

CLINICAL SCIENCE

Development and analysis of a low-cost screening tool to identify and classify hearing loss in children: a proposal for developing countries

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OBJECTIVES: A lack of attention has been given to hearing health in primary care in developing countries. A strategy involving low-cost screening tools may fill the current gap in hearing health care provided to children. Therefore, it is necessary to establish and adopt lower-cost procedures that are accessible to underserved areas that lack other physical or human resources that would enable the identification of groups at risk for hearing loss. The aim of this study was to develop and analyze the efficacy of a low-cost screening tool to identify and classify hearing loss in children.

METHODS: A total of 214 2-to-10 year-old children participated in this study. The study was conducted by providing a questionnaire to the parents and comparing the answers with the results of a complete audiological assessment. Receiver operating characteristic (ROC) curves were constructed, and discriminant analysis techniques were used to classify each child based on the total score.

RESULTS: We found conductive hearing loss in 39.3% of children, sensorineural hearing loss in 7.4% and normal hearing in 53.3%. The discriminant analysis technique provided the following classification rule for the total score on the questionnaire: 0 to 4 points – normal hearing; 5 to 7 points – conductive hearing loss; over 7 points – sensorineural hearing loss.

CONCLUSION: Our results suggest that the questionnaire could be used as a screening tool to classify children with normal hearing or hearing loss and according to the type of hearing loss based on the total questionnaire score.

KEYWORDS: Questionnaire; Hearing loss; Children; Low-cost screening; Hearing.

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INTRODUCTION

The prevalence of permanent mild or more severe hearing loss is estimated at one in every ten people.¹ In Brazil, many localities already require newborn hearing screening by law, but only in a few cities. However, because newborn hearing screening is not universally implemented in many areas,² permanent congenital and early-onset hearing loss in children may be detected late in developing countries, with the mean age varying from approximately 2 to 7 years.³

Otitis media is one of the most common disorders in childhood, and approximately 80% of school-age children suffer from temporary hearing loss every year.⁴

Chronic otitis media is considered the main cause of mild-to-moderate hearing loss.⁵⁻⁷ The prevalence ranges from 1% to 46% around the world and is higher in developing countries. This condition is considered by the World Health Organization to be a public health problem.^{5,8}

Children with slight/mild hearing loss (<40 dBHL in one or both ears) are an underreported and understudied group. The majority of studies have focused on children with higher levels of hearing loss,⁹ although studies have shown that even slight/mild hearing loss can result in negative consequences for the biopsychosocial development of children.^{9,10}

In Brazil, primary care does not address hearing health or even recognize this demand, which is partially due to the limited number of health professionals in this area.¹ Moreover, the territorial extension, the cost of equipment and a lack of human resources make it difficult to implement such programs. The Family Health Strategy Program has been growing in Brazil, but it is still lacking with regard to actions to address hearing health in primary

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care. Development of a strategy that involves low-cost screening may help fill the current gap in the care of children with hearing problems in developing countries. Therefore, it is necessary to establish and adopt lower-cost procedures, such as the use of screening and/or questionnaires, that are accessible to underserved areas of the country that lack the physical or human resources necessary to identify groups of children who are at risk for hearing loss as early as possible.¹¹

Although several studies on this topic have been conducted, many have neglected to consider slight or mild and/or unilateral hearing loss. Furthermore, most of the previous studies have been conducted using low-cost instruments and have failed to classify hearing loss by type. Therefore, the aim of the present study was to develop and analyze the efficacy of a low-cost screening tool to identify and classify hearing loss in children through comparison with the results of a complete audiological assessment.

MATERIALS AND METHODS

This study was carried out in the Hearing Health Primary Care Laboratory with the approval of the São Paulo University School of Medicine Ethics Committee (Protocol # 778/08). All participants provided written informed consent to participate in the study.

Study population

We carried out a hearing health survey among children who were 2 to 10 years of age in the District of Butantã, which is located in the western region of São Paulo, Brazil. The study area comprised São Remo community, a slum area with approximately 10,000 inhabitants attended by a public primary health care unit staffed by health community agents included in the Family Health Program (more than 2,000 indexed families). Children 2 to 10 years old constitute approximately 18% of this population, but there is no established hearing health care program despite a federal law guaranteeing people's access to all levels of hearing care. Universal newborn hearing screening and hearing screening in schools are not current practices in this region or across the country, and therefore, late identification of hearing loss in children is common.

