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Neonatal hearing screening in the process of diagnosis and hearing rehabilitation

A triagem auditiva neonatal no processo de diagnóstico e reabilitação auditiva

El examen de audición neonatal en el proceso de diagnóstico y rehabilitación de audición

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Abstract

Introduction: neonatal hearing screening aims the early identification of hearing loss. Despite the existence of Laws regarding it, there are still several cases of notable delays in the access of hearing loss services for diagnosis and intervention. **Objective:** describe the role of neonatal hearing screening in the process of diagnosis and hearing rehabilitation of zero to seven years old children attended in an Ambulatorial Service of Hearing Health on the North Coast of Santa Catarina.. **Methodology:** use of a questionnaire with 30 parents or legal responsible for children with hearing loss, using descriptive and inferential statistic as method of analysis. **Findings:** considering the 30 children, 86,66% underwent neonatal hearing screening, from which 96,15% had the first test done and 34,62% had the retest done during their first month of life; 15,38% had a diagnosis until they were three months old and 6,66% the intervention before becoming 6 months old. 16,66% of monitoring and intervention were identified regarding basic care, reinforcing the importance of hearing screening as a way of tracking and identification. **Conclusion:** the results indicate that the accomplishment of neonatal hearing screening has an important role for the access of an early hearing loss diagnosis, although the ideal ages for the diagnosis and for intervention are fall short from the expected. The importance of basic care capacitation is emphasized, in order to have efficient monitoring and support for children with risk or confirmed hearing loss.

Keywords: Neonatal Screening; Hearing Loss; Early Diagnosis; Public Health.

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Authors' contributions:
DFP: study design and preparation, methodology, critical review and guidance.
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Resumo

Introdução: a triagem auditiva neonatal objetiva a identificação precoce de perdas auditivas. Apesar da existência de leis, ainda se percebe diversos casos de atrasos no acesso aos serviços de diagnóstico e intervenção das perdas auditivas. **Objetivo:** descrever o papel da triagem auditiva neonatal no processo de diagnóstico e reabilitação auditiva de crianças de zero a sete anos atendidas em um Serviço Ambulatorial de Saúde Auditiva do litoral norte catarinense. **Metodologia:** aplicação de questionário com 30 pais/responsáveis de crianças com perda auditiva, sendo aplicada análise estatística descritiva e inferencial. **Resultados:** das 30 crianças, 86,66% realizaram a triagem auditiva neonatal, das quais 96,15% realizaram a primeira testagem e 34,62% o reteste dentro do primeiro mês de vida; 15,38% realizaram o diagnóstico até os três meses e 6,66%, a intervenção até os seis meses. Foi identificado 16,66% de acompanhamento e investigação na atenção básica, reforçando a importância da realização da triagem como forma de rastreio e identificação. **Conclusão:** os resultados indicam que a realização da triagem auditiva neonatal tem papel importante para o acesso ao diagnóstico precoce da perda auditiva, embora as idades idealizadas para o diagnóstico e a intervenção estejam aquém do esperado. Destaca-se a importância de capacitar a atenção básica para o suporte e o acompanhamento de crianças de risco ou com perda auditiva confirmada.

Palavras-chave: Triagem Neonatal; Perda Auditiva; Diagnóstico Precoce; Saúde Pública.

Resumen

Introducción: el examen de audición neonatal apunta a la identificación temprana de la pérdida auditiva. A pesar de la existencia de leyes, todavía hay varios casos de demoras en el acceso a los servicios de diagnóstico y la intervención de pérdida auditiva. **Objetivo:** describir el papel de la detección auditiva neonatal en el proceso de diagnóstico auditivo y rehabilitación de niños de cero a siete años que asisten a un Servicio de Salud Auditiva Ambulatoria en la costa norte de Santa Catarina. **Metodología:** aplicación de un cuestionario con 30 padres / tutores de niños con pérdida auditiva, aplicando análisis estadísticos descriptivos e inferenciales. **Resultados:** el 86,66% de los 30 niños se sometieron a un examen de audición neonatal, de los cuales el 96,15% realizó la primera prueba y el 34,62% volvió a realizar la prueba dentro del primer mes de vida; el 15,38% realizó el diagnóstico hasta tres meses y el 6,66%, la intervención hasta seis meses. Se identificó 16,66% seguimiento e investigación en atención primaria, lo que refuerza la importancia de la detección como un medio de detección e identificación. **Conclusión:** los resultados indican que el examen de audición neonatal desempeña un papel importante en el diagnóstico precoz de la pérdida auditiva, aunque las edades idealizadas para el diagnóstico y la intervención están por debajo de las expectativas. La importância de habilitar la atención primaria para el apoyo y el seguimiento de los niños en riesgo o con pérdida auditiva confirmada.

