



## Implementation of a neonatal hearing screening programme in three provinces in Albania



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### ABSTRACT

**Objectives:** The EUSCREEN study compares the cost-effectiveness of paediatric hearing screening programmes and aims to develop a cost-effectiveness model for this purpose. Alongside and informed by the development of the model, neonatal hearing screening (NHS) is implemented in Albania. We report on the first year.

**Methods:** An implementation plan was made addressing objectives, target population, screening protocol, screener training, screening devices, care pathways and follow up. NHS started January 1st, 2018 in four maternity hospitals: two in Tirana, one in Pogradec and one in Kukës, representing both urban and rural areas. OAE-OAE-aABR was used to screen well infants in maternity hospitals, whereas aABR-aABR was used in neonatal intensive care units and in mountainous Kukës for all infants. Screeners' uptake and attitudes towards screening and quality of screening were assessed by distributing questionnaires and visiting the maternity hospitals. The result of screening, diagnostics, follow up and entry into early intervention were registered in a database and monitored.

**Results:** Screeners were keen to improve their skills in screening and considered NHS valuable for Albanian health care. The number of "fail" outcomes after the first screen was high initially but decreased to less than 10% after eight months. In 2018, 11,507 infants were born in the four participating maternity hospitals, 10,925 (94.9%) of whom were screened in the first step. For 486 infants the result of screening was not registered. For the first screen, ten parents declined, eight infants died and one infant was discharged before screening could be performed. In 1115 (10.2%) infants the test either could not be performed or the threshold was not reached; 361 (32.4%) of these did not attend the second screen. For the third screen 31 (34.4%) out of 90 did not attend. Reasons given were: parents declined (124), lived too far from screening location (95), their infant died (11), had other health issues (7), or was screened in private clinic (17), no reason given (138).

**Conclusions:** Implementation of NHS in Albania is feasible despite continuing challenges. Acceptance was high for the first screen. However, 32.4% of 1115 infants did not attend the second screen, after a "fail" outcome for the first test.

### 1. Introduction

The EUSCREEN study [1] compares the cost-effectiveness of paediatric vision and hearing screening programmes and aims to develop a cost-effectiveness model for this purpose. When completed, the model can be used to assist introduction, modification or disinvestment of a screening programme in a country or region, taking into account the

local circumstances such as professionals available to screen and existing screening pathways. Alongside the development of the model and informed by its preliminary predictions, neonatal hearing screening (NHS) is being implemented in Albania, where hearing screening is not yet routinely performed. The implementation of NHS is being evaluated in three provinces in Albania: Tirana, Pogradec and Kukës. Information gathered by the implementation study will be used to validate and

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**Table 1**  
Studies on implementation of NHS in other countries, their protocol, attendance and referral rates.

Author	Country	Protocol	Attendance screen 1	Invited screen 2	Non-attendance screen 2	Referral diagnostic assessment
Augustine, 2014 [5]	India	aABR, aABR	97.70%	9.10%	17.40%	
Bouillot, 2019 [2]	France	aABR, aABR, (aABR)	99.50%	10.30%	0.10%	2.20%
Caluraud, 2015 [7]	France	OAE + aABR, aABR	99.40%	1.70%	7.90%	0.20%
Chen, 2017 [3]	China	OAE, OAE	93.60%	11.30%	31.30%	0.80%
Low, 2005 [8]	Singapore	OAE/aABR, OAE/aABR	99.80%			6.00%
Olusanya, 2008 [9]	Nigeria	OAE, aABR		14.30%	51.60%	4.10%
Saki, 2017 [10]	Iran	OAE + aABR, OAE + aABR		11.68%	4.23%	1.25%
Sun, 2009 [11]	China	OAE, OAE	90.85%	12.16%	34.32%	7.30%
Uilenburg, 2009 [12]	The Netherlands	OAE, OAE, aABR	94.00%	7.60%	6.30%	2.20%
Wood, 2015 [4]	UK	OAE + aABR	97.54%			< 3.00%

calibrate the model to improve its predictions.

Studies that report on implementation of NHS can be retrospective [2–4] or prospective [5–12] cross-sectional studies. They mainly report on the outcomes related to the screening programme, such as coverage, referral rates and attendance rates. Only a few describe the actual implementation process and describe their experiences with requisites and barriers for implementation [8,9,12]. In most countries where NHS has been implemented, most of the infants are born in maternity hospitals. Acceptance of the first screen has been found to vary between 90.85% (China [11]) and 99.8% (Singapore [8]) (Table 1), however, out of all infants screened in the first screen, the percentage that was invited for a second screen varied between 1.7% [7] and 14.3% [9], whereas non-attendance for the second screen varied between 0.1% in France [2] and 51.6% in Nigeria [9]. Out of all infants who attended the second screen, between 10.75% [10] and 31.3% [2] received a “fail” outcome (the test could not be performed or the threshold was not reached). Between 4.6% [2] and 64.6% [5] did not attend diagnostic assessment.

The Joint Committee on Infant Hearing (2007, 2019) [13,14] defined benchmarks to evaluate the implemented NHS programmes. They recommend to cover 95% of eligible infants and to refer less than 4% of infants to diagnostic assessment. They also recommend for an infant to complete screening within the first month after birth, to complete diagnostic assessment within 3 months after birth and to initiate intervention within 6 months after birth.