Children who met the inclusion criteria and were between the ages of 2 and 10 years old (the age criterion applied was selected based on the United Nations' definition of the beginning of adolescence, i.e., only children up to 10 years of age were included) were invited to participate in this study. The children were evaluated in their family homes, schools and kindergartens in the region, as well as at the health unit. A total of 214 2-10 year-old children of both genders took part in this study.

Questionnaire

To investigate the problem (late identification of hearing loss in children), we developed a questionnaire to identify children at risk for hearing loss (Appendix 1) with questions regarding health history (pre-, peri-, and post-natal), development, communication skills, and hearing complaints based on previous investigations.^{3,12-16} This questionnaire was validated in the present investigation and will be applied by health community agents of the family health program in the future to expand access to hearing health primary care services. This questionnaire will also be used

as a preliminary screening tool to decrease the number of audiological assessments that are necessary in Brazil.

Parents answered the questionnaire (Appendix 1), and each answer represented a risk factor for hearing loss. For each subject, every factor that was present was scored with a 1. The score for each question and the total score were calculated.

Prior to administering the questionnaires and conducting the other procedures, we studied the questionnaire test-retest reproducibility. For this stage, 20 parents of children belonging to the study area and not participating in the other procedures (corresponding to approximately 10% of the total sample) answered the questionnaire twice at an interval of three months. The questionnaire was applied by one researcher the first time and by another the second time. The aim of this step was to verify the clarity and replicability of the answers. The intraclass correlation coefficient showed moderate concordance (0.7) between the values of the total score in the two applications of the questionnaire, suggesting that the understanding and clarity of the questions was suitable.

Procedures

This study was double-blind, as different evaluators performed the questionnaire and the audiological assessment; the parents only knew the results at the end of the investigation.

An audiological assessment was conducted on all participants. Following otoscopic inspection, tympanometry was carried out using a GSI 38 Auto Tymp (Grason-Stadler). An audiometric evaluation for pure-tone thresholds was carried out with an Itera II Clinical Audiometer (Madsen) using standard audiometric techniques in a sound-attenuated testing room (from 250 to 8000 Hz). These results were compared with the questionnaire scores, establishing the sensitivity and specificity of this tool.

The results of the audiological assessment were considered normal when hearing the thresholds were normal, i.e., ≤ 15 dBHL, and when tympanometry presented a type-A curve and acoustic reflexes were detected in both ears. Hearing loss was indicated when the pure tone thresholds were elevated^{17,18} (> 15 dB HL), when the tympanogram was type B, C, As, or Ad and/or when there was an absence of acoustic reflexes with one or both ears.

Statistical analyses

The statistical analyses were performed using SPSS 15.0 for Windows. The Kolmogorov-Smirnov test was applied to determine the normality of the variable distribution. To assess the existence of an association between hearing status and gender and between hearing status and age group, the chi-square test (and Fisher's exact test, when appropriate) was used. The distribution of the total score on the questionnaire was compared between children with normal hearing and those with hearing loss and between children with normal hearing, children with conductive hearing loss and children with sensorineural hearing loss; the results were evaluated using the Kruskal-Wallis test. When necessary, the Bonferroni procedure was used to identify differences between the distributions. An ROC curve was constructed to establish a cutoff value of the total score that would classify a child as either having normal hearing or affected by hearing loss based on the questionnaire. The discriminant analysis technique was used to classify a child

in one of three categories of hearing (normal hearing, conductive hearing loss, and sensorineural hearing loss) based on the total score. The level of significance was set at 0.05.

RESULTS

Characterization of the sample

Girls composed 40.7% of the sample, and boys represented 59.3%. There was a significantly greater number of boys participating ($p < 0.01$).

Of these 214 children, 114 (53.3%) had normal hearing and 100 (46.7%) had hearing loss (Table 1). When the type of hearing loss was considered, 84 (39.3%) had conductive hearing loss (slight-mild to moderate), and 16 (7.4%) had sensorineural hearing loss (severe to profound).

Questionnaire scores (by age and type of hearing loss)

Children in the hearing loss category presented higher scores than children in the normal category (Table 1). Furthermore, the total score values were higher in the sensorineural hearing loss group than in the conductive hearing loss group ($p < 0.001$) and the normal group ($p < 0.001$) (Table 2).

