Clinical Risk Indicators for Early Childhood Development Questionnaire: evaluation of the sensibility for autism spectrum disorders

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ABSTRACT

Purpose: To assess the sensitivity of the Clinical Risk Indicators for Early Childhood Development (IRDI) questionnaire for parents in identifying autism spectrum disorders. Methods: Seventy-two subjects, which comprised parents of children aged 2 years 11 months to 7 years 7 months, participated in the study divided into two groups, namely the study and control groups. The IRDI questionnaire was administered in both groups, and the CARS-BR was applied in the study group. The data were subjected to statistical analysis, and the Pearson correlation coefficient between the IRDI questionnaire and CARS-BR was performed only for the study group to calculate validity. In defining the cutoff score generated by the IRDI questionnaire, the receiver-operating characteristic (ROC) curve was used, and sensitivity, specificity, and positive predictive value were calculated. In the reliability analysis for the IRDI questionnaire, the Kappa coefficient and overall concordance was used.

Results: The IRDI questionnaire was easy to use, quick to complete, and inexpensive. Analysis of the internal consistency of the questions of the instruments presented good content. The IRDI questionnaire was positively correlated with the CARS-BR. The ROC curve analysis revealed good performance. Conclusion: The evaluation of the sensitivity of the instrument for purposes of identifying autism spectrum disorders showed good performance according to the established cutoff point in the study.

Keywords: Speech, language and hearing sciences; Autistic disorder; Questionnaires; Language; Diagnosis

RESUMO

Objetivo: Avaliar a sensibilidade do IRDI-questionário para pais, para rastreamento de transtornos do espectro do autismo. Métodos: Participaram do estudo 72 sujeitos, familiares de crianças na faixa etária de 2 anos e 11 meses a 7 anos e 7 meses, divididos em dois grupos: grupo pesquisa e grupo controle. O IRDI-questionário foi aplicado nos dois grupos estudados e a CARS-BR foi aplicada no grupo pesquisa. Os dados foram submetidos à análise estatística e, para a validade, foi realizado o coeficiente de correlação de Pearson, entre o IRDI-questionário e a CARS-BR, somente para o grupo pesquisa. Na definição do ponto de corte do escore gerado pelo instrumento IRDI-questionário, foi utilizada a curva ROC e calculados os valores de sensibilidade, especificidade e valor preditivo positivo. Na análise de concordância para o IRDI-questionário, foi utilizado o coeficiente de Kappa e concordância total. Resultados: O IRDI-questionário mostrou-se de fácil aplicação, rapidez no preenchimento e baixo custo. A análise da consistência interna das questões do instrumento apresentou bom índice. Na comparação com a CARS-BR, apresentou correlação positiva. A análise da curva ROC identificou bom desempenho. Conclusão: A avaliação da sensibilidade do instrumento, para fins de rastreamento de transtornos do espectro do autismo, mostrou bons índices, a partir do ponto de corte estabelecido no estudo.

Descritores: Fonoaudiologia; Transtorno autístico; Questionários; Linguagem; Diagnóstico

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INTRODUCTION

First described by Leo Kanner in 1943, autism is considered a pervasive developmental disorder characterized by severe impairment in socialization, qualitative impairments in communication, and repetitive and stereotyped behaviors\(^{(1-4)}\).

Clinicians and researchers have been using, in the specialized literature, the concept of “autism spectrum disorders,” which includes autism, Asperger syndrome, and pervasive developmental disorders not otherwise specified\(^{(1-4)}\) and has been recently adopted by the American Psychiatric Association as the official nomenclature\(^{(5)}\).

Clinical signs of autism spectrum disorders can be observed in very young children, as they involve skills that typically develop in early life. During this phase, most diagnosed children have signs reported by parents and/or caregivers. In addition, many families express their first concerns to pediatricians around 18 months of the child’s life\(^{(6-8)}\).