Albania has a population of 2.9 million inhabitants and an annual birth rate of about 30,869 [15]. Assuming a prevalence between 1 and 3 per 1000 neonates [16–18], in Albania approximately 30–90 out of the 30,869 infants are born with a permanent sensorineural hearing impairment (HI) of more than 40 dB HL. Without a screening programme, hearing disorders usually are not detected or treated until infants are one to three years old [19,20]. Delays in detection and treatment of childhood hearing impairments have serious consequences for speech, language, cognitive and socio-emotional development [21,22] which can be prevented or significantly reduced by neonatal hearing screening, timely diagnosis and effective early intervention.

Albania is one of the few European countries currently without established NHS. White (2010) [23] reported that of the countries that have an NHS programme, seven screened over 90% of neonates, nine countries screened 25–89% of neonates and 46 countries screened less than 25% of neonates. Sloot et al. (2015) [24] reported that 33 of 38 European countries had a nationwide NHS programme.

An Albanian-Italian study, conducted in 2003–2004 by Hatzopoulos et al. (2007) [25] and an Italian study, conducted in 2009–2012 and funded by MAGIS by Beqiri and Nika (2015) [26] previously studied implementation of NHS in Albania. Hatzopoulos et al. (2007) [25] used a screening programme with two examinations in which infants who failed the first otoacoustic emission (OAE) screen performed in the maternity hospital 2–3 days after birth, underwent a second OAE after four weeks. Infants who failed the second OAE were asked to return a third time after 4–6 weeks for diagnostic assessment using click-auditory brainstem response (ABR). They screened 463 well babies (WB;

healthy infants without any overt diseases) and 1098 infants admitted to the neonatal intensive care unit (NICU). The programme had an attendance of 98,5% for the initial OAE. However, 40–50% of infants did not attend the second OAE. Two (0.18%) NICU infants of the 1098 and none of the 463 WB were diagnosed with a bilateral hearing impairment. Beqiri and Nika (2015) [26] screened 47,341 infants in the three main cities of Albania (Tirana, Shkodra and Fier) with an OAE-ABR protocol for WB and a single ABR for NICU infants. Of the infants screened, 93 had a bilateral hearing impairment. These NHS programmes were not continued. At the time the studies ended, the Albanian government and its Ministry of Health did not consider continuation of NHS to be a priority, resulting in no programme implementation at that time.

We report on the first year of implementation of a NHS programme as part of the EUSCREEN project in three provinces in Albania (Tirana, Pogradec and Kukës) that started on January 1st, 2018.

## 2. Methods

In what follows, we describe the prediction of the cost-effectiveness model, the demography, the screening protocol and methods to assess outcome of implementation.

### 2.1. Prediction of the cost-effectiveness model for Albania

A micro-simulation model has been developed using the Microsimulation Screening ANalysis (MISCAN) model structure [27]. This model was originally developed to simulate the natural history of diseases and to evaluate screening programmes such as for cancer. It was provisionally adapted to evaluate neonatal hearing screening programmes.

In the preparation phase in 2017, the cost-effectiveness model was used to evaluate several possible NHS programme approaches, taking into account local circumstances in Albania such as demography and geography, natural history of hearing impairment in Albania and costs. It proved to be impossible to make accurate predictions as the data needed for the calculation were insufficient or unknown. Since the model was still in the process of being developed, it was a challenge for the preliminary version of the model to take all of the local circumstances into account to calculate the most cost-effective NHS programme.

### 2.2. Demography

NHS was implemented in Tirana, Pogradec and Kukës in Albania (Fig. 1) because they offer an appropriate representation of the general population of Albania with respect to provision of health care in both urban and rural areas and a range of socio-economic circumstances. The NHS programme has been implemented in two maternity hospitals in Tirana: Mbretëresha Geraldine (MG) and Koço Gliozheni (KG), each having their own NICU department. It has also been implemented in the



Fig. 1. Map of Albania marked for Tirana, Pogradec and Kukës.

Map: Philippe Rekeawicz, Emmanuelle Bournay, UNEP/GRID-Arendal (<http://www.grida.no/resources/5360>).

maternity hospital in Pogradec and the maternity hospital in Kukës. The maternity hospitals in Pogradec and Kukës provide limited NICU care. In Albania the majority of the deliveries (> 97%) take place in a hospital [28]. Deliveries at home are rare and occur mainly in remote areas, where no adequate transport is available.

Whereas both Pogradec and Kukës are located in rural and mountainous regions, Kukës is one of the poorest regions in Albania and the inhabitants of the Kukës province are more spread out across the countryside. Parents have to travel far from remote places to reach the maternity hospital and roads are often blocked by snow in winter. Pogradec is situated in the South-East of Albania near the border with Macedonia and Kukës is situated near the border with Kosovo in the North-East of Albania.

### 2.3. Location and timing of screening

The total number of births per year in the maternity hospitals in the selected regions is approximately 11,500. The majority of deliveries (approximately 10,000) take place in the two maternity hospitals in Tirana. The maternity hospitals in Pogradec and Kukës each account for 400–600 births annually.