Cutoff scores of the questionnaire

To determine a cutoff value of the total score for ranking the results of the audiological assessment in normal children or children with hearing loss, ROC analysis was performed (Figure 1). The nearest point on the upper left corner of the curve represents the maximum sensitivity and specificity (0.44 sensitivity and 0.87 specificity). The area under the curve was 0.72, indicating good discriminatory power of the total score. Thus, the point that simultaneously provides the highest sensitivity and specificity corresponds to 5.5. Therefore, the classification rule was established as follows:

- An individual could be classified as having "hearing loss" when the total score was greater than or equal to 6;
- An individual could be classified as having "normal hearing" when the total score was less than 6.

A rule was also established to classify each subject as having normal hearing, conductive hearing loss or sensorineural hearing loss, and this rule was also based on the cutoff values of the total score. Table 2 shows that the total score distribution was not the same in the three groups (normal, conductive hearing loss and sensorineural hearing loss). Thus, the technique of discriminant analysis yielded the following classification rule:

- Total score from 0 to 4: normal hearing;
- Total score from 5 to 7: conductive hearing loss;
- Total score from 8 to 14: sensorineural hearing loss.

Based on the rule described above, without regard to questions 3 and 4 on the questionnaire, which refer to prior

Table 1 - Descriptive statistics for the total score in the questionnaire in both categories of audiological assessment (normal and hearing loss).

Children	Categories	n	Mean	Standard deviation	p-value
2-10 years	Normal	114	3.8	1.9	<0.001
	Hearing loss	100	6.1	3.0	
	Total	214	4.9	2.7	

Table 2 - Statistics describing the total scores for the questionnaire based on the type of hearing loss or normal audiological assessment.

Children	Type	n	Mean	Standard Deviation
2-10 years	Normal	114	3.8	1.9
	Conductive	84	5.2	2.3
	Sensorineural	16	10.6	1.6
	Total	214	4.9	2.7

hearing assessment, the questionnaire correctly classified 85% of children with normal hearing, 68% of those with conductive hearing loss and all of the subjects with sensorineural hearing loss.

DISCUSSION

This study developed a low-cost screening tool to identify hearing loss in children in developing countries that may be used by community agents of the Family Health Program in the future. Furthermore, we analyzed the efficacy of this tool when compared to the results of an audiological assessment. Our findings suggest that this screening tool could be used to classify children as having normal hearing or hearing loss and to identify the type of hearing alteration (conductive or sensorineural hearing loss) based on the scores of the questionnaire.

Characterization of the sample

In the studied sample, we observed a large number of cases of slight or mild-to-moderate conductive hearing loss (39.3%) and severe-to-profound sensorineural hearing loss (7.4%). This prevalence of hearing loss is consistent with other studies that have found high rates of hearing loss in "disadvantaged" groups in developing countries,^{1,3,5,19} such as the population studied here. Furthermore, Olusanya³ pointed out that several studies have shown a significant prevalence of hearing disorders in children in developing countries, but the authors did not consider the differences in the test protocols employed. The prevalence of hearing loss in developing countries varies from approximately 4% to 25%, with the middle ear disorders ranging from 7% to 36%, which is consistent with the findings of our study.

The largest number of slight-to-mild conductive hearing loss observed in our study is consistent with a previous study,²⁰ which found a lower prevalence of hearing loss in a population of children aged 6 to 12 years. It is known that the prevalence of otitis media is highest in children between 2 and 5 years of age,^{21,22} which coincides with the age range investigated here. Above the age of 5, the prevalence of otitis media decreases, and this may explain the low prevalence reported in this investigation.²⁰

As seen from our results, there is great variability regarding the prevalence of hearing loss reported in various developing countries. This variability is probably due to the different screening procedures and criteria for hearing loss, as well as socioeconomic, health, and other intrinsic and extrinsic factors of each population, which often are not directly related to hearing loss.²⁰

It is noteworthy that the prevalence of hearing loss in this study cannot be extrapolated to the general population because the research was developed within a specific district of São Paulo, Brazil. Furthermore, not all of those

