

Parental responses to autism classic signs in two screening tools

Respostas parentais aos sinais clássicos de autismo em dois instrumentos de rastreamento

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ABSTRACT

Purpose: To analyze the parental responses to questions investigating classic signs of autism using two different instruments (IRDI-questionnaire and M-Chat). **Methods:** Forty-one children, 80% male, with a mean age of 2 years and 8 months, who were evaluated with two autism screening tools recommended by the Brazilian Ministry of Health. After administration of both tools, seven questions were selected to make up the instruments and illustrate symbolically the classic signs of the Autistic Spectrum Disorder (ASD), answered by the parents of the subjects for further analysis. The subjects did not have a formal diagnosis of ASD, or any other diagnosis. **Results:** The main predictors of importance were questions about “make-believe” playing, interest of the child in other children, the child’s response to “motherese”, and exchange of glances between mother and child. **Conclusion:** Not all questions referring to the typical signs of autism showed to be good predictors of importance in the conducted analysis. The data lead us to reflect on a need to analyze a set of signs and not only isolated signs when facing a child with suspected ASD.

Keywords: Autistic disorder; Early diagnosis; Surveys and questionnaires; Speech, language and hearing sciences; Diagnosis

RESUMO

Objetivo: Analisar as respostas parentais a perguntas que investigam sinais clássicos de autismo, em dois instrumentos diferentes: Questionário de Indicadores de Risco para o Desenvolvimento Infantil (IRDI-questionário) e *Modified Checklist for Autism in Toddlers* (M-Chat). **Métodos:** Quarenta e uma crianças, sendo 80% meninos, com média de idade de 2 anos e 8 meses foram avaliadas com dois instrumentos de rastreamento de autismo, recomendados pelo Ministério da Saúde. Após a aplicação integral dos instrumentos, foram selecionadas sete perguntas que compõem os instrumentos e ilustram emblematicamente sinais clássicos de transtorno do espectro do autismo (TEA), respondidas pelos pais dos sujeitos, para posterior análise. As crianças avaliadas não tinham qualquer diagnóstico fechado de TEA ou outros transtornos. **Resultados:** Os principais preditores de importância foram questões sobre brincar de “faz de conta”, interesse da criança por outras crianças, resposta da criança ao “manhês” e troca de olhares entre mãe e criança. **Conclusão:** Nem todas as perguntas que abordam os sinais típicos de autismo mostraram-se bons preditores de importância na análise realizada. Há necessidade de analisar o conjunto de sinais e não apenas sinais isolados, quando se está diante de uma criança com suspeita de TEA.

Descritores: Transtorno autístico; Diagnóstico precoce; Inquéritos e questionários; Fonoaudiologia; Diagnóstico

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INTRODUCTION

The diagnosis of autistic spectrum disorder (ASD) has been established early, allowing a more immediate intervention and a better prognosis for the children^(1,2,3).

Parents of children with ASD often report that their concerns about their children's development begin around the age of 12 months⁽⁴⁾, although the diagnosis of ASD can only be established decisively around the age of 3 years^(1,5). Despite advances in genetic and biomedical research on ASDs in recent years, there is still no biological marker allowing its diagnosis^(3,6). Therefore, identification and diagnosis of ASD should be based on characteristics of the child's behavior, which often makes this process difficult and slow^(1,6,7).

The development of tracing procedures to be used at early ages required the establishment of a minimum set of signs that could serve as a reference. Parental reports, clinical anamneses, questionnaires, and family videos have been used for this purpose^(8,9,10).

The clinical anamnesis was the first area of identification of these signs. Parental reports of child behavior, mainly in the first and second years of life, driven by the clinical professional in sessions of anamnesis and even in those of general recommendations, was of great value in delineating this set of signs⁽¹⁰⁾.

This type of survey was transformed into a report guided by the clinical interview. Clinicians started to supplement the information on autistic signs provided by parental interviews with those collected by other instruments such as family videos, questionnaires applied by the clinicians themselves, and clinical observation of the child^(10,11).