In Tirana and Kukës, mothers are discharged within 24 h after delivery. In Pogradec, discharge takes place within 48 h after delivery. Unless infants are delivered by a caesarean section, mothers do not return for routine follow up appointments after they have left the maternity hospital. To reach high coverage, the best option was to perform the first screen in the period the mother and the infant were still in the maternity hospital. However, when screening shortly after birth, residual amniotic fluid in the neonates' middle ear has been shown to cause a higher rate of failed tests [29,30]. OAE measures the reactive sound emitted by the outer hair cells in the cochlea to a tone or click produced by the screening device and played in the ear canal. The residual amniotic fluid causes the middle-ear to be less sound-conductive and increases the number of "fail" outcomes during screening. Amniotic fluid disappears when the middle ear is aeriated and more optimal screening conditions may be reached between the third and fifth day after birth [29]. An aABR detects the change in brain wave activity of the auditory brainstem as a reaction to a tone or click. Screening tests performed with aABR are less sensitive to transient conditions such as amniotic fluid in the middle ear, which results in fewer false-positive results [31].

Most existing NHS programmes commonly use a combination of OAE and aABR in a screening protocol that consists of 2 or 3 subsequent screening steps [24,32]. Sensitivity and specificity are between 90% and 100% for both tests [33–35]. Although aABR is more sensitive and specific, less sensitive to transient conditions in the middle and external ear and more likely to detect auditory neuropathy, aABR may be more expensive and time consuming than OAE. Both OAE and aABR can be performed by a wide range of practitioners, provided that they are appropriately trained and have gained sufficient experience.

The Ministry of Health was consulted by the local study coordinator (BQ) to receive approval for the study and to decide on the maternity hospitals that participate in the study. Agreements were made between the Tirana University Hospital Centre (TUHC) and the maternity hospitals in Tirana, Pogradec and Kukës. All infants born healthy as well as all infants admitted to NICU in one of the four maternity hospitals were eligible for NHS.

### 2.4. Screening personnel and training

The first screen was performed in the maternity hospital after delivery and before discharge. Nurses and midwives who were already employed in the maternity hospitals were considered most suited to be trained to perform the screening. Other NHS programmes have trained a wide range of professionals for NHS such as health care professionals, paediatricians, speech language therapists, audiologists or dedicated screeners.

During the autumn of 2017 a training course of three days was set up for screening nurses and midwives participating in the programme. During the first course screeners were taught theoretical information regarding anatomy and physiology of hearing, hearing impairment, hearing (screening) tests, diagnostic assessment, factors influencing the testing, etc. In the second course, screeners were taught about the screening devices, and screeners were able to use the devices. In the last course, screeners were taught about communication skills. They were given information on how to approach and inform parents regarding their participation in the screening programme, how screening can affect the parents, how to explain the test they are performing, and how to explain the result to the parents. After the course, the knowledge of the participants was assessed by administering a test and their



screening skills were evaluated.

During screening the screeners were supervised by a team from the University of Tirana supported by technical staff of the manufacturers. After several months of screening a refresher course was organised so screeners could exchange experiences, get help with problems identified and have questions answered.

An additional one-day course was set up for health care professionals from Tirana, Pogradec and Kukës that did not perform screening: personnel (nurses and general practitioners (GP)) from health centres in the villages, midwives from maternity hospitals that did not perform screening, paediatricians from child consultancy centres and public health workers. During this course these health care professionals were informed about the purpose of the screening programme, how screening is performed, how infants are followed up and what kind of rehabilitation is provided. Furthermore, information on NHS was included in the continuing education courses provided for GP's nationwide. GP's will be informed on the screening outcome through the 'baby book' each infant receives in the maternity hospital. In this book, medical information such as birthweight is recorded, a page was added to document the result of hearing screening.

### 2.5. Information for parents of screened children

The awareness of parents about the importance of early detection and intervention and their acceptance of NHS is of great importance for the success of the programme [36,37]. This has previously been reported for NHS programmes in Albania [25,26]. Leaflets and posters were developed to inform parents about hearing impairment and the effect of hearing impairment on the development of their infants. One leaflet was developed about the existence, rationale for and the course of the screening programme. Another leaflet was developed about what happens when an infant is referred for diagnostic assessment. Information campaigns and interviews were broadcast on television and radio. Furthermore, information videos were produced and played on screens in the waiting rooms of the maternity hospitals.

### 2.6. Outcome criteria for implementation of the screening programme

In this ongoing implementation study, the following criteria for implementation of screening are to be assessed: acceptability, adoption, appropriateness, feasibility, fidelity, costs, coverage and sustainability of the screening programme [38,39]. These criteria can only be assessed after the implementation study has ended in 2020. The definitions and appraisal methods of acceptability, adoption, appropriateness, feasibility, fidelity, costs, coverage and sustainability of the screening programme are presented in the supplemental material (Appendix 1).

A questionnaire was used to measure the screeners' uptake and the acceptability of hearing screening, their level of responsibility, and their attitudes towards parents. It consisted of 155 multiple choice questions in seven domains: general information, attitudes of the screener towards hearing screening, hearing loss, parents of infants screened and subcultures, individual features of the screener and additional questions. This questionnaire was distributed amongst screeners at the start of the screening programme and after one year of screening. They will be asked to fill out the questionnaire again after two years of screening. On-site visits and observations were made to determine the adoption and adherence to the programme, to get an indication of the commitment, understanding and the skills of the screeners. The workflow in the maternity hospitals and the collaboration within the screening teams in the maternity hospitals was studied: how the teams experienced the training courses, how much they remembered after training, how well they used and handled the devices. Additionally, interviews with 8 parents and all 21 nurses who perform screening in each maternity were conducted. In these interviews, parents were asked how they experienced the screening and what information they received. Nurses were asked about their experiences and

opinions regarding the screening programme.

At the end of the implementation study in 2020, the acceptability, adoption, appropriateness, feasibility, fidelity, costs, coverage and sustainability of the screening programme will be assessed.

