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Original Research Article

A two-step aABR regime in neonatal hearing screening: an efficient alternative for remote and poor geographic areas

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ABSTRACT

Background: Neonatal hearing screening is a standard of care for the neonatal population. Nevertheless, lost to follow-up (LTFU) cases remain an issue for many new and mature programs. This study aims to evaluate the impact of hearing screening using an aABR algorithm in a neonatal population, with a particular focus on its effect on LTFU rates.
Methods: This retrospective study analyzed 2,072 infants selected from a total of 2,181 neonates born in the districts of Pogradec and Kukës, Albania, during 2018–2019. Two different newborn hearing screening algorithms were applied: a two-stage aABR protocol in Kukës and a three-stage TEOAE–TEOAE–aABR protocol in Pogradec. The "fail" rates and the proportion of infants LTFU were calculated and compared between the two screening approaches.
Results: LTFU rates were highest in Pogradec, where a traditional screening algorithm was used, increasing from 1.4% at the first test to 16.9% at the second. In Kukes, the corresponding rates were 0.7% and 15.5%. Fail rates ranged from 22.9% in Pogradec to 6.8% in Kukes. At both sites, fewer neonates failed the second screening than the first.
Conclusions: The algorithm based on two aABR stages appeared to produce a lower number of subjects who failed the first screening test and also a reduced number of LTFP cases. The 'fail' rates remained high in both sites, although lower in Kukes. Targeted strategies to reduce LTFU should be implemented to enhance the overall sensitivity and effectiveness of the hearing screening program.

Keywords: aABR, 'fail' rates, Lost to follow up, Neonatal hearing screening, Screening algorithms, TEOAE

INTRODUCTION

The protocol for hearing screening in "well babies" (WB) and "at-risk" newborns is both essential and complex. The choice between the two most commonly used screening methods otoacoustic emissions (OAE) and ## (aABR) as well as the number of screening steps or the combination of these technologies, depends on the specific objectives of each program and the inherent characteristics of the tests.[1-3] Although widely adopted in newborn hearing screening programs, there is still debate regarding the sensitivity and specificity of these tests (which vary according to the selected method), program costs and the percentage of cases lost to follow-up (LTFU). For newborns admitted to neonatal intensive care units (NICUs), aABR-based protocols are among the most widely used, as this method can detect auditory neuropathy spectrum disorder (ANSD), which occurs in approximately 1 in 1,000 NICU patients.[4,5] This is also the standard protocol applied in Albania for NICU populations. For well babies, OAE is typically used as the initial screening test before hospital discharge (the first screening stage). It is often followed by either a repeat OAE or an aABR test, either within the same step or after several weeks. In Albania, the approved protocol for well babies consists of two TEOAE tests, followed by an aABR in cases of persistent failure. Beyond the distinction between WB and NICU populations, both OAE and aABR have specific advantages and limitations.[2,6,7]

The EUSCREEN project is a European research initiative evaluating the cost-effectiveness of hearing and vision screening. It collected data from multiple European countries and also included contributions from outside the EU. Albania participated in this project by implementing a universal newborn hearing screening (UNHS) program across three provinces: Tirana, Kukës and Pogradec. These regions were chosen for several reasons.[8,9] Kukës and Pogradec are remote, mountainous regions, with Kukës being one of the poorest areas in Albania. Both provinces have a similar annual number of births, but neither maternity hospital has a fully equipped NICU. Instead, they operate smaller units that monitor newborns with temporary health concerns, while severe cases are referred to the capital for specialized treatment.

Analysis of the EUSCREEN data 8 revealed that the rate of LTFU was very high, despite different strategies to raise awareness about the importance of early detection of hearing loss. A possible explanation is that families were required to travel multiple times to maternity hospitals for second or third screening steps, which demanded time, financial resources and leave from work.

The aim of the present study is therefore to assess whether simplifying the screening protocol could reduce LTFU rates and thereby improve the sensitivity of the screening program. For this reason, data from the two Tirana maternity hospitals, where standard protocols were used, are not included in this analysis. By contrast, the protocols in Pogradec and Kukës although implemented in regions with similar geographic and demographic conditions differ in both their algorithms and their screening approaches.