Initially, the surveys were based on videos of family celebrations (birthday and/or religious parties, vacations, etc.), which are considered, to date, anchor points in the analysis^(9,10,11). Nonetheless, the studies indicated difficulties in discriminating signs of autism in very young children⁽¹⁰⁾.

A study on the analysis of videos for children from 0 to 6 months⁽¹²⁾ of age reported great difficulty in dealing with the data, since the signs are very fragile at this stage. At more advanced ages, the differences become more striking: from the age of 8 to 10 months, the child's gaze towards people and there is some atypicality in the child's gaze towards people and objects, and in some characteristics of his/her ability regarding social and communicative skills from the age of 9 to 12 months, there are differences in psychomotricity and signs of aversion to social interaction, absence of smiles and facial expressions, and dysfunction in intention and imitation^(10,11,13).

Questionnaires designed for parents are also often used to collect information on the development of children suspected of having autism. For tracing/screening purposes, some of the instruments used worldwide have been validated for use in Brazil: the Modified Checklist for Autism in Toddlers (M-Chat)⁽¹⁴⁾ and the Autism Behavior Checklist (ABC)⁽¹⁵⁾.

The M-Chat is a questionnaire used for ASD screening

comprising 23 questions for parents of children aged 18 to 24 months, with answers of "yes" or "no", indicating the presence of behaviors known as early signs of ASD. It includes items related to the child's interests in social engagement, ability to maintain visual contact, imitation, repetitive and "make-believe" playing, and the use of eye contact and gestures to direct the social attention of the partner or to ask for help^(7,14).

The Brazilian instrument – Clinical Indicators of Child Development Risk (*Indicadores Clínicos de Risco para o Desenvolvimento Infantil*, IRDI) – was developed by Brazilian researchers and validated for the use of health professionals, to observe behaviors of the mother-infant dyad in the period from 0 to 18 months of age. The IRDI aims to detect risks for the development of the child, although it is not a specific instrument for ASD⁽¹⁶⁾.

The IRDI-questionnaire was adapted by Brazilian researchers with the purpose of tracing cases of ASD. It is a retrospective questionnaire for parents, indicated to be used in children between the ages of 18 months to 7 years^(17,18).

The final document of the Brazilian Ministry of Health⁽¹⁹⁾ on this subject indicates the use of two instruments for ASD screening. These instruments, validated for use in Brazil, are the IRDI and M-Chat.

In a study⁽⁸⁾ that addressed the initial development of shared attention, exchange of looks, and affection during the first two years of life, using retrospective interviews with parents and analysis of family videos of children later diagnosed with ASD, the results showed that the interviews with parents confirmed the results of the observations from the videos, indicating that early problems in socio-communicative abilities are characteristics of children with ASD⁽⁸⁾.

There are three points of controversy regarding the questionnaires: parents may be influenced by information about the diagnosis, the recollection of facts may be compromised, and parents lack a perspective oriented by a knowledge of ASD.

The several instruments, in one form or another, highlight some signs of autism that may, therefore, be regarded as persistent, *i.e.*, signs with value for the identification of cases at risk. They include receptiveness of the child to "motherese", response when called by name, exchange of looks between child and mother, interest of the child in other children, and "make-believe" playing^(2,5,11,20,21,22).

On the assumption that the use of screening instruments for ASD by the speech and hearing specialist is more effective than the simple knowledge of classic clinical signs such as those described, the goal of this study was to analyze the parental responses to questions that specifically evaluate these classic signs of autism in two different instruments: the IRDI-questionnaire and the M-Chat.

METHODS

The study was approved by the Research Ethics Committee

of the *Pontifícia Universidade Católica*, where it was developed (opinion no. 766,311). All subjects signed the Informed Consent Form, authorizing the use of data for research.

The sample was obtained by convenience and consecutive selection. The selection criteria included subjects aged more than 18 months, without a diagnosis of genetic, neurological, or metabolic disorders.

In the period between May 2014 and May 2015, after verifying the exclusion criteria (age below 18 months and presence of genetic, neurological, or metabolic disorders), we evaluated 41 children, of whom 33 (80.5%) were boys. The average age of the sample was 2 years and 8 months (SD=0.8) and the median age was 2 years and 8 months, ranging from 1 year and a half to 4 years and 6 months. Of the respondents, 92.7% were mothers and 53.7% had completed secondary education.