### 2.7. Database of screened infants

To collect data about the screened infants, a database was developed for the implementation study. Data were filled out by local screeners and the database was monitored by the Erasmus Medical Centre in Rotterdam. All four maternity hospitals were assigned an account in the database. Screeners were able to create their own account that was linked to one of the maternity hospitals. Each eligible neonate was registered in the database by one of the screeners and was assigned an anonymous and unique number. Screeners filled out medical information about the neonates screened such as date of birth, duration of the pregnancy and possible risk factors for hearing impairment. They also registered the date and results of screening as well as the reasons given by parents to decline screening. All three screening outcomes were registered as well as subsequent diagnostics, follow up and hearing-aid fitting. The overall coverage of the programme was estimated from the number of eligible infants screened.

### 2.8. Care pathway and follow up strategies

Following referral from completion of screening, infants received diagnostic assessment at the TUHC or Child Centre for Rehabilitation (CCR) in Tirana. An audiometric booth was built in the TUHC. This room was equipped with one diagnostic ABR device that can be used for both diagnostic ABR and auditory steady state response testing. Another diagnostic ABR device was installed in the CCR.

Because Albania had no formal neonatal hearing screening programme to date, there was little experience with hearing aid fitting in very young infants. In the spring of 2018, a team consisting of speech language therapists, psychologists and paediatricians was trained to help guide parents through the care pathway and provide early intervention. This has helped to create a pathway for children with various levels of hearing impairment and to ensure that the families experience between detection of hearing impairment and appropriate follow up is streamlined.

## 3. Results

### 3.1. Screening protocol

It had been our intention to have the model predict the best screening protocol for the three provinces in Albania. However, the development of the model had to be completed within 7 months and it was not sufficiently finished when the decision on the protocol had to be made and the training of screeners began. Furthermore, because of the sparse and not very detailed data on demographics, current provision of health care, follow up and costs, it proved to be difficult to adapt the model specifically for the situation in Albania and to make differentiated regional predictions for Tirana, Pogradec and Kukës. For example, because of the lack of experience with NHS, the margin of error in the estimation of the costs for screening and diagnostic assessment was very large. This information is difficult to collect, even in countries with established NHS programmes.

As a consequence, decisions made for the screening programme relied much on sound clinical judgement, experience of implementation of NHS in other countries and conclusions made after the previous attempts to implement NHS in Albania. Based on these considerations, local circumstances, expected expenses associated with the screening programme and expected costs for the parents a multi-step programme including aABR was chosen to improve the specificity of the programme and to reduce the number of infants that have to travel to Tirana for full

diagnostic assessment. The sequence of the tests, the location, choice of screening regime and postnatal age at the first screen and the age at repeat screening were considered to be relevant to decide on the NHS programme implemented in Tirana, Pogradec and Kukës. This led to the use of a different protocol in the mountainous region of Kukës as this could reduce the need for repeat screening in recognition of the long journey parents have to undertake to reach the maternity hospital [40]. In anticipation of this problem, the number of steps were reduced and a more specific screening test was expected to result in lower referral rates.

The protocol for WB in Tirana and Pogradec was decided to be an OAE-OAE-aABR test sequence. This protocol was used in both maternity hospitals in Tirana and in the maternity hospital in Pogradec. This sequence was chosen because a three-step programme and the use of an aABR screen offers high sensitivity and specificity. A two- or three-step screening programme and a combination of both OAE and aABR are commonly used across Europe, and higher sensitivity and specificity are beneficial when high numbers of infants do not attend repeat screening. In hindsight, more in-depth consideration about factors influencing low attendance rates would have been advantageous.

For neonates admitted to NICU a sequence of aABR-aABR was chosen to enable detection of auditory neuropathy. Neonates who had a “fail” outcome after the final screen were referred to the ENT department of the TUHC for diagnostic assessment.

To ensure maximum attendance and minimal costs for the first screen, tests were performed in the maternity hospital immediately before discharge. When a “fail” outcome was obtained for one or both ears, a repeat screen was scheduled, after the result of the test was explained to the parents. Within the current programme, infants who lived outside Tirana were not tracked. When parents did not attend repeat screening, they were contacted by the screeners by telephone. When required, the second screen took place in the maternity hospital approximately two weeks after the first screen and the third screen took place in the maternity hospital approximately one week after the second screen. As with WB, the first screen for NICU infants took place before they left the maternity hospital. Parents who lived outside the Tirana area were given the possibility to return for repeat screening in Tirana, however, these parents were not actively reminded. These infants may have been followed up elsewhere.

### 3.2. Screening location and devices

A room was prepared in all maternity hospitals exclusively for hearing screening. A small bed for the neonates was installed as well as screening devices with consumables and a computer with internet connection to upload all screening data and fill out the case record form in the database. Initial problems were that there was too much environmental noise and some screening rooms were too cold for the neonates. No constructional acoustical improvements could be made in any of the maternity hospitals. Therefore, a room was chosen in a quieter part of the maternity hospital and measures were taken to improve screening conditions. Broken doors and windows were replaced to diminish environmental noise and electric heaters were installed to make infants more comfortable during winter months.

Nine Natus Echo-Screen III devices were obtained to perform OAE testing. These devices were distributed between the maternity hospitals in Tirana and Pogradec and the university hospital. Six Interacoustics Titan ABRIS devices to perform aABR testing were obtained for all maternity hospitals. Equipment to perform these tests is calibrated and validated regularly by the supplier to ensure sensitivity and specificity.