METHODS

This retrospective study examined the babies born at the maternity hospitals of Kukës and Pogradec over a 24-month period, from January 2018 to December 2020. Kukës and Pogradec are two districts located in the northern and eastern regions of Albania. Both have a similar population size and annual number of deliveries. The population in these districts is predominantly rural, with many villages situated in remote geographic areas, which can make repeated access to hospital services challenging when required.

A total of 1,255 subjects from Kukës and 926 subjects from Pogradec were included in the study. The database containing information on live births and subsequent screenings underwent quality control procedures to identify irregularities, including inconsistencies in date of birth, gestational age and other key variables. Only records with complete and reliable data were retained for further analysis. Consequently, for this investigation, 1,242 screened subjects from the Kukës cohort and 830 from the Pogradec cohort were included. These cases represented datasets in which all required variables were comprehensively included and documented. For all newborns enrolled and followed throughout all stages of the screening program parents have given written consent for testing.

In Pogradec, the screening protocol consisted of two TEOAE tests performed two weeks apart. If a newborn failed both TEOAE tests, an aABR test was administered two weeks later in the maternity hospital. Infants who failed the aABR were referred for audiological assessment at the University Hospital Center in Tirana (UHCT). In Kukës, the screening algorithm consisted of two aABR tests, also performed two weeks apart. Newborns who failed both tests were referred for audiological assessment at UHCT.

Both screening protocols took into consideration the adjustment of gestational age when scheduling the second or third test. Upon discharge, if a baby failed the first test, parents were provided with an appointment for the second or third test and were sometimes reminded by telephone. Both maternity hospitals had very simple NICU wards, where premature infants or babies with various minor neonatal issues were usually kept. However, neonates with serious health conditions are generally transferred to one of the two specialized NICUs in Tirana.

All subject data were entered into a database, with each infant assigned a unique code to ensure anonymity and prevent any possibility of linking the code to a specific child. The implementation of newborn hearing screening (NHS) in these maternity hospitals was not classified as a clinical trial by the national ethics committee, nor was it considered a novel procedure. Therefore, formal ethics approval was not required for its implementation. However, the study received approval from the University of Medicine in Tirana and the Ministry of Health and Social Protection. Informed consent was obtained from parents prior to screening their infants.

RESULTS

In the Pogradec area, 830 out of 926 live births underwent the first-step screening, representing a coverage rate of 89.6 %. In the Kukës area, the first screening test was performed in 1,242 out of 1,255 live births, achieving a coverage rate of 98.9%. The differences include newborns who were referred directly to Tirana due to medical complications or because parental consent was not provided or subjects whose data were recorded not correctly on the database. This indicates that, despite logistical challenges, both regions achieved high overall coverage. Figures 1 and 2 present the results during the first stage of screening for both sites in percentages and absolute numbers.

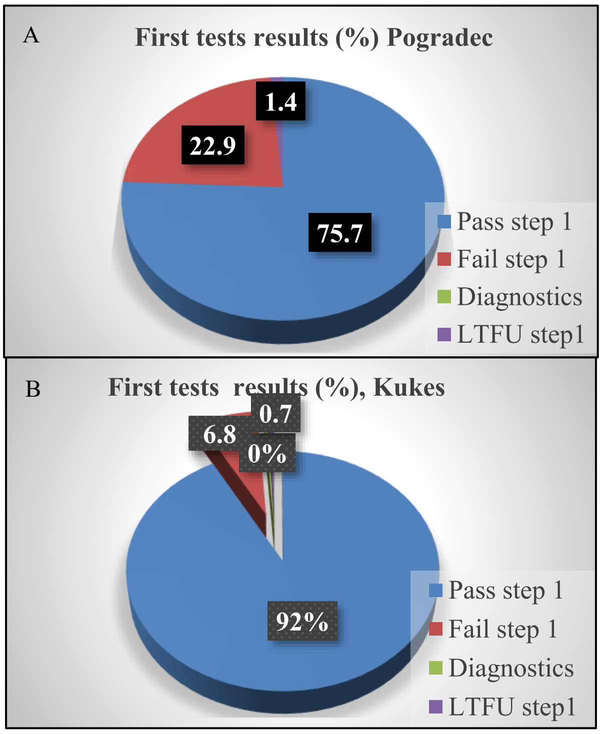

Figure 1 (A, B): Results of the first tests on both sites in percentage.