Palabras clave: Tamizaje Neonatal; Pérdida Auditiva; Diagnóstico Precoz; Salud Pública.

Introduction

The development of hearing skills is a complex process that begins in intrauterine life. Although the cochlea is already fully functional at birth, the central auditory system is still quite immature, while the central auditory pathways continue to develop during childhood and adolescence. Thus, hearing loss (HL) in childhood can cause morphofunctional changes in central auditory system neurons[1].

The prevalence of HL in neonates ranges from 0.1-0.6% in low-risk infants, increasing to 1-4% when the risks for HL are present[2]. In addition, the HL may cause impairment in different areas of the child's life, such as language development, speech, educational or emotional changes[3].

Studies suggest that, even after the onset of hearing loss, if stimuli are properly reintroduced, especially in the first six months of life, these can provide a development closer to that of their hearing peers when compared to cases of late HL diagnosis[4,5].

That said, the importance of monitoring the development of hearing skills should be highlighted, in addition to the early diagnosis and intervention of hearing changes and losses, as regulated by

the Guidelines for Attention for Neonatal Hearing Screening (NHS) of the Brazilian Ministry of Health[4,5,6].

The Guidelines of the Brazilian Ministry of Health (MH) were published in 2012, after the publication of Law No. 12,303 of August 2, 2010, which established the mandatory free performance of the Evoked Otoacoustic Emissions (OAE) test, commonly known as "baby hearing screening", in all hospitals and maternity hospitals in the country for children born in its facilities[7,2].

Designed from a joint action of several areas of public health and related bodies, the MH Guidelines were based on the definitions and recommendations of publications made by national and international reference institutions on hearing health, such as the Multiprofessional Committee on Auditory Health (COMUSA) and the Joint Committee on Infant Hearing (JCIH)[2,8,9].

The MH Guidelines and the JCIH recommend that the NHS should be performed in the first month of life and that the diagnosis of children who fail (in the test and retest) should be determined within three months. In addition, the intervention must start until six months of age in 90% of cases of HL[2,8].

With regard to the flow of care, the MH Guidelines indicate that the attention to children's hearing health should be distributed at different levels of health care, with primary care being responsible for the proper referral to specialized centers[2,6].

Primary care should monitor and refer children with Risk Indicators for Hearing Loss (RIHL) for diagnosis, as well as those who show less than expected development and those whose parents have suspected HL[2].

It is noteworthy that despite the constant efforts and legislation, the NHS coverage in Brazil is still low (37.2%)[10,11], thus resulting in a late diagnosis and initiation of intervention for most neonates with HL[6,12].

Therefore, this study aimed to describe the role of the NHS in the process of diagnosis and hearing rehabilitation of children from birth to seven years of age attended at a Hearing Health Outpatient Clinic (SASA) on the Northern coast of Santa Catarina.

Methodology

This was a cross-sectional and quantitative study and was carried out with parents/guardians of children attended at the SASA of the Universidade do Vale do Itajaí (UNIVALI), upon approval by the Research Ethics Committee of UNIVALI under consubstantiated opinion No. 3.086.560 of December 16, 2018.

The sample size was calculated for a cross-sectional study, with a significance level of 95%, a finite population of 129 children who would be in the age group from 0-7 years old, from March to July 2019 with a diagnosis of HL and users of hearing aids (Personal Sound Amplification Product - PSAP, or cochlear implants - IC) attended at the study center. The sample size ratio was 1, with a percentage of non-exposed positives (children at low risk for HL) of 0.3% and exposed positives of 1.6% (children at high risk)[13].

The study included 30 parents/guardians of children who attended for follow-up at the study center from February to August 2019 and who agreed to participate in the study by signing the Informed Consent Form (ICF). Study participants included 20 mothers, 7 fathers and 3 grandparents/great-grandparents. Among them, seven had complete or incomplete primary school; 17 had complete or incomplete high school; four had complete or incomplete higher education and two had graduate degrees.