### 3.3. Screening personnel and training

Both teams in Tirana consisted of six screeners and an administrator. The local administrators were in charge of organising the screening programme. They made sure sufficient consumables such as

probe tips and electrodes were available. Furthermore, the strategy used to track all infants depended mainly on the local administrator organising the follow up. In Pogradec, four nurses and one administrator took care of screening. The Kukës team consisted of four screeners who alternated shifts to ensure there was always someone present to perform the screening.

The screeners who participated in the project were paid approximately €150 per month by the study in addition to their salary, €400–500 on average. Screening as well as administration took place in overtime. All staff members selected to perform screening attended the training courses and passed the assessment. Screeners were supervised during the first screens and were only allowed to screen independently after they became sufficiently skilful.

### 3.4. Information for parents of screened children

Before the screening test was done, parents were informed about how the hearing screening test works and were able to ask questions before giving written informed consent. After screening, the result was explained. When the infant got a “fail” outcome for the screening test, screeners made an appointment for the next screen one or two weeks later in the maternity hospital.

Most parents were interested in participating in screening when asked to participate in the maternity hospital. Only a few parents declined screening based on their belief that their child could hear or that the test might hurt their child. Screeners reported that the better parents were informed about the project, the less they declined. This is consistent with the wider literature [36,37].

### 3.5. On-site observations

Albania was visited four times by the authors (AB, HH, AG): during the preparation phase, when screening had just started, a few months after screening had started and after one year of screening. Questionnaires were distributed to observe the implementation and to determine the adoption of the programme. During the first two visits and during the last visit all maternity hospitals were visited. During the intermediate visit only the two maternity hospitals in Tirana were visited. Screeners were interviewed. They expressed their ideas and concerns about the programme. Only a few interviews with parents were conducted, because the language barrier prevented an in-depth conversation and translators were not always available. A follow up visit will be made again after 24 months of screening.

During the first months of the screening programme, equipment failures such as malfunctioning OAE devices and broken OAE probes disturbed the screening process. The broken probes and devices were sent back to the manufacturer for replacement but it took several weeks for the equipment to be repaired. This did not delay screening since back-up screening devices were available. Steps were taken to ensure more care when handling the probes and transporting the devices.

Initially, it was difficult for the screeners to place the probe and make sure all conditions were right for screening. Many tests were paused and restarted when screeners experienced difficulties placing the probe, when the probe fell out of the infants ear, the probe got obstructed or when the infant was restless during the test. Initially “fail” outcomes occurred often when screeners had not yet gained sufficient experience in screening. These problems resulted in high failure rates (50–80%) for the first screen. In Pogradec, 78% of infants were referred for repeat screening in the first month. For most maternity hospitals the failure rates for the first screen decreased to less than 10% after eight months. The more neonates were being screened, the better the test seemed to be executed and the lower the failure rate was (Fig. 2).

Differences were noted between the two maternity hospitals in Tirana. The number of infants born in MB was 1.74 times higher than in KG while the number of screeners was the same. This resulted in different outcomes. In KG, screening was organised in a more efficient way

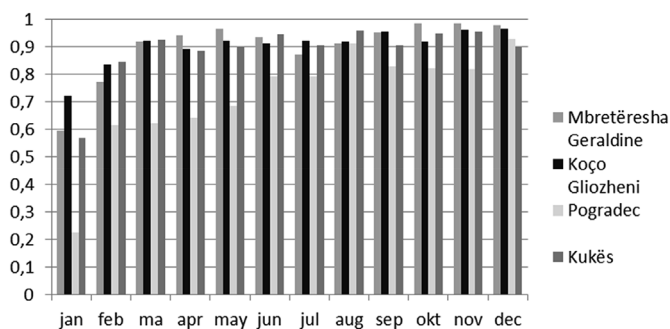


Fig. 2. Pass rate of the first screen for each maternity hospital per month.

resulting in a lower referral rate and less infants not attending repeat screening.

3.6. Screening outcome

All infants born in one of the four maternity hospitals involved in the study were eligible for screening. In 2018, 9583 infants were born in the Tirana province, 1634 in the Korçe province, in which Pogradec is located, and 952 in the Kukës province [15]. In the participating maternity hospitals, a total of 11,507 eligible infants were born. In principal, all infants were invited to participate in screening. For 11,430 infants a record was made in the database but for 486 infants no result of screening was registered. Out of the 10,944 remaining infants, 19 did not participate in the first screen because parents refused screening (7), the infants died (8), or the infant was discharged before screening could be performed (4). Out of all infants who participated in the first screen, an additional 392 (3.4%) did not reach screen completion. Reasons given were: the infant died (21) or had other health issues (8), parents were convinced their child could hear and did not think repeat screening was necessary (9), parents went to a private clinic for screening (17), parents lived too far away from the location where screening took place and were unable to return (61), parents declined screening but no further reason was given (157) or parents did not attend without giving a reason (138). The distribution of reasons given per maternity is shown in Table 2. In the first year, 97,28% of infants completed screening within the first month after birth.