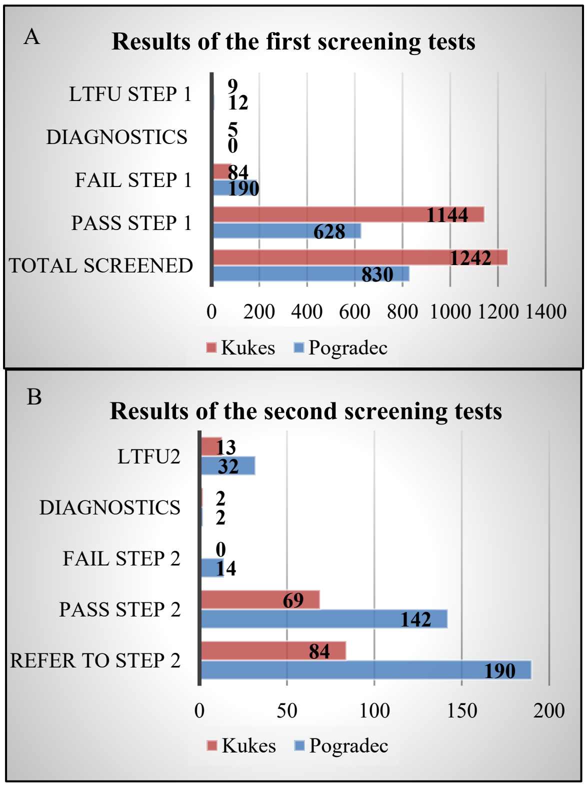

Figure 2 (A, B): Results of the first and second tests on both sites in absolute numbers.

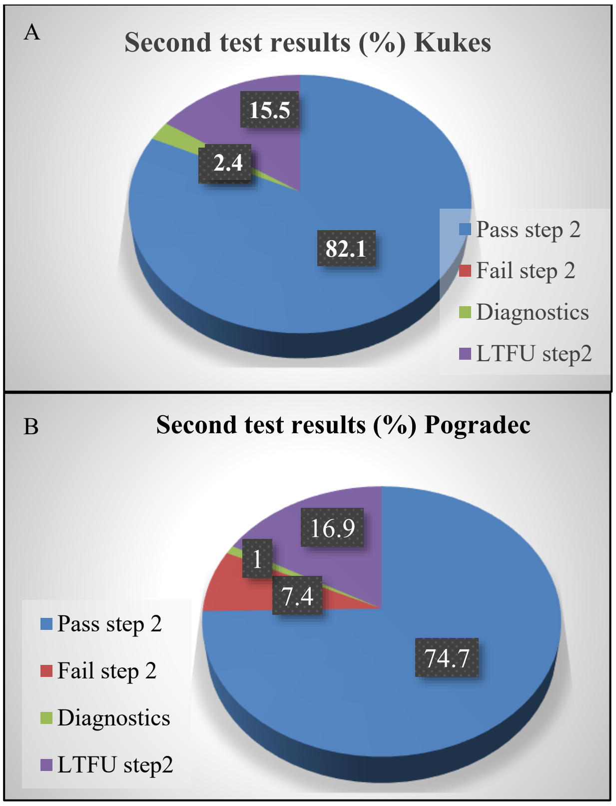

Figure 3 (A, B): Results of the second tests on both sites in percentage.

During the first stage of screening, 92.1% of subjects in Kukes and 75.7% of newborns in Pogradec passed the screening test and were therefore released from the programme. The ‘pass’ percentage during the second testing was 82.1 % in Kukes and 74.7% in Pogradec. 14 out of 190 subjects (7.4%) in Pogradec failed to pass the second screening test. 2 subjects from each site were directly sent from diagnostic evaluation in Tirana University Hospital Center.

The screening algorithm ended at the second stage in Kukës, whereas in Pogradec the algorithm included one additional test. In the latter site, 14 subjects were scheduled for further testing: 1 failed the test, 9 were sent directly for diagnostic evaluation and 4 (28.6%) did not show up and were therefore categorized as LTFU. It is also obvious that the ‘fail’ percentage from stage to stage remains high in the Pogradec area (from 22.9 % in the first testing to 7.4% in the second and 7.1 % in the third stage) while in the Kukes area only the first testing 6.8% of subjects failed the test and 0 subjects failed the second test).