The children whose parents/guardians were interviewed had the following characteristics: 5 children from zero to three years old, being 4 (13.33%) females and 1 (3.33%) male; and 25 children from three to seven years old, being 12 (40%) females and 13 (43.33%) males.

The interview with parents/guardians was carried out at the study center and included the application of a questionnaire that was developed by the researchers and the study supervisor. When the information provided by the parents/guardians was not sufficient to answer the variable studied, the researchers consulted the medical records at the SASA or the child's health booklet, as authorized and described in the ICF.

The questionnaire consisted of 30 closed questions, which were designed by the researchers from reading the MH Guidelines[2], the Guidelines for Hearing Health Care in the health care network for people with disabilities in Santa Catarina[14]the

JCIH recommendations[8]; as well as from reading of articles related to the topic and from the experience of the researchers regarding the assessment and diagnosis of children with HL at a SASA.

The study variables were related to age; risk for HL at birth; performance and importance of NHS; age at diagnosis of HL; ways to start the care process at the SASA; difficulties faced in the access to the SASA; waiting time; time elapsed between the first consultation at the SASA and the audiological diagnosis, and between the diagnosis and the intervention; family's reaction to the diagnosis of HL; hearing and language follow-up at the Primary Health Unit (UBS); type and degree of hearing loss and type of hearing aids.

Data were organized in an Excel® spreadsheet, and the information was processed and analyzed using the SPSS Statistics 21.0 (Chicago, IL 60606, USA). The data obtained were submitted to descriptive and inferential statistical analysis using the Fisher's exact test. A significance level of 5% ($p<0.005$) was adopted for all analyzes.

Results

As a starting point, participants were asked about the performance of the NHS. According to the report of the parents/guardians, 3 (10%) children did not perform the NHS, 1 (3.33%) did not know how to report and 26 (86.66%) reported that the child had performed the NHS.

Of these 26 who performed the NHS, 25 (96.15%) underwent the first test within the first month of life, and 18 (69.23%) underwent the test before hospital discharge. Retest was also performed in the first month of life in 9 (34.62%) children; between 1-3 months in 6 (23.08%) and between 3-6 months in 2 (7.69%). Nine (34.61%) children did not undergo the retest, as they had passed the initial test.

As for the procedures used in the NHS, on the one hand, it was found that the test was performed by OAE in 13 (50%) cases. However, on the other hand, this information could be obtained neither from the parents nor from the child's medical record in the SASA in the other 13 (50%) cases.

Regarding the results of the test and retest of the 26 children who underwent the test, 9 (34.61%) passed and 17 (65.39%) failed. Among those who failed, 2 (11.76%) passed the retest, 14 (82.35%) failed the retest and 1 (5.89%) did not know how to report the result.

All (100%) parents/guardians answered that they would have performed the NHS even if it were optional. However, there was no unanimity about its importance for the final diagnosis of HL: among the 26 parents/guardians who reported having performed the NHS, 20 (76.92%) agree that it was important for the audiological diagnosis, while 6 (23, 08%) disagree.

Of these six parents/guardians who reported not believing that NHS was important for the diagnosis, in 4 (66.67%) cases the child had passed the NHS and in 1 case the child had passed the retest. The other parent/guardian who reported not believing that NHS was important in the diagnostic process of the HL, even with his child having failed the test and retest, reported that although sad, he understood and was concerned on what would be best for the child from the diagnosis of HL.

Regarding the way that the care process was initiated at the study center, 10 (33.33%) children were referred through the NHS and 20 (66.67%) by other means. Of these, 16 (80%) were referred from public services (35% from primary care, 30% from specialized care, 5% from hospitals and 10% from other sources) and 4 (20%) from private care.

Regarding the presence of RIHL, according to the medical records and children's health booklet, it was found that 19 (63.34%) had a risk indicator, 10 (33.33%) did not have a risk indicator and 1 (3.33%) was not reported. Two of the 19 children who were at risk for HL had more than one indicator. Table 1 shows the risk indicators, considering that two children had more than one risk indicator.

Table 1. Risk indicators for hearing loss in the clinical history of children whose parents/guardians were interviewed (n=22).

Risk factor	Prevalence
ICU* stay for more than 5 days	6 (27.27%)
Craniofacial anomalies	4 (18.18%)
Family history of permanent deafness	4 (18.18%)
Genetic syndromes	3 (13.64%)
Postnatal bacterial or viral infections	2 (9.09%)
Congenital infections	1 (4.55%)
Other (pre-eclampsia**)	2 (9.09%)
TOTAL	22 (100%)

Legend: ICU=Intensive Care Unit; **Pre-eclampsia – although not reported as a RIHL in the literature (JCIH, 2007)8, it was listed as a possible cause for HL in the otorhinolaryngological assessment records.