The study was conducted at the Children Hearing Center (*Centro Audição na Criança*, CeAC) of the Division of Education and Rehabilitation of Communication Disorders (*Divisão de Educação e Reabilitação dos Distúrbios da Comunicação*, DERDIC) - *Pontifícia Universidade Católica* of São Paulo. This is a high complexity center, accredited by the Brazilian Unified Health System (*Sistema Único de Saúde*, SUS), which provides care to children suspected of having or have been diagnosed with hearing impairment. The center offers audiological diagnosis, selection and recommendation of individual hearing aids, hearing and speech therapy, and family monitoring and guidance.

As part of the service's routine, after the first appointment with an ENT doctor, the children undergo an initial interview with a speech and hearing specialist to gather survey data on audiological anamnesis, before undergoing the required audiological tests.

At the time of the interview, after checking the selection criteria, we also applied the instruments IRDI-questionnaire and M-Chat. The same researcher applied the instruments to all the guardians of the children.

Once the evaluated children had no conclusive diagnosis of ASD or other developmental disorders, those with ASD warning signs according to the instruments used, were referred for more in-depth diagnostic evaluations at the SUS network, according to the possibilities offered in the region closest to their homes.

The audiological tests performed were: impedance audiometry, otoacoustic emissions, play audiometry, Visual Reinforcement Audiometry (VRA), and Brainstem Auditory Evoked Potential (BAEP), according to the requirements of each case. Equipment used: Audiometer model AC33, Impedance Audiometer model AT235h and Eclipse Black Box- ABRIS software, all from Interacoustics®.

The instruments IRDI-questionnaire and M-Chat were applied in their entirety, following the scoring criteria characteristic of each one. After the instruments were scored and the risk criteria were verified, the children were divided into two groups: "risk" and "no risk". In other words, the application of

the instruments, following their specific scoring instructions, defined the groups.

After this procedure, we selected seven questions, four from the M-Chat and three from the IRDI-questionnaire. These questions were chosen for *a posteriori* research purpose, because they characterize those that are considered classic ASD signs, according to recurrent data in the literature, which highlight, among ASD signs, a difficulty in maintaining eye contact^(2,20), absence of response from the child to "motherese"^(22,23) and when he/she is called by name^(11,24), as well as a difficulty in social interaction⁽²⁴⁾ and playing "make-believe" games^(7,25,26). Such signs are represented in the following questions, which are part of the utilized instruments.

Questions selected from the M-Chat:

- "Does your child display interest in other children?"
- "Has your child already played 'make-believe', as, for example, pretending that he/she is talking on the phone or taking care of a doll, or any other game of 'make-believe'?"
- "Does your child look at you in the eye for more than one or two seconds?"
- "Does your child respond when you call him/her by name?"

Questions selected from the IRDI-questionnaire:

- "Did the mother speak with the child in a way particularly addressed to him/her (motherese)?"
- "Did the child react to motherese?"
- "Was there an exchange of glances between the child and the mother?"

It is worth noting that M-Chat defines as "at risk" those children scoring at least two of the six critical items that comprise the instrument. Even though this has not been the selection criteria of the questions analyzed in this study, two of the four questions listed comprise the critical items of the instrument.

To identify the relevance of the issues, we performed the Two-Step Cluster analysis, using the log-likelihood function and the Schwarz's Bayesian criterion (BIC) to measure distance. To identify the contribution of each variable in the clusters, we used predictors of importance, in probability, ranging from 0 to 1, in which closeness to 1 represented a greater relevance within the group. The Chi-square test was used to observe the associations between the clusters and the presence of risk verified by the instruments. We assumed the descriptive level of 5% ($p < 0.05$) for statistical significance.

The data were analyzed using SPSS, version 22.0 for Windows.

RESULTS

Of the 41 subjects evaluated, 7 (17.9%) presented hearing loss. Regarding the hearing loss cases, 71.4% were severe, and sensorineural loss was the most frequent type (85.7%) (Table 1).

