Automated auditory brainstem response hearing screening in NICU graduates
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Implementation of an AABR Hearing Screening Programme in the Neonatal Intensive Care Unit.

Based on the article:
*Implementation of an AABR Hearing Screening Programme in the Neonatal Intensive Care Unit.*
H.L.M. van Straaten, E.T.M. Hille, J.H. Kok, P.H. Verkerk and the Dutch NICU neonatal hearing screening working group.
Submitted for publication.
7.1 Abstract

Objective: As part of a future national neonatal hearing screening programme in the Netherlands Automated Auditory Brainstem Response (AABR) hearing screening was implemented in 7 out of 10 Neonatal Intensive Care Units (NICU). The objective was to evaluate key outcomes of this programme: capture rate, first stage success rate, pass/refer rates, rescreen compliance, diagnostic referral rates, age of first diagnostic evaluation and prevalence of Congenital Hearing Loss (CHL).

Subjects: This prospective cohort study collected data of 2513 survivors of the neonatal intensive care period between 01-10-1998 and 01-04-2000 in 7 out of 10 NICU’s in the Netherlands. NICU graduates with one or more risk factors according to the Joint Committee on Infant Hearing were included in a two-stage hearing screening programme.

Methods: The method used was an AABR hearing screening method performed bilaterally by a neonatal nurse before discharge. After a unilateral- or bilateral refer at the first stage a second screening was performed at term. After a second refer conventional ABR was used to establish diagnosis of congenital hearing loss.

Results: A total of 2513 newborns enrolled in the programme were alive at 3 months of age with a median gestational age of 31.6 weeks (range 24-43) and a median birth weight of 1450 gram (range 510-4820). In 25 (1%) cases parents refused the screening. In 4/2513 newborns were initially lost. Finally 2484 newborns have been tested. A 98% capture rate was obtained for the whole programme. After a median post-menstrual age at the 1st test of 33.7 weeks (range 27-54) a pass rate after the first stage of 2284/2513 (92%) resulted. The rescreen compliance after the first test was 92% (184/200). A referral rate for diagnostic ABR of 3.1% (77/2484) resulted. Of the 77 referrals 14 (18.2%) had no CHL, 15 (19.5%) had a unilateral CHL and 48 (62.3%) had a bilateral CHL. Therefore the prevalence of a unilateral CHL was established as 0.6% (15/2484) and for bilateral CHL 1.9% (48/2484).

Conclusion: A two stage AABR hearing screening programme can be successfully incorporated in NICU centres. Prevalence of congenital hearing loss in NICU infants is high. Neonatal hearing screening should become standard clinical practice for all NICU infants as soon as possible.
7.2 Introduction

The impact of permanent hearing impairment on a child and his family can be substantial and long-term. Since the 1950's it has been recognized that early detection and management of [permanent] hearing impairment will help lessen the impact of the condition on the child's social, emotional, intellectual, and linguistic development.\(^1\) In a prospective study Yoshinaga-Itano showed that habilitation of hearing loss before the age of 6 months may result in normal speech and language development, compared to habilitation started beyond 6 months.\(^2\) The long practice of universal distraction hearing screening in some European countries at the end of the first year is now gradually moving towards neonatal hearing screening. The NIH Consensus Development Conference in 1993 and the European Consensus Development Conference in 1998 were strong impulses for efforts to run universal or high-risk register screening programmes.\(^3,4\)

Congenital hearing loss occurs in approximately 0.1% of the normal population and much more frequently in defined high-risk populations (1-2%).\(^5,6\) Because of the high risk of congenital hearing impairment the NIH recommends hearing screening of all NICU graduates before discharge.\(^3\) We previously have shown that the use of AABR hearing screening in the premature newborn is successful even when used in the neonatal intensive care setting.\(^7,8\) An AABR neonatal hearing screening programme could adequately fulfil the principles laid down by Wilson and Jungner in a WHO report\(^9\) (see Table 2).

In this paper results are presented of the implementation of a NICU hearing screening programme in the Netherlands as a first step towards a nationwide neonatal hearing screening programme. This was financially supported by the government. Aim of this study was to investigate capture rate, success rate and referral rates of a two stage hearing screening programme in the NICU, as well as to establish the prevalence of congenital hearing loss in NICU graduates who fulfil the criteria of the Joint Committee on Infant Hearing (JCIH).\(^10\)

7.3 Subjects and Methods

**Subjects**

In the Netherlands neonatal intensive care has been centralized in 10 level 3 NICU's. Neonatal intensive care is provided to 2% of all newborns of whom approximately 70% are inborn and 30% outborn. Seven out of 10 NICU's participated during the study period. Newborns who fulfilled at least one of the criteria according to the JCIH were included in this study (see Table 9).
Chapter 7

Table 9, descriptives and distribution of risk factors in neonatal intensive care graduates enrolled in an AABR hearing screening programme.