The concerns expressed by parents of children with autism spectrum disorders are varied; however, complaints about delayed language development are among the most common and consensua\(^{(9-11)}\). Among such complaints, noteworthy as a main concern of parents is the absence of the child’s response when called by name, which is often interpreted as a hearing problem\(^{(11)}\).

Family concerns in relation to the language development of such children are one of the reasons why the speech therapist is, in many cases, the first professional sought by the family, even before the diagnosis of autism spectrum disorders is issued or evaluated.

Hence, the relevance of speech therapy in early identification of possible risk of autism spectrum disorders is evident. In addition, for diagnosis made by a competent professional, the availability of tools to assist the speech therapist in assessment procedures and necessary clinical referrals is mandatory.

The diagnosis of autism spectrum disorders is essentially clinical and is made from observations on the behavior of children and interviews with parents and/or caregivers. Scales and screening tools and standardized assessment have been useful and necessary tools that can contribute to referral for diagnosis\(^{(11)}\).

Various instruments and scales are used worldwide, each with a specific application, depending on the purpose and child’s age\(^{(11-15)}\). In addition to the issues related to translation and validation processes for use in Brazil, the use of these instruments is restricted due to the payment of royalties to authors and publishers responsible for their development and publication.

The Clinical Risk Indicators for Early Childhood Development (IRDI) tool was developed and validated by a group of Brazilian experts for use by health professionals so that the behavior of the mother-infant dyad can be observed; thus, it identifies signs of risk for child development in general. It consists of 31 indicators that are observed in the first 18 months of the child’s life\(^{(16,17)}\). The IRDI is not a specific tool for autism spectrum disorders but broadly for problems in child development. It focuses on the observation of behaviors involved in the interaction of the mother-child dyad, between 0 and 18 months old. In a previous study\(^{(18)}\), the adaptation of IRDI to an IRDI questionnaire format was proposed for parents of children diagnosed with autism spectrum disorders. The results obtained in the sample proved that it was possible to differentiate children with autism spectrum disorders from typical development children.

The present study focused on the applicability of the IRDI questionnaire\(^*\) for early diagnosis and, by extension, effectiveness of treatment in children with autism spectrum disorders.

The aim of this study was to evaluate the sensitivity of the IRDI questionnaire for parents in identifying autism spectrum disorders.

METHODS

The study was approved by the Ethics Committee in Research of the Pontifícia Universidade Católica de São Paulo and Hospital das Clínicas, Faculty of Medicine, Universidade de São Paulo (protocol nos. 0813/11 and 7598/12). All the subjects signed a free and informed consent form authorizing the use of data for research.

Sample and locations of the study

The sample was a convenience sample obtained by consecutive selection. Among the 72 subjects who participated, 65 were mothers and 7 were fathers of children aged 2 years 11 months to 7 years 7 months.

The sample size was calculated considering the efficiency level of the instrument as good when higher than 75%. Thus, the efficiency index was assumed to be approximately 85%, with an accepted error of 10%, to obtain estimates with 95% confidence levels. It was estimated that it would take at least 49 subjects in the study (adding up the two groups).

The subjects were divided into two groups as follows: the study group, composed of parents of children with autism spectrum disorders, and the control group, composed of relatives of typical development children. As this is a retrospective instrument (with questions about the initial development of the child as an infant, from zero to 18 months) that relies exclusively on the memory of the parents for the answers, the age of 7 years

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\* Although the IRDI was developed by a group of researchers who were mostly from the area of psychology, it is an instrument recommended for use by health professionals in general (physicians, health workers, speech therapists, and nurses, among others). To that extent, the proposal of the instrument assumes a multidisciplinary clinic as essential for timely diagnosis by a qualified professional.

It is worth noting, though, that the study in question had as coauthor a psychologist who was also coauthor of the research that validated the IRDI instrument.
was set as the maximum limit to minimize problems associated with reliability of responses\textsuperscript{(19)}. The diagnosis of autism spectrum disorders, based on the \textit{Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV)}, and the \textit{International Classification of Diseases, Tenth Revision}, can hardly be safely established before 3 years of age\textsuperscript{(20,21)}, which is why this age group was established for the selection of subjects in this study.