Staying in the Neonatal Intensive Care Unit (NICU) was the most common RIHL in the studied children. With respect to the results of the NHS, 8 children had passed the NHS and had RIHL, of which 2 had genetic syndromes (biliary atresia and Pierre-Robin), 2 had previous meningitis, 2 were from the NICU, 1 had an external ear malformation (microtia) and 1 had a history of rubella and cytomegalovirus.

Of the 19 children who had RIHL, 17 underwent the NHS. According to the child's medical record or health booklet, 47.06% of these 17 children underwent the NHS through the OAE.

The audiological diagnosis for children who underwent NHS was performed between one and three months for 4 (15.38%) children; between three and six months to 6 (23.08%); between six and 12 months for 4 (15.38%); between 18 and 24 months for 2 (7.69%) and 10 (38.47%) children were diagnosed with hearing loss only after 24 months of age. In this regard, all children (4) that the parents/guardians reported that they did not perform the NHS, or did not know how to report their performance, had the diagnosis after 24 months of age.

When the 20 parents/guardians who felt that the NHS was important for the diagnosis were asked about the family's reaction to the diagnosis, 15 (75%) reported that, although sad, they understood the diagnosis of HL and were concerned; 2 (10%) were already waiting for the confirmation of a HL, 2 (10%) believed that the diagnosis of HL was incorrect, even with the previous change in the NHS and 1 (5%) reported that the diagnosis of HL "was peaceful" .

Table 2 shows the results regarding the age at the first test, retest, diagnosis and the beginning of the intervention, regardless of the place where it was performed, according to the recommendations in the literature.

Table 2. Distribution of children according to the age at the first test, retest, diagnosis and beginning of the intervention, according to the recommendations in the literature (n=30).

Recommended age in months (JCIH, 2007)	Yes	No	Did not perform/ did not know how to report	p-value
1st test (< 1)	25 (83.34%)	1 (3.33%)	4 (13.33%)	--
Retest (< 1)	9 (30.00%)	17 (56.67%)	4 (13.33%)	--
Diagnosis (< 3)	4 (13.33%)	26 (86.67%)	--	0.0001
Start of intervention (< 6)	2 (6.66%)	25 (83.34%)	3 (10%)	--

Legend: JCIH=Joint Committee on Infant Hearing; <: less or equal; Fisher's exact test was used. p<0.005=significant.

Table 2 shows that the majority of children (83.34%) performed the NHS within the period recommended in the literature[2,8], contrasted to retest (56.67%), diagnosis (86.67%) and the beginning of the intervention (83.34%), which did not follow the recommendations of the literature[2,8].

Table 3 shows the comparative results of the NHS performance in the period recommended by the literature in relation to the period recommended for the audiological diagnosis.

Table 3. Comparison of the age at which the NHS was performed with relation to the age at diagnosis, following the recommendations in the literature (n=30).

Age at the recommended NHS[2,8]	Age at the recommended diagnosis[2,8]	
	Follows the recommendation	Do not follow the recommendation
Follows the recommendation	4 (13.33%)	21 (70%)*
Do not follow the recommendation	--	5 (16.67%)

Legend: NHS=Neonatal Hearing Screening; *Fisher's exact test was used. p<0.005=significant.

In turn, Table 3 shows that, regardless of whether or not they performed the NHS following the recommendations of the literature[2,8], most children had their diagnosis determined beyond the recommended period[2,8]. However, children who underwent NHS were diagnosed earlier than those who did not.

Nine (30%) of the 30 research participants at the study center were referred with a previous diagnosis of HL, of which three had already undergone some type of intervention. As for the children who were diagnosed with HL at the study center, the average age at diagnosis was 31 months and the average age at the beginning of the intervention was 34 months.

Table 4 shows the comparative results between the findings in the NHS test (n=30) and retest (n=17) with the occurrence of unilateral or bilateral HL.

Table 4. Comparative results between the findings in the NHS test (n=30) and retest (n=17) with the occurrence of unilateral or bilateral hearing loss.