Of the 10,925 (94.9%) infants participating in the first screen, 1115 (10.2%) infants had a “fail” outcome. Six were referred to Tirana for diagnostic assessment and all 1109 remaining infants were invited for the second screen. Out of the 1109 infants invited, 361 (32.6%) did not participate of whom 124 did not provide a reason. The remaining 748 (67.4%) were screened in the second screen, 90 (12%) of whom received a “fail” outcome. Out of these 90 infants, ten were referred to Tirana for diagnostic assessment and 31 did not participate in the third screen, of which 14 did not provide a reason. Forty-nine infants participated in the third screen. A total of 33 (0.30%) infants were referred for full diagnostic assessment in Tirana (Fig. 3). The low referral rate in general and the low referral rates for each screener were looked at in more detail during the second year of screening. Results from the diagnostic assessment will be reported after the implementation study has ended in December 2019.

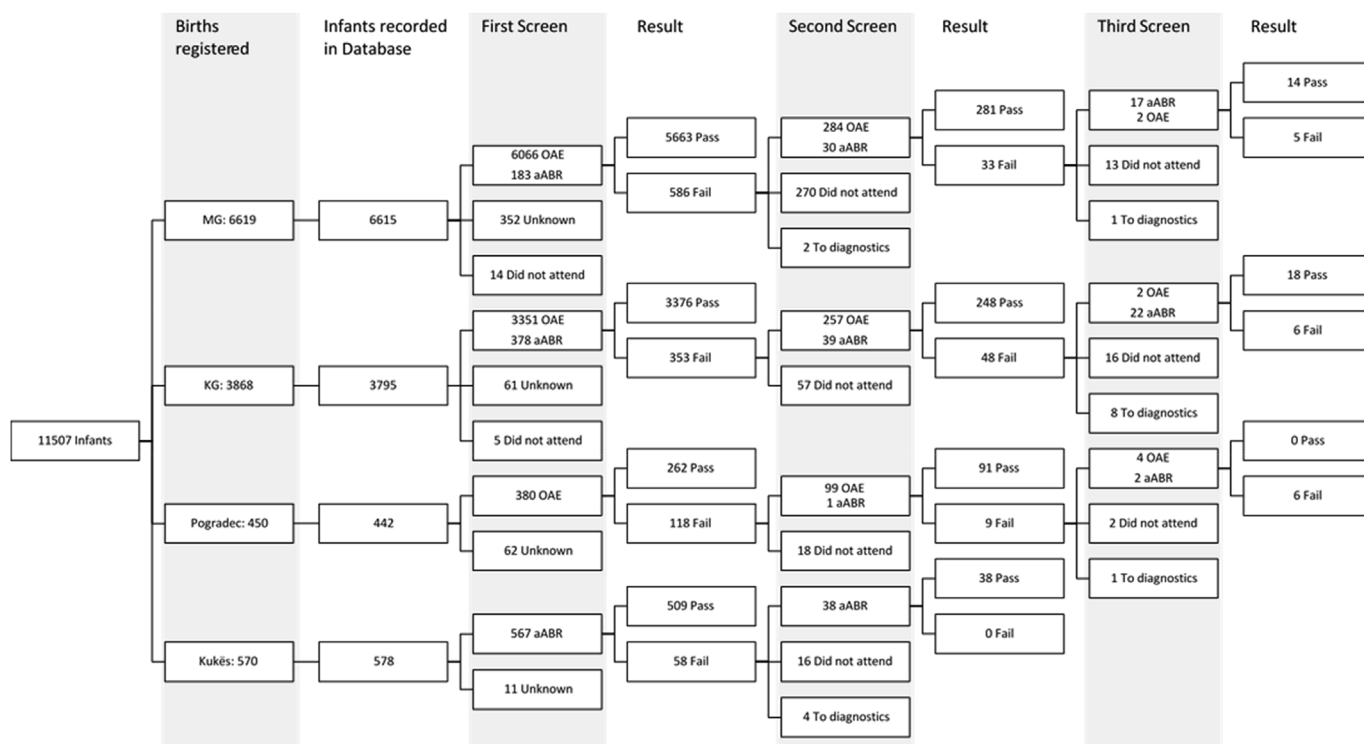
Despite the use of a different protocol, the proportion of infants referred in the Kukës maternity hospital was much higher than should be anticipated [40]. This implies that further training may be needed. This inconsistency has given rise to more in-depth exploration of the situation in Kukës and what is required to maintain screener quality.

3.7. Assessment of screening implementation

Acceptability and adoption by the screeners were assessed by means of the questionnaire. Nineteen of the twenty-one screeners returned a

Table 2 The number of infants screened in the first, second and third screen in four maternity hospitals in Albania. The table shows the number of “Pass” and “Fail” results per screen, the number of infants referred for full diagnostic assessment and the number of infants who did not attend follow up screening including the reasons given. “Fail” results include tests that could not be performed or did the threshold could not be reached.