The study group consisted of 36 subjects (34 mothers and 2 fathers) who were parents of children, aged between 2 years 11 months and 7 years 7 months, who attended at the autism outpatient service (Hospital das Clínicas, Faculty of Medicine, Universidade de São Paulo) and at a private practice, and were selected according to the following inclusion criteria:

- had a diagnosis of autism spectrum disorder according to the DSM-IV criteria, without genetic, metabolic, or neurological disorders documented in medical records;
- the IRDI questionnaire was answered by a parent;
- the parents signed the Free and Informed Consent Form.

All the subjects were selected in order of arrival at the two study sites between August 2011 and September 2012.

It is noteworthy that data collection in private practice constituted a minority of the data (11%). Moreover, most of the patients had previously been diagnosed with autism spectrum disorder. Only 10\% had a diagnosis concomitant to the application of the questionnaire.

The control group consisted of 36 subjects (31 mothers and 5 fathers) who were parents of children attending the Municipal Curumim I Preschool in Santana de Parnaíba, all with typical development, aged between 3 years 3 months and 6 years 10 months and selected according to the following inclusion criteria:

- parents and the school had no complaints about their development; no diagnosis of genetic, neurological, or metabolic disorders;
- parents answered the IRDI questionnaire;
- parents signed the Free and Informed Consent Form.

The groups were matched for age and education of the respondents.

Materials

- The IRDI questionnaire is self-administered and contains 31 retrospective questions addressed to parents, whose responses were in accordance with a 5-point Likert scale (never, rarely, sometimes, often, and always) and an option “do not remember.”
- The Brazilian version of the Childhood Autism Rating Scale (CARS-BR)\textsuperscript{(22,23)}. CARS is composed of 15 items and is considered as the criterion standard in the identification of children with autism. Its importance also stems from the possibility to differentiate between mild/moderate and severe autism. It is brief and suitable for use with any child older than 2 years\textsuperscript{(23)}. In Brazil, CARS-BR is currently considered as the criterion standard rating scale because the Autism Diagnostic Interview–Revised and Autism Diagnostic Observation Schedule (scales considered as the criterion standard abroad) have not gone through the whole validation process and therefore not available for use.

Procedures

Application of the instruments in the study group

As a routine part of the outpatient autism service of the Institute of Psychiatry, Hospital das Clínicas, Faculty of Medicine, Universidade de São Paulo, a team of researchers developed a protocol for multidisciplinary evaluations. Patients who attended the service for psychiatric clinical practice were invited to participate in this survey. Once the interest of the parents was confirmed, a revisit was scheduled for assessments. Thus, the study group was composed of the parents of these children, selected according to the inclusion criteria described earlier and diagnosed with autism spectrum disorder by the psychiatrist based on the DSM-IV criteria.

On the scheduled date, the researcher gave a brief explanation of the contents of the IRDI questionnaire, and this was delivered to a parent (the one who came forward) for completion in writing, in accordance with the instructions contained in the instrument.

The application of the CARS-BR was conducted in the form of a semistructured interview by the researcher with a parent. First, a brief explanation was provided about the content of the interview to ascertain whether the informant would be able to provide the necessary information. The average time of duration of the interviews was 30 minutes.

In the private practice, subjects who met the inclusion criteria for this study were consulted to determine whether they were interested in participating. If so, the questionnaire was delivered to one of the parents, and the CARS-BR was administered by the researcher, for an average duration of 30 minutes. The diagnosis of autism spectrum disorder was conducted by a psychiatrist based on the DSM-IV criteria.

One must stress that the CARS-BR and IRDI questionnaire were applied by the same researcher.

Application of the IRDI questionnaire to the control group

After acquiring approval from the Secretary of Education of the Municipality of Santana de Parnaíba to conduct this research, the researcher attended the school designated by the municipality to contact with the directors. It was agreed that the IRDI questionnaires would be distributed by the researcher on the parents’ meeting day, according to the school calendar.