Result	HL	
	Bilateral	Unilateral
Approved at NHS	9 (30%)	--
Failed at NHS and approved at retest	1 (3.33%)	1 (3.33%)
Failed at NHS and failed at retest	14 (46.67%)	--
Failed at NHS and did not report the result of the retest	1 (3.33%)	--
Did not perform or did not know how to report	3 (10%)	1 (3.33%)
TOTAL	28 (93.33%)	2 (6.67%)

Legend: NHS=Neonatal Hearing Screening

Table 4 also shows that, considering the best ear, 28 (93.33%) of the children had bilateral HL after testing and retesting and only 2 (6.67%) had unilateral HL. In addition, despite passing the first test, 9 (30%) children were diagnosed with HL.

In turn, Table 5 shows the comparative results between the findings in the NHS test and retest with the type of hearing loss in the best ear.

Table 5. Comparative results between the findings in the NHS test (n=30) and retest (n=17) with the type of hearing loss in the best ear.

Result	Type of hearing loss			
	No HL	Conductive	Sensorineural	Mixed
Approved at NHS	--	1 (3.33%)	7 (23.33%)	1 (3.33%)
Failed at NHS and approved at retest	1 (3.33%)	--	--	1 (3.33%)
Failed at NHS and failed at retest	--	1 (3.33%)	12 (40%)	1 (3.33%)
Failed at NHS and did not report the result of the retest	--	--	1 (3.33%)	--
Did not perform or did not know how to report	1 (3.33%)	--	3 (10%)	--
TOTAL	2 (6.66%)	2 (6.66%)	23 (76.66%)	3 (9.99%)

Legend: NHS=Neonatal Hearing Screening

In addition, Table 5 shows that 23 (76.67%) children had sensorineural HL, of which 7 (23.33%) passed the NHS and 12 (40%) failed both in the first test and retest.

Table 6 shows the comparative results between the findings in the NHS test (n=30) and retest (n=17) with the degree of hearing loss in the best ear.

Table 6. Comparative results between the findings in the NHS test and retest with the degree of hearing loss in the best ear.

Result	Degree of hearing loss				
	No HL	Mild	Moderate	Severe	Profound
Approved at NHS	--	1 (3.33%)	2 (6.66%)	4 (13.33%)	2 (6.66%)
Failed at NHS and approved at retest	1 (3.33%)	--	1 (3.33%)	--	--
Failed at NHS and failed at retest	--	3 (9.99%)	4 (13.33%)	3 (9.99%)	4 (13.33%)
Failed at NHS and did not report the result of the retest	--	--	1 (3.33%)	--	--
Did not perform or did not know how to report	1 (3.33%)	--	--	1 (3.33%)	2 (6.66%)
TOTAL	2 (6.66%)	4 (13.33%)	8 (26.66%)	8 (26.66%)	8 (26.66%)

Legend: NHS=Neonatal Hearing Screening

Table 6 also shows a similar distribution between moderate, severe and profound HL, with 8 (26.67%) children in each grade, respectively.

The time between the first consultation and the audiological diagnosis was less than one month for 5 (16.66%) children; between one month and three months for 8 (26.67%) children and over three months for 8 (26.67%) children. It is noteworthy that 9 (30%) children have already been referred to the study center with an audiological diagnosis established by other professionals/institutions.

Regarding the time between the diagnosis and the beginning of the intervention, 2 (6.67%) children started the intervention within one month, 13 (43.33%) between one and three months, and 5 (16.67 %) between four and six months, while 1 (3.33%) took more than 6 months to start the intervention and 9 (30%) were referred to the SASA with a diagnosis of HL.

With respect to the type of hearing aids, 22 (73.33%) used a bilateral PSAP and 4 (13.33%) used a unilateral PSAP. On the other hand, one child had a bilateral cochlear implant (CI) and 3 (10%) did not use any type of device at that time.

As for the difficulties in the access to the SASA, 9 (30%) parents/guardians reported having faced difficulties, 19 (63.34%) did not and 2 (6.66%) did not know how to report. Regarding the waiting time, 4 (13.33%) reported that they waited less than a month for the first consultation, while 14 (46.67%) reported that they waited between one

month and three months, 9 (30%) waited more than 3 months and 3 (10%) did not know how to report.

