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# Quality indicators of hearing screening and evaluation of neonatal lingual frenulum

## *Indicadores de qualidade de triagem auditiva e de avaliação do frênulo lingual neonatal*

### Keywords

Newborn Screening  
Lingual Frenulum  
Ankyloglossia  
Quality Indicators  
Health Care

### Descritores

Triagem Neonatal  
Freio Lingual  
Anquiloglossia  
Indicadores de Qualidade  
Assistência à Saúde

### ABSTRACT

**Purpose:** To analyze the quality indicators of the hearing screening program and to evaluate lingual frenulum in newborns, in terms of coverage rates, duration of hearing screening, referral indexes and prevalence of ankyloglossia in a university hospital specializing in mother and child care. **Method:** This is a cross-sectional study, in which we analyzed the database of the newborn hearing screening and lingual frenulum assessment program for the period between September 2015 and August 2016. **Results:** During the study period, 2,345 babies were born at the institution, 1,380 (58.8%) underwent newborn hearing screening and 1,350 (57.6%) were diagnosed with a lingual frenulum. Mean gestational age was 39 weeks ( $\pm 1.6$ ), birth weight 3478g ( $\pm 469.2$ ) and 69% were boys. In newborn hearing screening, 95.7% of the 1,380 babies screened were discharged with guidance, 2.4% were referred for auditory monitoring and 1.9% of babies were referred for auditory diagnosis. In lingual frenulum assessment, 123 (9.1%) of the 1,350 evaluated, had ankyloglossia, 85 were boys and 47 girls. **Conclusion:** The indicators of the time of life in which the hearing screening is performed, the referral indices and ankyloglossia prevalence are in line with those reported in the literature; however, coverage rates were lower than recommended and do not comply with Brazilian law.

### RESUMO

**Objetivo:** analisar os indicadores de qualidade do programa de triagem auditiva e avaliação do frênulo lingual em neonatos, quanto às taxas de cobertura, tempo de vida em que são realizadas as triagens auditivas, índices de encaminhamentos e prevalência da anquiloglossia, em um hospital universitário especializado em assistência materno-infantil. **Método:** trata-se de um estudo transversal, no qual foi analisado o banco de dados do programa de triagem auditiva e da avaliação do frênulo lingual em neonatos, referente aos atendimentos realizados no período de setembro de 2015 a agosto de 2016. **Resultados:** no período analisado, nasceram 2.345 neonatos na instituição, 1.380 (58,8%) realizaram a triagem auditiva neonatal e 1.350 (57,6%), a avaliação do frênulo lingual. A média de idade gestacional foi de 39 semanas ( $\pm 1,6$ ), peso de nascimento igual a 3.478 g ( $\pm 469,2$ ) e 69% eram do sexo masculino. Na triagem auditiva neonatal, dos 1.380 neonatos, 95,7% receberam alta com orientação, 2,4% foram encaminhados para monitoramento auditivo e 1,9% foram encaminhados para diagnóstico auditivo. Na avaliação do frênulo lingual, dos 1.350 avaliados, 123 (9,1%) apresentaram anquiloglossia, 85 eram do sexo masculino e 47, feminino. **Conclusão:** os indicadores de tempo de vida em que são realizadas as triagens auditivas, índices de encaminhamentos e prevalência da anquiloglossia estão em consonância com a literatura, porém as taxas de cobertura estão inferiores ao recomendado e não seguem a legislação brasileira.

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## INTRODUCTION

Newborn screening is a preventive measure that can identify certain pathologies in asymptomatic individuals by applying tests during the first 28 days of life. The primary objective of screening is to refer newborns who most likely exhibit the disease investigated for more elaborate diagnostic procedures, in order to provide early treatment, thereby decreasing or eliminating associated sequelae<sup>(1,2)</sup>.

Newborn hearing screening (NHS), also known as early hearing detection, and lingual frenulum assessment, or the tongue-tie test, are part of neonatal screening procedures. NHS is considered the most effective and recommended way for early hearing loss detection and intervention<sup>(3)</sup>. Lingual frenulum assessment makes it possible to identify ankyloglossia, which restricts tongue movements and may interfere in breastfeeding<sup>4</sup>.

This early diagnosis has a significant effect on newborn growth and development<sup>(2)</sup>. Early interruption of breastfeeding, common in ankyloglossia<sup>(4)</sup>, can compromise the immune function of newborns and negatively affect nutritional gain<sup>(5)</sup>. Hearing disorders may delay language development and cause cognitive, emotional and social disturbances in the affected individuals as well as their families and communities<sup>(3)</sup>.

For this reason, these screenings are mandatory in all hospitals in Brazil, as stipulated in Federal laws 12.303/2010<sup>(6)</sup> and 13.002/2014<sup>(7)</sup>, governing NHS and lingual frenulum assessment, respectively.