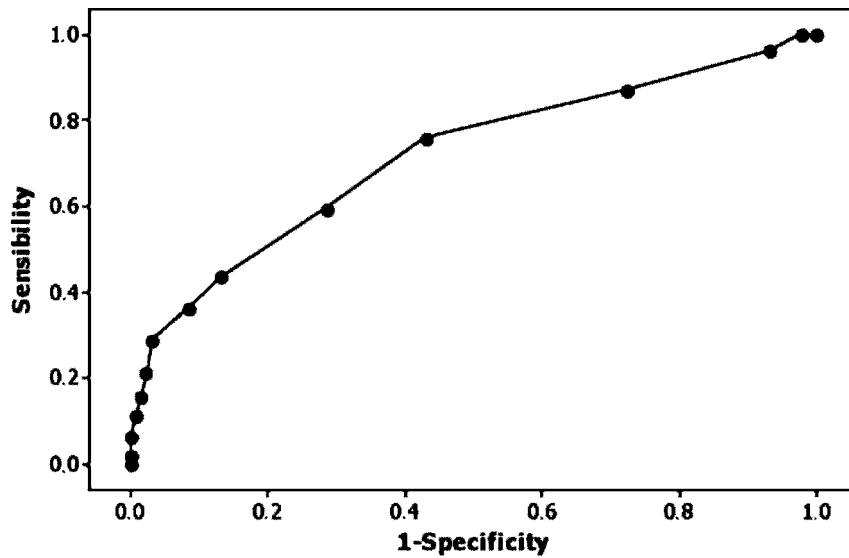


Figure 1 - ROC Curve.

invited to participate in the study attended the audiological assessment, which may constitute a limitation of this study; it can be hypothesized that the parents of children suspected of suffering from hearing loss were more willing to participate in the study, and thus the true prevalence of hearing loss could be somewhat lower. However, identification of hearing loss prevalence in the general population was not a goal of the present study.

Scores on the questionnaire (by age and type of hearing loss)

The difference between the scores of children with normal hearing or hearing loss was evident among the children in this study (2-10 years of age). We also detected differences between the scores for children with normal hearing, those with conductive hearing loss and those with sensorineural hearing loss. Thus, we verified that children with sensorineural hearing loss presented significantly higher scores than children with conductive hearing loss, who in turn presented significantly higher scores than children with normal hearing.

Cutoff scores of the questionnaire

Based on the ROC curve, we established a cutoff value that simultaneously provided the highest sensitivity and specificity (5.5 points) for the total score of the questionnaire. This analysis indicated that children who obtained a total score greater than or equal to 6 were more likely to have hearing loss detected at the audiological assessment. Meanwhile, children with a score lower than 6 are more likely to have normal hearing based on the audiological evaluation.

For this cutoff value (5.5 points), the sensitivity of the questionnaire was 44% and the specificity was 87%, which are similar to values obtained in other studies^{12,16} that used questionnaires as a form of screening. However, our results differ from the findings of one investigation,¹⁵ which obtained the highest values of sensitivity. This was because the authors considered only hearing loss greater than 40 dBHL, whereas in the present study, we also considered slight and mild hearing losses, expanding the range of hearing loss involved.

Slight and mild hearing losses are the most difficult to detect and are often overlooked in studies of hearing health.

Taha et al.²⁰ emphasized the use of questionnaires as a method of screening to address parents' concerns regarding hearing loss, although previous studies have not reached a consensus on this subject because of variations in the sensitivity of the proposed instruments. Furthermore, the authors stressed the importance of further investigations on the cost-effectiveness of using these screening tools in some communities. They reported that instruments based on information collected from parents can improve parents' awareness about hearing loss and may increase parents' motivation to access the appropriate hearing health care resources.

Our analysis of the total scores for the questionnaire also allowed for a subdivision of the subjects based on the results of their audiological hearing loss assessment: a total score of 0 to 4 points indicated a higher chance of having normal hearing, 5-7 points a higher chance of having conductive hearing loss, and more than 7 points a greater likelihood of sensorineural hearing loss. According to this classification, the questionnaire correctly classified 85% of children with normal hearing, 68% of those with conductive hearing loss and all children with sensorineural hearing loss.

It is important to emphasize that this kind of classification based on a low-cost tool that can distinguish children with normal hearing from those with hearing loss and can differentiate between different types of hearing loss (conductive or sensorineural hearing loss) represents a novel contribution that may improve health care in developing countries by reducing the number of referrals to specialized services. Such specialized services are often inadequate, especially in regions far from major urban centers and at the periphery of large Brazilian cities.²

It is important to note that all children with sensorineural hearing loss were classified correctly, indicating that, at least for this type, this low-cost instrument is effective. With regard to conductive hearing loss, the losses are usually intermittent and are often slight or mild and/or unilateral, making it harder for parents to recognize the factors presented in the questionnaire. This

may explain the low level of agreement between the questionnaire data and the audiological assessment in these cases.^{3,15,16,19,21,22} However, the instrument showed a high sensitivity concerning the identification of children with normal hearing.