With respect to the investigation of the global development and hearing of the child by the primary care team, 9 (30%) parents/guardians reported that this follow-up was conducted, 17 (56.67%) reported that it was not conducted and 4 (13.33%) reported that they do not use the public health service or do not use it that much. As for the primary care professionals most reported by parents/guardians as interested in global development and hearing, 4 (13.33%) mentioned the doctor, 2 (6.66%) mentioned the nurse, 5 (16.66%) mentioned the community health agents and 3 (10%) mentioned the speech-language pathologist of the NASF. In addition, 13 (43.34%) parents/guardians reported that no professional showed interest in this subject and 3 (10%) did not know how to report.

Regarding the monitoring of global development, hearing and language in primary care, 5 (16.66%) parents/guardians reported that it was conducted, while 25 (83.34%) reported that there was no follow-up. In addition, 10 (33.33%) parents/guardians reported that the community health agent (CHA) confirmed the screening and vaccination records in the child's health booklet during home visits, while 20 (66.67%) did not.

Discussion

This study found that 86.66% of children underwent NHS, which is similar to studies carried out in Curitiba[15] and Batatais[16], which reported 86% and 90.84%, respectively. On the other hand, a reference center for hearing health in São Paulo[17] found that only 57.8% of children underwent NHS between 2010 and 2015.

Among the children who underwent NHS, it should be noted that the majority (96.15%) underwent the first test within the first month of life, including 69.23% before the hospital discharge, as recommended by the JCIH, the Guidelines for Hearing Health Care at the Health Care Network for People with Disabilities in Santa Catarina and by the MH Guidelines[2,8,14]. These guidelines state that the test must be performed in the first days of life in hospitals and maternity hospitals or, within the first month, if performed in other institutions[2,8,14.]

All parents/guardians responded that they would choose to perform the NHS, even if it was optional, and 76.92% of the subjects recognized the relevance of the NHS for the diagnosis. It can be compared with a study carried out in Nigeria[18], which found that 85.4% of mothers of newborns understand that the NHS is of paramount importance for childcare. This data should be highlighted, since the lack of recognition on the part of families may affect the performance of necessary hearing tests[19]. It should be noted that these responses may have been influenced by the presence of the researchers in the application of the questionnaire in a closed room in the SASA, in which the children are monitored on the use of the PSAP/IC, which may lead the subjects to respond positively to this question.

The presence of RIHL was reported in 63.34% of the children, with NICU stay as the most reported indicator, followed by craniofacial anomalies and family history of deafness. Similar data were found in a study carried out at a hearing health reference center in São Paulo[17], which found RIHL in 65.5% of children attended for audiological diagnosis, with the stay in the NICU for more than five days as the most frequent indicator, followed by family history of HL.

A study carried out in a Family Health Strategy (FHS) in Curitiba[15] through screening and monitoring of newborns, found that only 12% of the cases had a RIHL, but with emphasis on the child's stay in the NICU as the main indicator. A lower occurrence of RIHL (25%) was also found in a study[16] carried out from 2009 to 2015 with neonates who were diagnosed with HL. In this case, the main indicator was the use of ototoxic medication, which differs from this study.

Some indicators reported in the literature, such as those most associated with failure in the NHS, had a low occurrence in this study, such as low Apgar score in the first and fifth minutes, birth weight below 1,500 kg, craniofacial malformations, syndromes associated with HL or suspect of syndromes and use of mechanical ventilation[20,21].

The presence of RIHL is a determining factor for the referral of newborns for auditory monitoring[2]. In addition, the hearing development of children with risk indicators must be monitored in the first year of life by primary care[14].

The use of the correct procedure for the NHS, according to the presence or absence of RIHL, is also worth mentioning, as it was found in the child's medical record or health booklet that 47.06% of children with RIHL were tested only through OAE.

For children with IRDA, the literature recommends performing the test directly with automated Brainstem Auditory Evoked Potential (BAEP-A) or in screening mode at 35 dB NA, due to the higher prevalence of retrocochlear hearing losses in this population, which is not identified with OAE[2,8]. This inadequacy of the procedure performed may be related to the lack of equipment of the institutions that carry out the screening, as well as to the non-mandatory character of the BAEP.

It is recommended that the audiological diagnosis should be performed until the age of three months and the intervention with the use of sound amplification until the age of six months[2,8], However, most children in this study did not have a diagnosis (86.67%) nor initiated the intervention (83.34%) in the recommended times. As for the diagnosis, data similar to this study were found in a study conducted in São Paulo[17], in which 81.60% of the children were diagnosed at an older age than recommended. In contrast, 55% of participants in a study in North Carolina (USA)[22] completed the diagnostic process within the time recommended by the JCIH.