Quality indicators stipulate that all newborns be assessed in the first months of life<sup>(7)</sup>, recommending at least 95% NHS coverage<sup>(8)</sup> and universal lingual frenulum assessment. In the national and international literature, there is a lack of studies on the concomitant application of both screenings.

In this respect, the present study aims to analyze the indicators of NHS quality and lingual frenulum assessment in terms of coverage rates, time of life that hearing screenings are carried out, referral indices and prevalence of ankyloglossia in a university hospital specialized in mother and child care.

## METHOD

This is a cross-sectional study in which we analyzed the database of 2,730 newborns assessed by the NHS program and lingual frenulum evaluated by the Ana Bezerra University Hospital (HUAB), in the city of Santa Cruz, Brazil. Around 3000 births per year are recorded at the hospital, which serves a population of a little over 200,000 inhabitants.

Healthy, full-term newborns, adequate for gestational age (GA), born at HUAB, were included in the study. Those from other institutions, with syndromes or congenital malformations, were excluded. In line with Resolution no. 466/2012, chapter IV, this research dispensed with written informed consent, without compromising the subsequent clarification process, given that it used secondary data from the database of the project, which was approved by the Research Ethics Committee, under protocol number 2.413.853, and ensured the privacy of the medical records analyzed.

NHS and lingual frenulum assessment were conducted during the mother and newborn's hospitalization or at the neonatal follow-up outpatient facility by a speech therapist and three dentists, the latter only to evaluate lingual frenulum. In order to perform hearing screening, the transient evoked otoacoustic emissions (EOAT) protocol of the Accuscreen Madsen<sup>®</sup> screener, duly calibrated for each assessment, was applied only by the speech therapist, a specialist in clinical and educational audiology.

The Martinelli et al.<sup>(9)</sup> protocol, applied in only one stage (during hospitalization) was used to assess lingual frenulum. The protocol was applied by a speech therapist or dentist, and in the event of uncertainty, the case was discussed with two professionals specialized in orofacial motricity, always in the presence of the speech therapist. The speech therapist and dentist trained the author of the protocol and the rest of the team.

NHS presents two results: "pass" or "fail". According to the equipment's manual, the following parameters must be reached to achieve a "pass" result: eight response frequency peaks, artefact rate below 20% and probe stability above 80%. If one of these parameters is not obtained, a "fail" result is recorded in screening. A "pass" result means that the newborn exhibits a hearing threshold less than or equal to 30 dB.

NHS was conducted in two stages. At the first testing, carried out at hospital discharge or the outpatient facility, a "pass" was considered when the newborn showed the presence of otoacoustic emissions in both ears and "fail" when this did not occur in at least one of the ears. When "fail" occurred, a retest was scheduled in the outpatient facility of the maternity hospital, approximately 15 days later. If the retest also failed, the baby was referred for audiological diagnosis at the Auditory Health Reference Service, for hearing assessment. If the infant displayed a risk indicator for hearing loss (RIHL), it was referred to the reference service for auditory monitoring, consisting of audiological assessment, including brainstem auditory evoked potential (BEAP) and follow-up of hearing and language development. Finally, when the newborn had no risk indicator and the result was "pass", it was discharged with guidance, and the parent or guardian received verbal orientation regarding the auditory and linguistic development of the child.

Lingual frenulum assessment was performed in only one stage (during hospitalization). Newborn screening of the lingual frenulum assessment protocol in babies consists of four items: oral rest posture, with lips closed, slightly parted or open; tongue position while crying, on the midline, raised, on the midline with the sides raised or with the tip of the tongue lowered and the sides raised; the shape of the tip of the tongue was also assessed while the newborn was crying or raising its tongue, the shapes being round, with a slight groove or heart-shaped; finally, the frenulum itself, where thickness (thin or thick), fixation site on the tongue (in the middle third, between the middle third and the apex or at the apex), and fixation site on the floor of the mouth, on the sublingual caruncles or inferior alveolar crest, were evaluated. The protocol contains a progressive scale, where a score of zero means normality, while one to three, indicates altered characteristics<sup>(10)</sup>.

The diagnosis of altered frenulum was based on the anatomofunctional assessment score on the Lingual Frenulum Assessment Protocol in Babies. When the total anatomofunctional assessment score was greater than or equal to 7, the frenulum was deemed to have interfered with tongue movements<sup>(10)</sup>.