	Mbretëresha				Geraldine				Koço Gliozheni				Pogradec				Kukës			
	First screen		Second screen		First screen		Second screen		Third screen		First screen		Second screen		Third screen		First screen		Second screen	
	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)
Total	6615		584		32		3795		40		442		118		8		578		54	
Unknown	352	(5.3%)	270	(46.2%)	13	(40.6%)	61	(1.6%)	16	(40%)	62	(14%)	18	(15.3%)	2	(25%)	11	(1.9%)	16	(29.6%)
Did not attend	14	(0.2%)	73		1		5	(0.1%)	12		-		16		1		-		16	
Reason Unknown	-		117		7		-		18		-		2		1		-		-	
No parental consent	5		40		1		5		2		-		17		2		-		-	
Infant lives in different city	1		15		2		-		-		-		-		-		-		-	
Infant screened in private hospital	-		6		1		-		-		-		-		-		-		-	
Parents convinced infant can hear	-		13		1		-		-		-		-		-		-		-	
Infant died	8		6		-		-		-		-		-		-		-		-	
Other health issues	-		6		1		-		-		-		-		-		-		-	
Infants screened	6249	(94.5%)	314	(53.8%)	19	(59.4%)	3729	(98.3%)	296	(83.9%)	380	(86%)	100	(84.7%)	6	(75%)	567	(98.1%)	38	(70.4%)
Pass	5663	(90.6%)	281	(89.5%)	14	(73.7%)	3376	(90.5%)	248	(83.8%)	262	(68.9%)	91	(91%)	6	(100%)	509	(89.8%)	38	(100%)
Fail	584	(9.3%)	32	(10.2%)	5	(26.3%)	353	(9.5%)	40	(13.5%)	118	(31.1%)	8	(8%)	2	(25%)	54	(9.5%)	4	(9.5%)
Fail + refer to diagnostics	2	(0.1%)	1	(0.3%)	1		8	(2.7%)	6	(25%)	1	(1%)	1	(1%)	6	(100%)	4	(0.7%)	4	(9.5%)



**Fig. 3.** The number of infants screened is depicted for the first, second and third screen in the four maternity hospitals in Albania. The figure shows the number of “Pass” and “Fail” results per screen, the number of infants referred for full diagnostic assessment and the number of infants who did not attend follow up screening. ‘MG’: Mbretëresha Geraldine, maternity hospital in Tirana; ‘KG’: Koço Gliozheni, maternity hospital in Tirana; ‘OAE’: number of infants screened with OAE; ‘aABR’: number of infants screened with aABR; ‘unknown’: infants for whom no results were recorded in the database; “Pass”: infants who reached threshold on the screening test; “Fail”: infants for whom the test could not be performed or the threshold could not be reached; “Refer to diagnostics”: refer for diagnostic assessment in Tirana; “Did not attend”: Infants who did not participate in screening due to various reasons.

completed questionnaire at the start of the screening programme. The questionnaire identified a positive attitude from the screeners towards the programme. They indicated their belief screening should be provided to all Albanian citizens. Screeners feel confident and able to participate in the programme, they were ready to take on extra screening tasks because they strongly believed in this screening programme. They found the training course to be very informative but would like to have more follow up training.

After 12 months the questionnaire was distributed again and all twenty-one screeners returned a filled-out questionnaire. The answers provided by the screeners after one year of screening were similar to the previous questionnaire in relation to attitudes and general feelings towards hearing screening. After one year, the screeners felt more confident, did not have to spend as much time preparing for screening and considered screening to be an obvious part of their daily work. Screeners spent more time on informing the parents about hearing screening and noticed parents understood the aim of the screening programme better because of their explanation. The screeners indicated their interest in expanding their knowledge on hearing and screening. The questionnaire will be filled out again after 24 months of screening.

**4. Discussion**

In the first year of implementation of a NHS programme in four maternity hospitals in urban and rural areas in Albania, both the uptake by the screeners and the acceptance by the parents for the first screen was good. Screeners were keen to improve their skills in screening and considered NHS valuable for Albanian health care. The number of “fail” outcomes - the test could not be performed or the threshold was not reached - on the first screen was high in the beginning but decreased to less than 10% after eight months. Devices and probes initially broke often. However, after a “fail” outcome of the first screen, a high number

of infants did not attend the second (32.4%) and third (34.4%) screen, thereby forming the most important obstacle for implementation of NHS in Albania. Possible reasons for not attending the second screen were: not enough information given to parents, administrative problems such as incorrect contact information and the parents could not be reminded of repeat screening, follow up examination took place at another location, the health condition of the infant, parents assumed their infant could hear, travel distances, time between rescreens and costs.

The first screen had high acceptance (94.9%), comparable to acceptance rates in Turkey [6], Italy [41], Poland [42] and Greece [43]. However, 32.4% of infants with a “fail” outcome in the first screen did not attend the second screen for all four maternity hospitals. Non-attendance was especially high in the MG maternity hospital in Tirana and in the maternity hospital in Kukës. For all maternity hospitals together, 34.4% of the infants with a “fail” outcome in the second screen did not attend the third screen. In retrospect, planning all screening steps before discharge, reducing the time between screening steps or choosing a protocol with fewer steps may have prevented these high non-attendance rates.

The aABR-aABR protocol in Kukës resulted in a lower proportion of “fail” outcomes compared to Pogradec, similarly rural but less mountainous and more affluent. This may indicate that the use of a two-step aABR protocol has a higher specificity and reduces the number of “fail” outcomes. The proportion of infants that did not attend the second screen was, however, similar to results found in the other maternity hospitals. The advantage of better specificity for aABR screening may be offset by larger travel times in mountainous areas.

High non-attendance rate were previously found in newly implemented NHS programmes in Italy (29.9% for the second screen) [44], Nigeria (43.1% for the second screen) [45] in Turkey (30%) [46] and in Shanghai (34.32% for the second screen) [11]. Other newly



implemented NHS programmes have reported non-attendance rates that are much lower as for example 0.2–0.4% in France [2,7] 4.23% in Iran [10] and 6% in the Netherlands [12]. Three studies previously reported that non-attendance reduced when experience in screening was gained. In Greece non-attendance reduced from 72.2% to 58.2% after three years of screening. In Colorado 52% of infants did not attend repeat screening between 1992 and 1996 compared to 24% in 1999 [43,47]. More recently, a non-attendance rate of 19% was reported for infants born in Colorado in 2005 and 18% for WB born between 2007 and 2012 [48]. Similar rates of non-attendance have been reported for referral to diagnostic assessment. During the early phase of the UK implementation the target of diagnostics within four weeks of screen completing was not met in 60% of cases. However, after seven years this improved to 17.5% [4].