Therefore, it can be said that the compliance with the goals established by the MH Guidelines is below expectations concerning the continuity of the process after the first NHS test.

The results of this study show that there is a significant difference ($p<0.005$) regarding the early diagnosis of HL for the group of children with HL who did not perform the NHS when compared to the group that performed it, which suggests that performing the NHS was a decisive factor for the early diagnosis of HL. Although a study conducted in São Paulo[12] showed similar results, the authors of this study observed that the average age at diagnosis and intervention was approximately 45.4 months and 57.6 months before the implementation of the NHS in care, and 41.2 months and 45.5 months after its implementation. These values are higher than that found in this study, in which the average age at diagnosis and intervention was 31 and 34 months, respectively, among all children surveyed.

Although statistically the children who underwent NHS completed the diagnosis of HL earlier than those who did not, it can be noticed that 38.46% completed the diagnosis after 24 months of age, with only one having a postnatal RIHL (meningitis). This late diagnosis can be explained by several factors, including difficulties in completing the diagnosis, lack of qualified professionals in the area to perform the exams[22], the family's acceptance/denial process for regarding the potential HL; abandonment of treatment; number of absences due to work and problems with displacement or health of the child or family.

This study identified a difference between the final diagnosis and the result of approved-failure in the NHS, with 34.61% of those who passed the first test and 11.76% of those who passed the retest having HL. Some factors that affect the performance of the test/retest may explain the difference in the result in the NHS compared to the audiological diagnosis, such as: the professional's ability to conduct the test, calibration of the screening equipment, background noise where the tests are carried out, algorithms used as a reference for failure and approval in the equipment and number of attempts to perform the test[22]. The presence of late[22] HL or HL due to postnatal infections such as meningitis, as in two children in this study, may also have influenced the results. Furthermore, it is noteworthy that this study did not identify retrocochlear changes, so that no false negative results would be explained by the failure to perform the procedures recommended by the MH Guidelines[2] for children with RIHL.

Regarding the diagnosis, there was a prevalence of bilateral HL in the study participants, with only 6.67% having unilateral HL. The low occurrence of unilateral HL was also observed in a study conducted in the USA[23], with 8.9% of unilateral HL in children who failed the first NHS test.

There was a prevalence regarding the type of HL considering the best ear, with 76.66% of sensorineural HL; while other studies recorded a prevalence of 86.56%, 50% and 59.10% of sensorineural HL[24,16,21].

This study found a similar distribution between the moderate, severe and profound HL in the children studied. In contrast, other studies show a predominance of moderate (30.72%)[25] or severe-to-profound HL (68%)[25].

As for the reaction of families who understand that NHS is important for the diagnosis, it should be noted that 75% of them reported that, although sad, they understood and were concerned with what was best for their child. A study[26] carried out regarding the parents' attitude towards the early detection and intervention of HL and NHS, also found the prevalence of a positive attitude. It is

noteworthy that the positive reaction reported by the families to the diagnosis in this study may have been influenced by the present, since the families have already gone through the initial mourning period of the diagnosis and are being assisted by the study center.

It should also be noted that after diagnosis, it is important for the family to accept the intervention and provide hearing aids to the child, if necessary. It was possible to find that 90% of the children at the time of the study used some type of adaptation, be it a Personal Sound Amplification Product (PSAIP) or a cochlear implant (CI). This number is higher than the acceptance rate found in another study[26], which reported an acceptance rate of 80% of families regarding the use of hearing aids, allowing children to use it, if necessary. The high adherence found in this study may be overestimated due to the study being carried out in a highly complex care service, which provides devices and constant monitoring of the children attended.

According to the parents/guardians of the children who failed the first test and retest, only 57.14% were referred for a diagnosis through NHS, which is higher than the rate found in a study conducted in São Paulo[17] that identified that only 29.6% of referrals for diagnosis of HL in a specialized center came from NHS.

The fact that only half (57.14%) of the children are referred for audiological diagnosis from NHS, shows a large gap in referrals. According to the recommendations, if the retest fails, the newborn should be immediately referred to otorhinolaryngological and audiological diagnostic assessment in Specialized Rehabilitation Centers (CER) and in the High Complexity Hearing Health Outpatient Clinic licensed by the Ministry of Health[2]. This gap is also reported in the literature. A survey conducted in a hospital in São Paulo found that 28% of neonates referred for intervention stage did not attend the designated service[27].