Table 1. Number and percentage of children, according to hearing characteristics

Variable	Category	n	(%)
Hearing	Normal	32	(82.1)
	Hearing loss	7	(17.9)
	Total	41	(100.0)
Degree of loss	Mild	1	(14.3)
	Moderate	1	(14.3)
	Severe	5	(71.4)
Type of loss	Neurosensorial	6	(85.7)
	Conductive	1	(14.3)
	Mixed	0	(0.0)
Loss	Unilateral	2	(28.6)
	Bilateral	5	(71.4)

Out of the 7 subjects with hearing loss, 4 (57%) were at risk for ASD according to the instruments used. Out of those 4, 1 with mild hearing loss and 1 with moderate hearing loss presented a risk of ASD according to the IRDI-questionnaire and the M-Chat. Two subjects with severe loss displayed a risk of ASD by the IRDI-questionnaire.

Of the 41 subjects evaluated, 22 (53.7%) were at risk of ASD according to the M-Chat and 24 (58.5%) by the IRDI-questionnaire, with a good level of agreement between the instruments ($Kappa=0.41$; $p=0.009$).

The clusters, M-Chat, and IRDI-questionnaire showed a good quality. For the M-Chat, among the issues discussed, the main predictors of importance were question 5 (has your child already played “make-believe”, as, for example, pretending that he/she is talking on the phone or taking care of a doll, or any

other game of “make-believe”?) and question 2 (does your child display an interest in other children?), respectively. Questions 10 (does your child look at you in the eye for more than one or two seconds?) and 14 (does your child respond when you call him/her by name?) did not show significance (Figure 1).

When analyzing the groups, we verified that in cluster 1, for the statistically significant variables – questions 5 and 2 – 100% of the records had “yes” as an answer, whereas in cluster 2, the majority of the responses was for the “no” category. We should emphasize that negativity comprised 89.5% of the answer to question 5 and 78.9% of those to question 2.

Thus, we observed that children in cluster 2 had a higher risk probability, according to the M-Chat, when compared with children in cluster 1 (89.5% versus 31.8%; $p<0.001$) (Table 2).

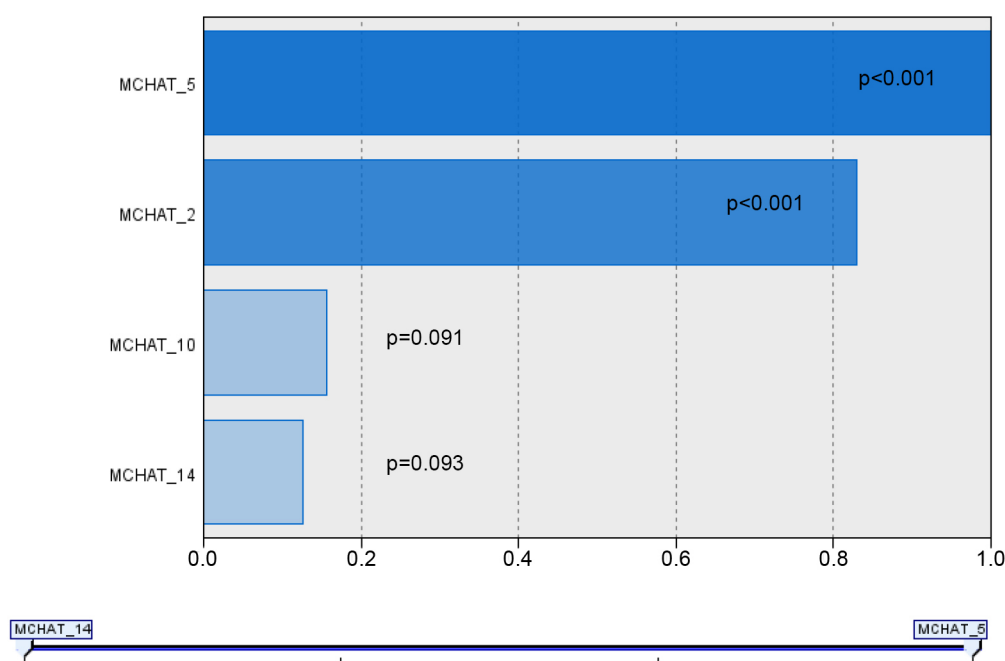
Table 2. M-Chat - Number and percentage of children, according to presence of risk and clusters