Acceptance is usually high when the first screen is performed in the maternity hospital before discharge. In contrast to the first screen, attendance to repeat screens may be much lower when parents have to overcome practical problems such as larger travel distances and costs [3,11,12,25,46]. Other reasons for non-attendance described in previous research were also similar to reasons given by parents and screeners in Albania: parents did not understand the importance of screening [49], negative attitudes and lack of awareness in parents or screeners [25,43], programme organisation [43], cultural reasons [26] and type of health insurance [50]. Another possible explanation may be that parents did not receive an invitation for repeat screening after their infant received a “fail” outcome or that the health condition of the infant did not allow for screening to be performed yet. In the Netherlands, attendance was found to be higher when NHS was combined with other health care measures thereby also improving the cost-effectiveness of NHS [12]. In developing countries mothers often do not give birth in regular maternity hospitals [45], and screening in combination with other mother or infant health care services has proven to be more efficient in these countries [45,51]. Similarly, when NHS is organised within an existing maternal and infant health programme and the professionals involved are more successful in explaining the importance to the parents, the number of infants not attending is reduced [2]. To further reduce the number of non-attendants in Albania, the number of screening steps may be reduced or repeat screening may be combined with existing maternal or infant health care services such as vaccination. When NHS will be implemented in the whole of Albania, multiple screening locations will be set up across the country so parents do not have to travel as far.

In previous implementation studies results indicated that parental awareness is very important to increase the acceptance of screening [36,37,45,52]. Therefore, the local study coordinator (BQ) developed leaflets and informative videos that were played in hospital waiting rooms to inform parents. Furthermore, she appeared in several national television and radio shows to explain the importance of NHS. The efforts made to inform parents improved their willingness to participate.

The number of “fail” test results in Albania was high initially. This may have been caused by the early timing of screening or the inexperience in screeners. As is described by Berninger and Westling (2011) [30], amniotic fluid that is present in the neonates’ middle ear in the first days of life may cause increased failure rates when performing OAE screening before the third post-natal day. Despite the fact that a more optimal timing for the first screen may be five days after birth [30], most NHS programmes perform the first screen in the maternity hospital before discharge because high attendance can be achieved. A number of experienced screening programmes (in the UK, USA and Italy) have reported low repeat rates despite screening within 48 h after birth [4,41,42]. Lack of experience with screening may have contributed to high screening repeat rates in Albania, in areas with low birth numbers like Pogradec and Kukës in particular.

Out of 10,925 infants that participated in the first screen, 411 (3.8%) did not complete screening and for 486, no results were registered. This may be an issue with database records not being completed.

The proportion of infants that did not reach screen completion was 4.8% in MG, 2.1% in KG, 5.3% in Pogradec and 2.8% in Kukës. Only 33 infants of the 10,925 who started screening were referred for a full diagnostic assessment. The outcomes of the screening programme show that more than 95% of eligible infants were covered and less than 4% of infants were referred, according to the JCIH recommendations. Low referral rates to full diagnostic assessment may be associated with a high number of false positives (for example due to multiple screens within one step), high numbers of infants lost to follow up and high-quality screening resulting in very low repeat rates between steps. The referral rates for each screener revealed that a number of screeners did not refer any infants. Screeners may choose to repeat screening within one step to be able to give good news to the infants’ parents. This is an issue that may be addressed in further training. Both the high number of infants that did not reach screen completion and the low number of infants referred to diagnostic assessment remain areas of concern. These issues will be followed up in the second year of screening. Low referral rates to diagnostic assessment were previously found in Turkey (0.01%) [6], France (0.2%) [7] and China (0.8%) [3]. The number of infants lost to follow up varied between studies: Caluraud et al. (2015) [7] reported 0.2% for both screening steps while 10% was reported for the same number of steps by Chen et al. (2017) [3]. Infants lost to follow up were not reported by Bolat et al. [6]. Another difference found was the protocol used in these studies. In the study performed by Caluraud et al. (2015) [7], aABR is used in the first step, Bolat et al. [6] uses aABR only in the second step and Chen et al. (2017) [3] does not use aABR in any of the screening steps.

Based on what we have learned from the first year of implementation, both the successes and the continuing challenges, an implementation plan is being developed to extend NHS to the whole country. This plan will take into account the areas in which the programme is still experiencing difficulties. It will combine current experiences with the experiences from previous implementation, that still need to be addressed such as the number of infants that do not complete screening.

In a country where previous implementations of NHS were not continued because NHS was not considered a priority at that time, significant successes were achieved in the number of infants reached in the first screen, improving the awareness in parents and developing a care pathway. However, the number of infants that do not attend repeat screening and consequently, the number of infants that does not complete screening remains too high. This issue will be further explored and addressed in the second year of screening and in the plan for nationwide implementation. Additionally, information gathered during this implementation study will be fed back into the model to improve its prediction for a nationwide NHS programme in Albania.

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## Declaration of competing interest

The authors have indicated they have no financial relationships relevant to this article to disclose. The authors have no conflicts of interest relevant to this article to disclose.

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## Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.ijporl.2020.110039>.

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