Surgery to release the lingual frenulum was performed by two dental surgeons. The technique consists of the following phases: drying and anesthetizing the frenulum membrane with topical anesthetic (ethyl aminobenzoate); puncturing the lingual frenulum with surgical scissors; and compressing the puncture site for hemostasis diffusion of membrane tissue, until reaching the adequate point to release the tongue<sup>(11)</sup>. Sutures were not needed and after the procedure, the baby was allowed to breastfeed, since mother's milk accelerates healing<sup>(12,13)</sup>. A return visit was scheduled for 30 days post-surgery for reassessment and orientation from one of the professionals.

The data obtained were tabulated and submitted to statistical analysis, using SPSS software, version 17.0. The chi-squared test was applied and a 5% significance level was adopted ( $p < 0.05$ ).

## RESULTS

Between September 2015 and August 2016, 2,345 babies were born, 179 at the institution under study. Of these, 1,380 (58.8%) were submitted to NHS, 1,350 (57.6%) of whom underwent lingual frenulum assessment. All the newborns evaluated (69% boys and 31% girls) were hospitalized in the rooming-in unit, with average gestational age of 39 weeks ( $\pm 1.6$ ) and birth weight of 3.478 g ( $\pm 469.2$ ).

The tests were applied at an average age of 12.6 days ( $\pm 19$  days). Most of the babies (96%) underwent NHS in the first 30 days of life. Only 50 infants were tested between 31 and 107 days of life and were excluded.

The referral index for the NHS retest was 7.6%, 40% of whom did not attend. Of the 1,380 babies screened, 1,320 (95.7%) were discharged with guidance, 34 (2.4%) were referred for auditory monitoring and 26 (1.9%) for auditory diagnosis (Figure 1).

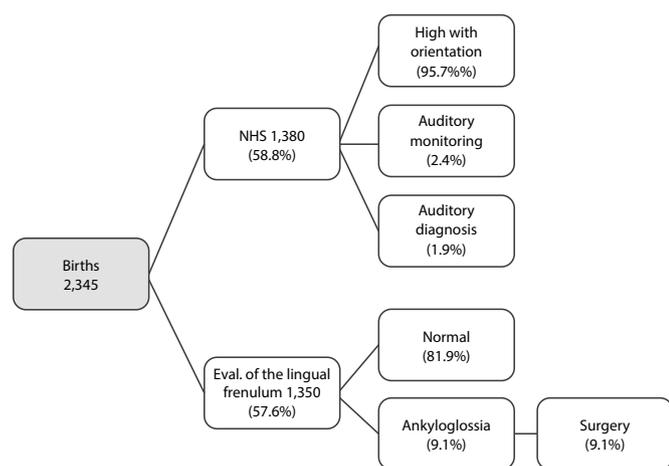


Figure 1. Flowchart of hearing screening and lingual frenulum assessment

With respect to lingual frenulum assessment, of the 1,350 evaluated, 123 exhibited alterations, which generated an ankyloglossia prevalence of 9.1% (Figure 1). Of these, 85 (69%) were boys and 47 (31%) girls. All were referred for surgery, but only 85 (69%) attended. The chi-squared test ( $\chi^2$ ) was used to compare proportions, revealing no sex prevalence in the results ( $p = 0.07$ ).

## DISCUSSION

Our results demonstrate that although the indicators regarding the time of life that auditory screenings take place and the referral index for auditory diagnosis are in line with literature recommendations and Brazilian laws, the coverage data are below recommended levels<sup>(8,13)</sup>. Lingual frenulum assessment coverage was also far from universal and the prevalence of ankyloglossia corroborates literature findings<sup>(14-16)</sup>.

The average NHS coverage rate in Brazil is 37.2%, varying according to the regions and programs<sup>(13)</sup>. In the present study, despite exhibiting an NHS coverage rate above the national average (58.8%), the result was far from the value recommended in the literature, which is more than 95%<sup>6</sup>. In relation to lingual frenulum assessment coverage, no studies describing this indicator were found.

The first neonatal hearing screening programs in Brazil occurred in the 1990s. Later, municipal laws were enacted, making the examination mandatory in some municipalities, culminating in Federal Law no. 12.303/10<sup>(6)</sup>, which mandated it throughout the country. In regard to mandatory lingual frenulum assessment, several municipal and state laws were approved in 2012, concluding with Federal Law no. 13.002/14<sup>(7)</sup>.

Corroborating Ministry of Health guidelines<sup>(8)</sup> but diverging from the results reported by Januário et al.<sup>(17)</sup>, in the present study, 96% of the newborns were assessed by 30 days of life. This indicator is directly related to early diagnosis and treatment of hearing loss<sup>(17)</sup>. One hypothesis to explain the high coverage rate in our results was to recommend assessment in the first days of life, while the newborns are still hospitalized.

A study conducted at a Brazilian facility confirmed that NHS provided early diagnosis and intervention in children with hearing loss and that factors such as families not attending outpatient consultations and diagnostic peculiarities hindered early diagnosis in most of the children<sup>(17)</sup>.