<table>
<thead>
<tr>
<th>Risk Factor</th>
<th>Total (range)</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Family history positive</td>
<td>53</td>
<td>2.1</td>
</tr>
<tr>
<td>TORCH/Syphilis</td>
<td>18</td>
<td>0.7</td>
</tr>
<tr>
<td>Cranio-facial anomalies</td>
<td>70</td>
<td>2.7</td>
</tr>
<tr>
<td>Birth weight &lt; 1500 g</td>
<td>1393</td>
<td>54.0</td>
</tr>
<tr>
<td>Hyperbilirubinaemia → Exchange transfusion</td>
<td>48</td>
<td>1.9</td>
</tr>
<tr>
<td>Ototoxic medication</td>
<td>216</td>
<td>8.4</td>
</tr>
<tr>
<td>Cerebral complications</td>
<td>327</td>
<td>12.7</td>
</tr>
<tr>
<td>Asphyxia</td>
<td>841</td>
<td>32.6</td>
</tr>
<tr>
<td>Artificial ventilation &gt; 4 days</td>
<td>761</td>
<td>29.5</td>
</tr>
<tr>
<td>Syndromal malformation</td>
<td>93</td>
<td>3.6</td>
</tr>
</tbody>
</table>

Data of all children were centrally registered at TNO- Prevention and Health (TNO-PG) in Leiden. The study period started at 01-10-1998 and was limited to those newborns consecutively registered until 01-04-2000. All medical ethical committees of the participating NICU's approved the study protocol. All parents were informed prior to the AABR hearing screening with a brochure in their admission packet. Written parental consent was obtained when required. Also, parents were informed for the need to return for rescreening after discharge from the hospital in case of a first failure. The clinical data of the included children are listed in Table 9.

Screening method
The screening method used was the ALGO 1E Automated Auditory Brainstem Response neonatal hearing screening device. (Natus Medical Inc. California USA). This AABR hearing screening technique is extensively described by Jacobson and Kileny. It uses 35 dB HL click stimuli, presented mono-aurally at a rate of 37 pulses/second. The clicks have an acoustic spectrum, which is flat from 750-5000 Hz. After artifact rejection for ambient noise and myogenic activity, an internally programmed template-matching...
algorithm measures ongoing EEG activity for the presence or absence of an ABR. After reaching a likelihood ratio of 160, the ALGO-1E stops collecting data and displays a "pass" for the ear being tested. This indicates that the data collected were sufficient to discriminate between the presence of a "response plus noise", versus the presence of pure noise, or a "no response" condition at better than the 99.80% level of confidence. The AABR hearing screening technique device is safe, simple to operate and quick to administer to large populations. It results in an objective "refer" or "pass" and can be used by personnel who have no special audiological training.8

Screening programme design
The neonatal hearing screening programme in the NICU's was designed as a two-stage hearing screening programme. The child underwent the first test as late as possible during admission on the NICU. During the implementation neonatal nurses were free to choose the most appropriate time for the first test before discharge. In failed cases at the first test with an unexpected prolonged stay in the NICU a "repeated first test" was performed before discharge. Both the first test and the repeated first test forms the first stage of this screening programme. When failed at the first stage, a second stage test was performed in an outpatient setting at least 4 weeks later or when the child reached term. After failing the second stage the child was referred for further diagnostics including conventional Auditory Brainstem Response. Those children who passed the screening were assumed to have normal hearing thresholds at that time. All children enrolled in the regular follow-up programme of each NICU.

Implementation programme
Hearing screening during admission was performed on the ward by a team of 4-7 neonatal nurses of each NICU. Prior to the study period implementation of neonatal hearing screening started with a four-hour central training of these teams. This training not only consisted of the manual skills but also included a theoretical background of the method, the psychosocial impact of the screening for both the parents and the child as well as the need for central programme monitoring. In each NICU the study coordinator informed the medical staff. Also on each location, a research nurse performed clinical lessons for nurses and medical students, guided performance and supported logistic problems during the first two days at the start of hearing screening. Further evaluation of the screening programme took place after 1 month and after 3 months in each participating NICU.