At first, after the selection of the classrooms where the questionnaires would be administered (according to age groups established as inclusion criteria for the study), the school board
was inquired if they had complaints about any student regarding their general language or behavioral development. Students who were subject to a complaint of this nature by the school were excluded from the study.

On the date of the parents’ meeting, the researcher briefly explained about the study and inquired at the time of delivery of the questionnaires if parents had any complaints about the overall, language, or behavioral development of their children so as to exclude those with complaints of this nature.

An explanation of the contents of the IRDI questionnaire was provided to a parent (the one who came forward), who responded in accordance with the instructions contained in the instrument. The average time to complete the questionnaire was 15 minutes.

Analysis of the results

The cutoff point for calculating the score of the IRDI questionnaire was established by the sum of responses according to the following scoring system: always, 0; often, 1; sometimes, 2; seldom, 3; never, 4; and do not remember, not rated.

Descriptive statistical analysis of data using absolute and relative frequencies, measures of central tendency (mean and median), and dispersion (standard deviation, range) was performed.

Analysis of normality for quantitative variables was performed using the Komolgorov-Smirnov test, and as the data were normally distributed, parametric tests were used. To verify homogeneity between the groups, we used the chi-square association and the mean comparison Student’s t test. For validity, the Spearman correlation coefficient analysis was performed between the IRDI questionnaire and CARS-BR for the study group.

In defining the cutoff score derived from the IRDI questionnaire, the receiver-operating characteristic (ROC) curve was used, and the sensitivity, specificity, and positive predictive value were calculated. In the reliability analysis for the IRDI questionnaire, the Kappa coefficient and overall concordance was used. For comparison of the IRDI questionnaire and CARS-BR, the sensitivity and positive predictive value were calculated.

A statistical significance of 5% (p≤0.05) was assumed. Data were entered into the database, and analyses were performed using SPSS version 17.0 for Windows.

RESULTS

The study population consisted of 72 children homogeneously distributed (n=36 for each group), among whom the percentage of boys was higher in both groups (p=0.114). No difference was found between the mean age of the children in both groups (p=0.199).

Regarding parental education, also no difference was found between the groups (p=0.690). Although the subjects were from a public clinic and private practice, no difference with regard to socioeconomic and cultural levels were found, as 89% of the subjects in the study group were treated at the same hospital (Table 1).

After checking for homogeneity between the groups, the assessment of the consistency of the IRDI questionnaire was performed using the Cronbach alpha, which showed a value of 0.89, featuring good consistency.

As to validity, the Spearman correlation coefficient was r=0.42, with a positive correlation between the results of both instruments (p=0.011); that is, the more severe the framework within the autistic spectrum disorders (according to CARS-BR),

<table>
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<th>Variable</th>
<th>Category</th>
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<th>Study Group</th>
<th>p-value</th>
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<td></td>
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</tr>
</tbody>
</table>

Chi-square test (p<0.05)
the higher the scores on the IRDI questionnaire. Figure 1 shows the relationship between the instruments.

The ROC curve analysis for the data from this study identified an area of 84.6%, which is excellent and significant (p<0.001; 95% confidence interval, 75.3% – 93.8%; Figure 2). The sum of the scores of the IRDI questionnaire responses ranged from 0 to 124. The ROC curve was used to establish the best cutoff point; that is, we sought the best sensitivity value and a moderate specificity, as the intention of the questionnaire is to identify/screen for potential cases.

The cutoff was defined from the score of 32.5 and showed a sensitivity of 88.9%, specificity of 63.9%, and positive predictive value of 71.1%. A moderate agreement (Kappa=0.58; p<0.001) was observed between the presence/absence of a clinical diagnosis of autism (DSM-IV) and presence/absence of a risk (indicated by the IRDI questionnaire), with overall concordance of 76.4% (Table 2).