Cluster	M-Chat				p-value
	No risk		Risk		
	n	(%)	n	(%)	
1	15	(68.2)	7	(31.8)	<0.001
2	2	(10.5)	17	(89.5)	
	17	(41.5)	24	(58.5)	

Chi-square test ($p<0.005$)

Subtitle: M-Chat = Modified Checklist for Autism in Toddlers

As for The IRDI-questionnaire, the three questions analyzed were relevant, respectively: 3 (does the child react to “motherese”); 5 (were there exchanges of glances between the child and the mother?), and 2 (did the mother speak with the child in a way particularly directed to him/her “motherese”?) (Figure 2).

**Figure 1.** Two-Step Cluster for the M-Chat

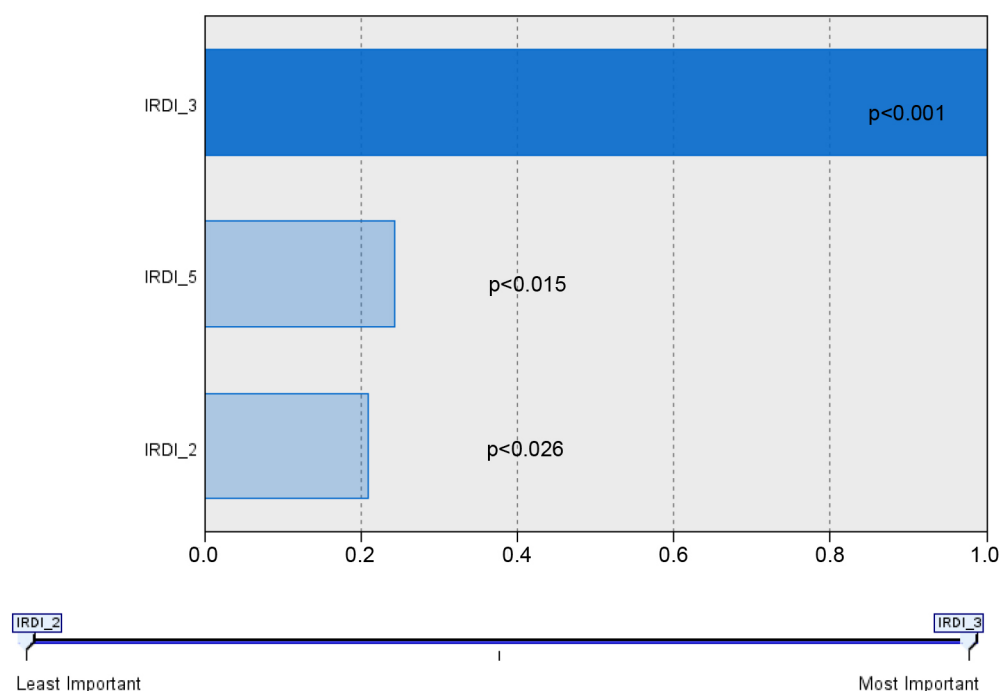


Figure 2. Two-Step Cluster for the IRDI-questionnaire

As regards the groups, we identified that in cluster 1, the answer “always” was 100% present in question 3. For questions 5 and 2, “always” appeared in 86.7% of the responses. On the other hand, in cluster 2, 45.5% of responses were for the option “never” in question 3. As regards questions 5 and 2, the answer “always” stood out, but in proportions that were lower than those in the responses for cluster 1, respectively, 36.4% and 54.5%.

We verified that children in cluster 2 had a higher risk probability, according to the instrument IRDI-questionnaire, when compared with children in cluster 1 (100.0% *versus* 36.7% respectively; $p < 0.001$) (Table 3).

