) 8.00 (3.00) | 8.50 (0.50) |
| | Group 2 (without regression) 8.91 (0.79) | 9.25 (0.58) |
| COM/INT | Group 1 (with regression) 14.00 (2.00) | 14.50 (1.55) |
| | Group 2 (without regression) 14.83 (1.20) | 15.25 (0.83) |

Legend: M = mean; SD = standard deviation; COM = communication domain; INT = social interaction domain; COM/INT = communication and social interaction domains

For purposes of examining the possible relationship between the regression of oral language skills and the subsequent social and communicative development of participants of this study, the comparison between Groups 1 and 2 was performed using the U Mann-Whitney test, considering a significance
levels≤0.05. The total of dimensions of communication (COM) and social interaction (INT) were compared separately, as well as the combined total of both dimensions (COM/INT).

There was no statistically significant difference in any of the analyses, indicating that the level of social and communicative commitment was similar in patients with and without regression of oral language skills. The results of these comparisons are presented separately, according to the ADOS module.

Thus, in Module 1, the comparison between the groups with and without regression in the communication dimension resulted in U=11 (p>0.05), and the dimension of social interaction U=10 (p>0.05). When both dimensions were analyzed together, the result was U=9 (p>0.05).

With respect to module 2, the comparison between groups with and without regression in the dimensions of social interaction and communication resulted, respectively, in U=23 (p>0.05) and U=17.5 (p>0.05). In turn, after the combined comparative analysis of both dimensions, the result found was U=19.5 (p>0.05). Table 3 shows the values of Z and p in each of the analyses.

Table 3. Between-group comparison of mean scores in the investigated dimensions

<table>
<thead>
<tr>
<th>Dimensions</th>
<th>Module 1</th>
<th>Module 2</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>z (p-value)*</td>
<td>z (p-value)*</td>
</tr>
<tr>
<td>COM</td>
<td>-0.18 (0.66)</td>
<td>-0.12 (0.95)</td>
</tr>
<tr>
<td>INT</td>
<td>-0.38 (0.79)</td>
<td>-0.81 (0.45)</td>
</tr>
<tr>
<td>COM/INT</td>
<td>-0.55 (0.92)</td>
<td>-0.55 (0.60)</td>
</tr>
</tbody>
</table>

* Mann-Whitney U test

Legend: M = mean; SD = standard deviation; COM = communication domain; INT = social interaction domain; COM/INT = communication and social interaction domains

DISCUSSION

The results showed that 20% of children showed regression of language skills in the first three years of life. This finding is close to the proportion found in previous studies. However, it is observed that there is still no consensus on the prevalence of regression in ASD, as several factors seem to influence its establishment such as the conceptual definition, the sample size and the subgroup studied.

The co-occurrence of oral language regression and loss of social skills was observed in the majority of children in this study, which has also been reported in previous findings. Furthermore, co-occurrence of regression of language and epilepsy was not identified, in line with other studies, although this association has been previously shown.

It should be noted that the results of this study showed no statistically significant difference between groups with regard to the investigated behaviors in the dimensions of communication and social interaction. This means that, in the current study, the regression of oral language skills did not seem to present as a risk factor for later social-communicative development in individuals with ASD. While corroborating previous studies, this finding also agrees with others showing the negative influence of regression in the later development of individuals with ASD. This controversy highlights the fact that the nature of the regression and its consequences for the overall development of children still remain poorly understood, in addition to pointing to methodological limitations of the different studies - such as the small number of participants and the use of non-standardized tools for the investigation of regression.

CONCLUSION

The findings of this study seem to not confirm the relationship between the occurrence of regression of oral language and subsequent impairment in the social-communicative development of children with ASD. Although this result is in line with previous research, some methodological points should be addressed. First, the small number of participants with regression of language is highlighted, which precluded the use of parametric tests, which are more statistically robust, influencing therefore the effect found. Moreover, it was not possible to pair the groups by chronological age. Although all the children were at the same age range, the statistical analysis showed that the age difference between the groups with and without language regression was significant. Moreover, the limitation of the use of the database, which restricts access to additional information such as the cognitive ability of the participants, the type of words lost and recovered, the frequency and duration of speech therapy are emphasized. These and other information could assist in understanding the phenomenon.

As a suggestion for future studies, the development of research with more expressive samples as well as with Brazilian participants is emphasized. Furthermore, longitudinal studies that enable researchers to assess the impact of regression on the global development of the child more accurately should be considered. At the qualitative research line, case studies are also desirable for understanding the context of the language loss in families.

Finally, the importance of understanding these findings based on theories of language development — as, for example, from the perspective of social pragmatics approach — is highlighted. This understanding is critical for speech and language therapy, considering that most studies are epidemiological.

ACKNOWLEDGEMENTS

We thank Coordenação de Aperfeiçoamento de Pessoal de Nível Superior (CAPES) and Fulbright for their support to carry out this research. We are also grateful to Patricia Manning-Courtney, MD, Associate Professor of Clinical Pediatrics, University of Cincinnati.

*BB was responsible for the study project and delimitation, data tabulation, analysis and interpretation and for the manuscript writing; RBZ collaborated with data analysis and interpretation, besides the critical review of the manuscript; CAB carried out the project elaboration and was responsible for the general orientation of the manuscript elaboration and execution.
REFERENCES