The main finding concerns the cases lost from one test to the next. This category again differs between the two sites, varying from test to test. In Pogradec, the percentages of LTFU were 1.4% in the first test, 16.9% in the second and 28.6% in the third values that are notably higher than the corresponding rates in Kukës, which were 0.7% and 15.5% in the respective stages.

DISCUSSION

While discussing a hearing screening program, the sensitivity and specificity of both test types and of the program as a whole require careful consideration. The sensitivity of a screening test is difficult to be established correctly, but it is generally accepted that aABR has slightly higher sensitivity than OAE (approximately 98% versus 95%), although it remains a more expensive test.[10,11] aABR also requires more time to be performed than OAE and is influenced by the electromagnetic environment. aABR is also believed to have better specificity than OAE, both tests being highly influenced by the testing time after the baby being born.[12]

For these reasons, many programs especially those targeting well-babies are primarily OAE-based. In contrast, aABR is universally accepted as the preferred test in NICU settings because it can identify auditory neuropathy, a condition in which OAE responses remain present despite underlying neural dysfunction.[4,5] Different countries adopt varying newborn hearing screening program algorithms depending on budget, staff training and other logistical considerations.[13]

In the data, the 'fail' rate of all stages of hearing screening was lower in Kukës than in Pogradec, varying from 22.9% to 7.4% in the first and second stage, respectively in Pogradec and 6.8% to 0% in Kukes for the same stages. Although the numbers are small, the pattern is clear. This finding is consistent with the meta-analysis by Manz et al, which reported that a two-stage screening using aABR–aABR had a lower referral (fail) rate (1.3%) compared to TEOAE–TEOAE (2.7%).[1] The high failing rate is also connected with the experience of the staff. In a fresh program, it requires some time for the midwives to become competent with testing and then the failure rate begins to decrease.

Apart from the 'fail rate', LTFU remains a major challenge even in well-established programs, regardless of the algorithm used.[14,15] In the data, LTFU percentages were particularly high in Pogradec, where a more traditional algorithm was applied. It rose from 1.4% in the first testing to 16.9% in the second one. In Kukes this interval was 0.7 to 15.5%. High LTFU rates have also been reported in other programs and tend to be more pronounced in newly implemented programs compared to mature ones.[1,16,17] Factors such as rural residence, harsh winters and difficult terrain may further discourage parents from returning for follow-up, delaying diagnosis.[1,18] Mackay et al study, supports the idea that aABR tends to reduce false positives (or referrals), but also highlights that reducing LTFU is multifactorial and not just screening method.[19]

Several strategies could reduce LTFU, including minimizing the number of testing stages until diagnosis, maintaining regular contact with families, implementing home visits and improving tracking systems.[14,15] In the study, although the dataset was relatively small, authors observed notable differences in both 'fail' rates and LTFU percentages between the two sites, favoring the aABR-only algorithm over the TEOAE-TEOAE-aABR sequence. The aABR algorithm requires one fewer stage and, despite higher cost, appears more robust in geographically challenging regions such as Kukes and Pogradec. Other countries, such as Scotland, have successfully implemented similar approaches, with pre-discharge education and nurse-led follow-up improving adherence.[20]

Other tactics for reducing LTFU might be to join the last two tests in one visit (second TEOAR with aABR), to combine hearing screening with other medical checks that the family has to do, like genetics, pediatrics, vaccinations etc. All depends on the structure of each individual screening program or health system characteristics of the country. What works for one program might not be efficient for another one. A recent study of Velarde et al suggests combining several aABR detection algorithms, both in time and spectral domains, in order to improve the detection rate in neonatal hearing screening.[22] Thus, they report a 100% sensitivity and 94.1% specificity in their trial. It demonstrates that the technology of aABR is improving, making it more robust, accurate and effective for screening.

This study focused solely on the outcomes of the screening program and did not include the results of the subsequent audiological assessments for infants who failed the screening. This represents a limitation of the study, along with its relatively short duration. Following the same population over a longer period assuming that cases lost to follow-up are eventually diagnosed, even with some delay would allow for more accurate results and more reliable comparisons.

CONCLUSION

Overall, LTFU remains a persistent issue in newborn hearing screening programs, highlighting the need for context-adapted algorithms and robust follow-up strategies to ensure timely identification and intervention for infants with hearing loss. In this line, an AABR-based algorithm appears to be more robust for remote and isolated areas.

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