Another noteworthy finding is the auditory diagnosis referral index. Although we studied a medium-complexity maternity hospital in a rural area of Northeastern Brazil, the index corroborates the value reported by the Ministry of Health<sup>(8)</sup> and those obtained in larger programs, such as in the Southeastern region<sup>(18,19)</sup>.

A study performed in the municipal maternity hospitals of Rio de Janeiro found a coverage of 40.5% and only 1.7% failed, requiring referral for auditory diagnosis<sup>(18)</sup>. In another program, also in the Southeastern region, the coverage and referral rates were 90.9% and 0.5%, respectively<sup>(19)</sup>.

An integrative review conducted in 2014 found that most newborn hearing screenings occur in public maternity hospitals. Fewer than half of these facilities achieve 95% coverage and the referral index for diagnosis was below 4%<sup>(20)</sup>.

No studies reporting lingual frenulum assessment coverage in Brazil were found, likely because the examination was only recently introduced in the country. The few studies are limited to clinical investigations<sup>(21,22)</sup>. In the present study, we found lingual frenulum assessment coverage of around 60%, which may increase by training more professionals and optimizing patient flow.

The prevalence of ankyloglossia was 9.1%, in line with the data of other studies, which exhibited a prevalence between 0.8 and 16%<sup>(4,23,24)</sup>. In these, the authors used the same assessment protocol reported here and deemed it reliable for diagnosing lingual frenulum changes<sup>(24)</sup>.

An interesting observation was our finding no association between sex and lingual frenulum changes, given that 85 (69%) of the newborns identified with ankyloglossia were boys. This corroborates the findings of Ferrés-Amat et al.<sup>(25)</sup>, who assessed 171 infants diagnosed with ankyloglossia, 60 of whom (35.1%) were girls, and 111 (64.9%) boys, showing no statistically significant difference between sexes. One reason would be its hereditary nature, which is independent of sex<sup>(26)</sup>. Indeed, the correlation between sex and ankyloglossia remains poorly explained in the literature. In some studies, the data demonstrate that boys seem to be more affected than girls<sup>(27)</sup>, while others report a similar<sup>(28)</sup> or inverse proportion<sup>(29)</sup>.

Although we surmised that access to the hospital is easier for families living in rural areas of the state because of their proximity to the health units, compared to those living in large urban centers, outpatient treatment adherence in the present study was low. In the case of hearing screening, 42 newborns did not undergo the retest.

This failure to submit to retesting may be due to the lack of knowledge regarding diagnostic possibilities, disinterest of the family, or difficulty in scheduling an appointment because of family obligations<sup>(30)</sup>. In light of this, our team recorded the screening results and retest appointments on the vaccination card. In addition, telephone contacts were made in an attempt at rescheduling appointments, including the newborn hearing screening retest, frenotomy or post-surgery return.

A limitation of this study was not submitting newborns with a risk indicator for hearing loss (RIHL) to BEAP. The low number of speech therapists precluded applying the test during hospitalization. As such, in order to guarantee the coverage recommended by the Ministry of Health, the infants with RIHL were referred for monitoring at the Audiological Reference Service. NHS is only the start of the Auditory Health Care Program for newborns. Integrating the auditory health care network among the different complexity levels is certainly a denominator of comprehensive newborn care<sup>(19)</sup>.

Newborn screening is an important health promotion and disease prevention tool, guiding early diagnosis and treatment. Although our study demonstrates that indicators of the time of life that auditory screenings take place and the referral index for auditory diagnosis are in line with those recommended by the literature and Brazilian law, the coverage data are below the stipulated levels. These indicators make it possible to analyze the care provided and improve the program.

However, a further limitation of the present study was not analyzing the relationship between sex and the changes found

in NHS. Thus, new studies should be conducted to identify the prevalence of hearing loss in newborns and establish relationships between ankyloglossia and breastfeeding in order to justify and strengthen newborn screening.

## CONCLUSION

The newborn hearing screening and lingual frenulum assessment program at the university hospital analyzed exhibited indicators of the time of life screenings are carried out, referral indexes and ankyloglossia prevalence that corroborate the literature, but the coverage rates of these screenings are lower than recommended levels and do not comply with Brazilian law. We underscore the importance of newborn screening to provide early diagnosis and treatment of hearing loss and ankyloglossia.

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### Authors contributions

*MRPD and SAP were responsible for the idealization and work design; AGFA was responsible for statistical analysis; CCSX and NSOH wrote the initial version of the article; SAP, JCSL, MRPD and AGFA reviewed and contributed to the writing of the final version of the article. MRPD, CCSX, JCSL and NSOH participated in the data collection. All authors read and approved the final version of the manuscript.*