Although access to health professionals is a relevant factor for families to adhere to diagnostic and audiological intervention procedures after failure in NHS, 30% of parents/guardians reported difficulties in entering specialized care: the greater the access, the greater the adherence[28].

According to the report of the parents/guardians, the wait for specialized care was longer than three months for 30% of respondents, which is not in line with the immediate care in cases of failure in NHS so that the times recommended in the literature[2,8,14] are duly complied: if the diagnosis must be made within the first three months of life, how can a child wait this same time in line for care?

In addition to the difficulty reported concerning the waiting time for care, other factors reported in the literature as barriers to adherence were mobility difficulties, such as the distance from home to the service; the need to use more than one public transport mode; financial difficulties; lack of social support; forgetting the scheduled date; restriction of service hours; difficulties to leave work and the need to take care of other children[25,29].

The Guidelines for Hearing Health define that the Municipal Health Department is responsible for ensuring compliance, encouraging the performance of the NHS and ensuring that children can have access to care, whether in public institutions or in partnerships with the private sector or with other municipalities[14].

The guidelines also highlight the role of primary care as responsible for the care of children's hearing health, which includes from the verification of the performance of NHS in the child's health booklet to the monitoring of development. Thus, the low follow-up (16.66%), reported by parents/guardians concerning the global development, hearing and language of children by primary care is worrying. Even with satisfactory results in the NHS, the Municipal Health Department is responsible through primary care for monitoring the hearing development of children with or without RIHL in the first year of life and, if HL is suspected, these cases must be referred for audiological evaluation and have access to diagnosis in specialized centers [14].

One of the possibilities reported in the literature for the low monitoring of children in primary care is the divergence in the conduct to be adopted by the Family Health Strategy (FHS) in the face of a HL, which can cause delays in referrals. Among the divergent conducts reported by the author, there is a question about which professional to refer to, whether to request exams or guide family members. In addition, the deficit of counter-referral of cases raised as suspected HL makes it difficult for the team to monitor and provide comprehensive care for the child.[6].

It is understood that there is fragility in the development of health care networks with regard to people with disabilities, in which medium and high

complexity services should train the primary care to serve these children. Although the Guidelines for Hearing Health Care at the Health Care Network for People with Disabilities in Santa Catarina[14] and the Guidelines for Attention for Neonatal Hearing Screening of the Ministry of Health[2] are available on the Internet, these professionals are not always able to search for or consult such documents in daily practice. Thus, a better structuring of the care network for people with disabilities could positively change this scenario.

Although the results of diagnosis and intervention are better than a previous study[12], they are still below expectations[2,8]. It is believed that it is necessary to improve the articulation between the different levels of care (primary care, medium and high complexity) on the operation of the NHS and its stages, since it is not limited to the ear test. It is essential that the families of children at risk for, or with confirmed HL, have the necessary guidance and support, in order to access the specialized services at the recommended times, aiming at a better auditory and language development of these children. To this end, teams at all levels of care need to know the referral flows, the existing laws and recommendations, the RIHL, as well as the characteristics of the typical language and hearing development in children.

One of the possibilities suggested in the literature to better equip the other points of the network, especially the community health agents, who are the main link between primary care and families, is the organization of face-to-face training with expository classes and workshops or via videoconference[30].

Final Considerations

At the end of this study, it was possible to verify that the first stage of the NHS (test) was carried out within the period recommended by the literature for most children. However, the other procedures (retest, diagnosis and start of intervention) are not in line with the recommended periods.

The findings indicate that the performance of the NHS has an important role for the early diagnosis of HL. Although these are still below the recommended, the children who underwent NHS were referred in a significantly shorter time than those who did not.

The study also identified low follow-up and investigation by primary care for children with, or at risk for HL. This indicates the importance of carrying out the NHS as a way of screening and identifying these subjects, since the network shows weaknesses to provide adequate support to the families of children at risk or confirmed HL.

It should be noted that this study will continue with the aim of increasing the sample and providing data that can serve as a basis for the implementation of programs and projects for the improvement and effectiveness of actions and flows of the NHS, thus enabling an early diagnosis and intervention that are relevant for the development of children with HL, so that they can be as close as possible to their hearing peers.

Further studies are also suggested in order to identify the journey and the difficulties faced by families in the process of accessing the diagnosis and intervention of a HL, with a special focus on the role of primary care and public policies.

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