Monitoring of the project
This implementation study was designed as part of a future nationwide hearing screening programme. This programme consists of hearing screening in a target population in the NICU's as well as hearing screening in all other newborns in the well baby clinic setting in
the Netherlands. Central registration provides the opportunity of integration between both hearing screening systems. Of more importance is the tracking of NICU-children who move between NICU's and local hospitals. Even moving between NICU’s is possible due to centralization of special functions like ECMO, neurosurgery, and pediatric (reconstructive) surgery. The monitoring function included a recall function. A reminder was sent:
1. if no test result was obtained within 30 days after registration,
2. if no result of a second screening was obtained 40 days after referral on the first test provided that the newborn had reached term and,
3. if within 40 days no result of diagnostic ABR was obtained after a referral on the second test.
A child was considered lost to follow-up when parents refused further investigations or when the child moved to a non-participating NICU.

Statistical analyses
Data were collected in a central database and analysed with SPSS (version 10.0; SPSS Inc, Chicago, Ill). This statistical software programme was used to obtain descriptive data for success rate screening, first stage pass/refer rates, rescreen compliance, diagnostic referral rates, age of diagnostic evaluation and to establish prevalence of CHL.

7.4 Results

During the study period a total of 2895 consecutive newborns were admitted to the NICU's and centrally registered. The mortality rate before the first stage was 329/2895 (11.4%). Another 53 initially screened newborns did not survive at least 3 months post term and were left out of the analysis. Thus a cohort of 2513 newborns resulted fulfilling one or more of the JCIH criteria. In 25 out of 2513 (1%) newborns parents primarily refused neonatal hearing screening.
Twenty-three children (0.9%) were lost during the study period: 4/23 after no test, 16/23 after 1 failed test and 3/23 after 2 failed tests. Most of the lost newborns after a failed test were transferred to a non-participating hospital and/or had complicated congenital malformations with cardio-respiratory dysfunction in which further screening or diagnostic testing had low priority.
During each stage 0.5% of the tests did not succeed mostly due to myogenic disturbance. The median post-menstrual age at the first test was 33.7 weeks (range 27-54). After the first test a repeated first test was performed in 427/2484 (17.2%) of the children before discharge of the NICU. The combined results of the first test and the repeated first test are from here on presented as the first stage results. Table 10 shows the programme structure and the results of the 2513 children enrolled at the first stage.
Implementation of an AABR Hearing Screening Program in the Neonatal Intensive Care Unit

Table 10  
Programme design and results of AABR neonatal hearing screening in NICU graduates.

The pass rate after the first stage was 92% (2284/2484) resulting in a first stage programme refer rate of 8%. In 21 cases of a prolonged period of illness a diagnostic ABR was performed after the first failed stage in order to reach a reasonable age of detection of hearing loss. The rescreen compliance after the first refer for a second test and/or a diagnostic ABR was 92% (n=184/200). At the second stage 104/163 (63.8%) of the tested newborns passed. The referral rate for diagnostic ABR for the whole programme was 3.1% (77/2484). Fourteen out of 77 (18.2%) had no hearing loss at diagnostic evaluation, 15/77 (19.5%) had a unilateral CHL and in 48/77 (62.3%) a bilateral CHL was established. Therefore the prevalence of unilateral CHL in this study turned out to be 0.6% (15/2484) and for bilateral CHL 1.9% (48/2484). According to the hearing level detected at the first diagnostic ABR 19/48 newborns with bilateral CHL had profound hearing loss (>80dB), 24/48 moderate to severe CHL (40-80 dB), and 3/49 mild CHL (<40dB). In 2 newborns central auditory neuropathy was established.

The false positive rate after the first stage (the number of positive test results when there was no hearing loss) was 4.9%. The false positive rate after the second stage was 0.6% and the positive predictive value 82% (63/77). The true-positive rate (sensitivity) of the programme is unknown.
Corrected post-term age at diagnostic ABR (weeks)
devolution during implementation

![Graph showing corrected post-term age at diagnostic ABR development during implementation.](image)

25 Percentile
75 Percentile
□ Median

corrected post-term age at diagnostic ABR during implementation.

Fig 9 Development of corrected post-term age at diagnostic ABR during implementation.