The lower sensitivity of the method of assessing hearing loss by questionnaire in general for slight or mild hearing loss as well as conductive hearing loss has been previously reported.²⁵ The authors indicated that children with slight or mild hearing loss were not significantly different with respect to the measures of speech, language, reading, academic performance, behavior, or quality of life when compared to children with normal hearing, and this could explain why parents did not suspect hearing loss in these cases. This may also account for the higher sensitivity of the questionnaire for sensorineural hearing loss in our study, as these children presented severe-to-profound hearing loss and therefore may also present the largest deficits in development; this could facilitate the parents' ability to recognize the signs of hearing loss.

With respect to the reproducibility of the results from the questionnaire, we administered the questionnaire to a group of test subjects twice at an interval of three months. It is possible that the interval between the two applications should be shorter. Over a period of three months, it is possible that health problems could arise that could affect answers given on the questionnaire. This would reduce the level of agreement between the answers given at the two time points. For future studies aiming to evaluate reproducibility of this type of questionnaire, we suggest an interval of 15 days between applications²⁶.

Although the instrument requires some adjustments to increase its sensitivity to conductive hearing loss, we believe it can be introduced into the practice of the community agents of the Family Health Program and the nurses and pediatricians in primary health care as a means of initial screening. This would allow the detection of possible hearing loss and also serve to attract the attention of parents and health professionals to the issue of hearing care, as it is known that hearing loss, even slight or mild, can have important negative consequences for children and society.^{19,20,25,27}

Actions that improve primary health care are important to enhance this level of attention and prioritize the resources according to the needs of each country, as has been stated by the World Health Assembly (1995) and other authors.^{3,5,19,20,29} It should be emphasized that hearing loss presents an additional and unique challenge because of its often invisible nature. Therefore, the efforts of countries to create effective prevention programs are essential,^{19,28} and these programs are complemented by early hearing detection and intervention for children (infants, preschool, and school-aged children).²⁹

Our results suggest that the questionnaire we developed could be used as a screening tool to classify children as having normal hearing or hearing loss as well as to distinguish the type of hearing loss (conductive or sensorineural).

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AUTHOR CONTRIBUTIONS

Samelli AG conceived and designed the study, was responsible for the study material and recruitment of subjects, collection and assembly of data, data analysis and interpretation, manuscript writing and final approval of the manuscript. Rabelo CM was responsible for the collection and assembly of data, data analysis and interpretation, manuscript writing, and final approval of the manuscript. Vespasiano APC was responsible for the collection of data, and final approval of the manuscript.

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APPENDIX 1 - Questionnaire to identify the risk for hearing loss (children 2-10 years of age).

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1. Identification
 2. Birth locality (maternity/city)
 3. Did the child undergo any hearing screening at birth?
 - Yes
 - No (1 point)
 4. Has the child ever undergone a hearing test?
 - Yes
 - No (1 point)
 5. Were any risk indicators for hearing loss present?[14] – All risks are enumerated.
 - Yes (1 point)
 - No
 6. In what position was the child breastfed? (to describe)
 - Lying down (1 point)
 - Sitting
 7. Family history: Is there any history of deafness in your family? Who?
 - Yes (1 point)
 - No
 8. Has the child had any health problems? (Some diseases are enumerated: meningitis, brain injury, frequent otitis media or ear infection).
 - Yes (1 point)
 - No
 9. Does the child pay attention to loud noises?
 - Yes
 - No (1 point)
 10. Does the child pay attention when he/she is called by name?
 - Yes
 - No (1 point)
 11. Does he/she require a gesture or a voice used at high intensity to understand?
 - Yes (1 point)
 - No
 12. Does your child hear as well as other children of the same age?
 - Yes
 - No (1 point)
 13. Does your child speak as well as other children of the same age?
 - Yes
 - No (1 point)
 14. Does your child understand orders, even if he/she is not looking at the speaker? For example: Bring the spoon to your mother (without pointing to the object)?
 - Yes
 - No (1 point)
 15. Does your child like music?
 - Yes
 - No (1 point)
 16. Has anyone commented that your son/daughter does not hear well or that his/her speech is very bad?
 - Yes (1 point)
 - No
-