The relationship between the instruments was also evident in the high values of sensitivity and positive predictive value (respectively, 87.9% and 90.6%) obtained in the comparison between the CARS-BR and IRDI questionnaire (Table 3).

DISCUSSION

The scarcity and need to use screening instruments for autism spectrum disorders in Brazil are mentioned in the literature as key points to improve the effectiveness of referrals in public health and to initiate epidemiological surveys of the pathologies involved\(^{[14,15,24]}\).

The identification of candidate patients for diagnosis of autism spectrum disorders has been gaining importance in recent years. Methods for accomplishing this task have become essential, and studies show that the clinical tools used should be relatively quick and at the same time be able to gather data that may contribute to an early intervention\(^{[6,25,26]}\).

The present study aimed to remedy the aforementioned shortcomings by using a new instrument, the IRDI questionnaire, that had been established and validated for the Brazilian population.

In a scenario of suspicious diagnosis, where the speech therapist has an extremely important role in the multidisciplinary team, the IRDI questionnaire can be a useful tool for referral to a diagnosis of autism spectrum disorders, which by itself, can be accomplished by professionals qualified to do so.
The IRDI questionnaire presented good internal consistency and results consistent with other studies of reliability of instruments used for screening and diagnosis of autism spectrum disorders, namely the Autism Spectrum Questionnaire (ASQ), and CARS-BR and CARS\(^{14,19,23}\).

As in other similar studies, it was possible to establish a cutoff for the IRDI questionnaire, with good levels of sensitivity in identifying children with autism spectrum disorders, in relation to both the DSM-IV and CARS-BR criteria. Moderate agreement was observed between the presence/absence of a clinical diagnosis of autism (DSM-IV) and presence/absence of risk (indicated by the IRDI questionnaire). Similar results were found in studies on the Modified Checklist for Autism in Toddlers, Autism Behavior Checklist (ABC/ICA), and ASQ\(^{13-15}\).

In the comparison between the IRDI questionnaire and CARS-BR, high values were observed for sensitivity and positive predictive value. These values are close to those obtained in the validation study of the Autism Spectrum Questionnaire\(^{14}\). They are also consistent with the literature, which is consensual to point out that because the purpose of screening instruments/screening is to identify children at higher risk for referral to more detailed assessments without a pretense for precipitated diagnosis, they should seek to maximize sensitivity to lose the least number of possible cases\(^{11,27,28}\).

To this extent, it can be stated that the established cutoff point for the IRDI questionnaire in this study allowed that goal to be reached.

Some limitations may be identified, such as the fact that we used a convenience and small sample, which should be increased in future studies to examine if the results agree with those obtained herein. We could not establish a negative predictive value, as the CARS-BR was not applied to the control group. We know that it would be needed to validate the IRDI questionnaire, and we suggest that it be calculated in future research using the instrument.

Similarly, international literature has been indicating that instruments able to differentiate children with autism spectrum disorders from those with mental retardation and delayed language development have greater strength than those that differentiate only between children with autism spectrum disorders and children with typical development\(^{8,29,30}\).

In this study, the IRDI questionnaire showed good results in terms of applicability and sensitivity levels, inaugurating a process of validation and refinement of this instrument.

It should be stressed that the use of the IRDI questionnaire by the speech therapist or other health professionals does not dismiss a clinical diagnosis. It only enables the fulfillment of the purpose of the instrument, which is to identify, raise suspicion, and screen for possible cases. It is worth remembering that the diagnosis, in turn, should be performed by qualified and trained professionals (psychologists and neurologists, or psychiatrists).

**CONCLUSION**

The evaluation of the sensitivity of the IRDI questionnaire for identifying autism spectrum disorders showed good performance levels based on the established cutoff point in the study, making it useful for this purpose. Furthermore, it was easy to apply, quick to complete, and inexpensive.

We note, however, the need for future studies to effectively validate the instrument, which would allow more effective referrals for assessment, diagnosis, and treatment. The complementary use of two assessment tools to ensure better conditions for referral is recommended.

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