Before implementation of this neonatal hearing screening programme there was no tradition of early diagnostic work-up in the first months after birth at the audiological centers. During the implementation the mean age of the first diagnostic work-up decreased and finally reached equal or less than 3 months of corrected age. (see Fig 9)

7.5 Discussion

This study is the first European study that presents the results of a programme wise implementation of neonatal hearing screening in NICU graduates supported by the government. The method selected was an AABR hearing screener appropriate for use on the ward and providing the opportunity to screen for auditory neuropathology. Our results show that it is possible to incorporate a two-stage AABR hearing screening programme in Dutch NICU’s with a capture rate of 98%. AABR hearing screening could be easily performed by neonatal nurses. Only 0.5% of all tests did not succeed at any stage even when performed at an early median age at the first test (33.7 weeks). This early age of screening was due to a conflict of interest i.e. testing as late as possible before being transferred and to avoid discharge of unscreened children to the local
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hospitals in a national health care setting characterized by strong limitation of centralized neonatal intensive care facilities. The early age of the first test counts for a 17.2% repeated first tests. We accept this proportion of repeated tests to save the larger expense of efforts made to track unscreened newborns, to avoid the quantity of second tests in an outpatient setting and to avoid the risk of loss to follow-up. Further main results are a pass rate of 92% after the first stage and a final refer rate for a diagnostic work-up of 3.1%. At the first diagnostic audiologic evaluation a prevalence of 1.9% of bilateral CHL and another 0.6% of unilateral CHL was established. After the first stage a false positive rate of 4.9% resulted. The false positive rate for diagnostic work-up (0.6%) was considered low. The rescreen compliance of 92% in this NICU population may be a reason for concern. The unknown consequences of this new facility counts for refusals in the beginning and for children lost to follow up during the programme. Increased knowledge among both professionals and the general public about the introduction of AABR hearing screening in all other NICU's will increase the capture rate and the rescreen compliance from 2001 onwards. This AABR hearing screening programme in NICU's fulfil the quality standards of the National Deaf Children's Society concerning the following markers: initial coverage of 99%, maximum false alarm rate of 20% for NICU/SCBU babies and a false alarm rate after repeated screening tests of 5%. Further follow-up data are necessary to assess the sensitivity of this AABR neonatal hearing screening programme. Those who intend to start a neonatal hearing screening programme should consider the test performance, the tracking of referred newborns, as well as the need for appropriate auditory intervention in those infants identified with CHL. Lack of capacity and experience at the audiologic centers, despite supplied information and the availability of a standard diagnostic protocol at the start of the project, resulted in this study in a learning curve of 1.5 years before the ultimate goal, diagnostic work up ≤ 3months of post-term age, could be reached. Even when hearing loss was established in time not all children started yet with habilitation within 6 months of age. Also, entering a diagnostic work-up before ≤ 3 months of age doesn’t guarantee an equal age of identification. Difficulties with interpretation of the diagnostic investigations and in some cases delayed neuronal maturation contribute for this delay. This programme has a tracking function until the age that a first diagnostic ABR establishes the hearing status in referred newborns. Central administration provides monitoring and quality control of the programme, which is an essential part of this programme. The habilitation of the hearing impaired child should be incorporated in this tracking system to improve the quality of this programme. Implementation of a hearing screening programme is never just the application of a diagnostic procedure. It inevitably includes complex organizational tasks, data management, and many informational problems to be solved. Initial experience with a screening programme for a high-risk population may facilitate the introduction of a more universal programme. Several other arguments contribute to the support of organizing a
neonatal hearing screening programme in NICU graduates. This high-risk group is centralized during a certain period and thus accessible for screening and the prevalence of CHL is a tenfold of the normal population. This makes a reliable screening very cost-effective. Also, it forms an indissoluble part of the neurologic evaluation after a prolonged neonatal intensive care period that sometimes exceeds 2-5 months. This may be too long for an easy access into a universal neonatal hearing screening programme in the well baby clinics. Finally, for those countries with limited financial support, neonatal hearing screening may be limited to the high-risk population. With regard to the high-risk newborn screening, Fortnum and Davis found that almost 30% of the hearing-impaired children have a history of neonatal intensive care and in total 60% have a major risk factor.\textsuperscript{15}

Detection of unilateral hearing loss was not the primary goal of this hearing screening programme. We found a prevalence for unilateral CHL of 0.6%. Although no accepted standard habilitation exist for infants with a unilateral hearing impairment the child may have benefit of the early knowledge of this unilateral hearing loss. Parents are counselled and the infant is monitored yearly to ensure that the hearing status has not changed. In the Netherlands no unilateral amplification or a contra lateral routing of signal instrument is considered when the child reaches school age. The child is properly positioned at school and parents are informed about the risk of hazards of noise exposure and contact sports.