Table 3. IRDI-questionnaire - Number and percentage of children, according to the presence of risk and clusters

Cluster	IRDI- questionnaire				p-value
	< 32.5 (no risk)		≥ 32.5 (risk)		
	n	(%)	n	(%)	
1	19	(63.3)	11	(36.7)	<0.001
2	0	(0.0)	11	(100.0)	
	19	(46.3)	22	(53.7)	

Chi-square test ($p < 0.005$)

Subtitle: IRDI-questionnaire = Clinical Indicators of Child Development Risk (*Indicadores Clínicos de Risco para o Desenvolvimento Infantil*)

DISCUSSION

The study of the parents’ perception regarding early signs of autism may bring important information about how clinicians may access such signs. This research is based on the assumption

that, in addition to knowing the key traits of autism, clinicians must use identifying instruments standardized for this purpose.

We observed a long period between the first concerns expressed by the parents and the age of diagnosis itself^(4,27). Increased awareness and information about the initial manifestations of ASD, as well as the use of specific tools, could help decrease this time^(2,28).

The instruments used in this study were able to identify deaf children at risk for ASD, indicating the presence of possible comorbidity, which would be in accordance with another study⁽²⁹⁾ that stated that there are no differences in autistic behaviors displayed by children who are deaf and those who have normal hearing. Nonetheless, some studies have identified difficulties in the diagnosis of ASD in deaf children, which have been linked to a lack of specific instruments for the diagnosis and the need for an increased awareness by health professionals^(29,30).

Even though a lack of eye contact is one of the most well-known classic signs of ASD, the question about this topic in the M-Chat was not so significant in the analysis conducted, nor was the question of the same instrument on the child’s response when called by name. Such data draw attention because they differ from those in the literature describing these signs as main concerns reported by families and observed by clinicians^(8,11,20). On the other hand, the question in the IRDI-questionnaire about the same topic – exchange of glances – proved to be relevant in the analysis performed, indicating a good prediction index.

The question that investigates the use of “motherese” by the mothers was also not a good predictor of importance, which draws similar attention since it diverges from the data from studies on this topic, in which there was a reduction in the use of “motherese” in cases of ASD^(22,23).

It is worth noting that the studies that addressed the issue of “motherese” were conducted from analyses of family videos, while the data for this study were collected from questionnaires directed to the parents. This difference in the collection of the information deserves attention and, perhaps, justifies the difference pointed out.

The first concerns of parents about their children with ASD were evaluated in a study⁽²⁰⁾ with an open-ended questionnaire which found that concerns about social development or autistic behaviors were frequent, but not exclusive. Specific characteristics of autism (changes in socioemotional development, language development delay, autistic behaviors) and warning signs that are not specific (difficulties in behavior not specific to autism, changes in perception and in motor milestones, medical problems, etc.) were represented in equal proportions. These findings are in agreement with those obtained in the present study, which did not verify a predictive power of some of the classic signs of autism in parental responses, using questionnaires directed for this purpose.

We verified that the questions about the child’s interest in other children, as well as the game of “make-believe”, were characterized as good predictors, which is in agreement with the information provided by the studies on warning signs and tracing of ASD^(2,7,11,20).

The results of the analysis of all these questions, separately, although different, indicated the importance of recognizing that the instruments, when fully used, are capable of fulfilling their function, *i.e.*, detecting potential cases of ASD^(2,7,14,17,18).

We observed an important movement in the sense of promoting increased information and awareness of the population in general, and health and education professionals about ASDs and their clinical manifestations^(2,28). Nonetheless, the results described in this study pointed out to a need for considering not only the way to access the classic signs of autism, but also the importance of analyzing a set of signs and not isolated signs, preferably using specific instruments for this purpose.

CONCLUSION

Not all questions that address the typical signs of autism were good predictors of importance, even if the result of the application of the instruments, as a whole, has been that of tracing possible cases of ASD, *i.e.*, although the evaluations by the instruments have indicated a possible risk of ASD, in some cases, the responses from the parents to specific questions did not disclose similar information. There is a need to analyze the set of signs and not only isolated signs when one faces a child with suspected ASD. The use of specific and standardized instruments by hearing and speech specialists can help them fulfill their role in identifying possible cases of ASD and, therefore, providing consistent referral so the diagnosis can be established by competent professionals.

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