Disadvantage of hearing screening at a very early age may be missing those cases with progressive hearing loss. The process of damage to the hearing can extend beyond the neonatal period. For example, progressive hearing loss due to congenital cytomegalovirus infection and in the group of children with persistent pulmonary hypertension and/or in the group of children treated with extra corporal membrane oxygenation (ECMO) may occur beyond the first year of life.\textsuperscript{16-18} In this study 4 out of 17 initially unilateral referrals turned out to have a bilateral hearing loss suggesting a progressive hearing loss. This strongly supports bilateral rescreening even in unilaterally referred child. It also draws attention to monitoring of hearing loss after neonatal hearing screening. The Health Technology Assessment programme of the Department of Health in the UK concluded that the optimum screening strategy is universal neonatal hearing screening supplemented by a targeted infant screening within 1 year for babies not screened at birth or about whose hearing there is parental or professional concern.\textsuperscript{19} This Dutch NICU neonatal hearing screening programme will be complementary to a universal hearing screening programme in the nearby future. Supplementary target screening in infants at the end of the first year is still subject of discussion in the Netherlands.

The annual costs of running an AABR hearing screening programme to detect bilateral CHL in all NICU's on annual bases are listed in Table 11.
Implementation of an AABR Hearing Screening Program in the Neonatal Intensive Care Unit

<table>
<thead>
<tr>
<th>Costs associated with AABR screening programme in the Dutch NICU's</th>
<th>no infants</th>
<th>total costs ($)</th>
</tr>
</thead>
<tbody>
<tr>
<td>infants screened annually</td>
<td>5000</td>
<td></td>
</tr>
<tr>
<td>first stage costs</td>
<td></td>
<td></td>
</tr>
<tr>
<td>personnel</td>
<td></td>
<td>58886</td>
</tr>
<tr>
<td>disposables (+17,4%)</td>
<td>5870</td>
<td>50314</td>
</tr>
<tr>
<td>overhead</td>
<td></td>
<td></td>
</tr>
<tr>
<td>personnel</td>
<td></td>
<td>60952</td>
</tr>
<tr>
<td>equipment</td>
<td></td>
<td>20622</td>
</tr>
<tr>
<td>supplies</td>
<td></td>
<td>7143</td>
</tr>
<tr>
<td>total predischarge</td>
<td></td>
<td>197916</td>
</tr>
</tbody>
</table>

| refer rate at discharge 8%                                    | 400        |
| second stage costs                                            |            |                 |
| personnel                                                    |            | 6095            |
| disposables                                                  |            | 3429            |

| refer rate for diagnostics 3,1%                               | 155        |
| diagnostic evaluation costs                                   | 25833      |
| total postdischarge costs                                     | 35357      |
| Total pre-postdischarge costs                                 | 233274     |
| average costs per birth                                       | 47         |
| Cost per identified child with bilateral CHL                 | 2456       |

Table 11: Costs associated with running an AABR hearing screening programme to detect bilateral CHL in the NICU population. Data management, monitoring and tracking of the programme are included. Equipment and overhead costs were annualized and amortized over 5 year. *a 17,4% extra use of disposables has been included due to repeated tests at the first stage.

The costs for data management, nation wide monitoring and tracking as well as diagnostic evaluation for all children with a unil- or bilateral failure have been included. This accounts for the total of $47 per infant, which favours the reported costs per infant in the literature.\textsuperscript{20,21} Based on a prevalence rate of 1.9% of bilateral CHL the actual costs of identifying an infant with bilateral CHL were $2456. A two stage screening process is beneficial for reduction of emotional stress for families and the number of time-consuming and costly diagnostic evaluations. The costs listed for first and second stage screening are costs incorporated in the Dutch preventive health care system. No additional charges are made for participants. The costs for diagnostic evaluation are covered by health care insurance in our country with a high participation rate for this.
insurance. Cost savings obtainable are costs from speech and language acquisition, avoidance of special educational programmes and improved quality of life. The cost of not identifying hearing impairment in one person may reach 1 million dollars.\textsuperscript{22}

Considering the cost of screening and the incidence of hearing loss we conclude that the favourable cost effectiveness of this target-screening programme supports continuation. Neonatal Intensive Care Unit (NICU) graduates generally underwent several screening tests to investigate potential existing neurologic sequelae, due to acquired or congenital pathology. Ophthalmologic screening for retinopathy, cranial ultrasound scans as well as neurologic function tests are already routine. Hearing was the sense of which the status only could be assessed using conventional ABR. Conventional ABR, although gold standard for establishing hearing loss, is not available for routine screening. This study has shown that AABR hearing screening can be implemented on a large scale in NICU’s in favour of high quality standards of a modern screening programme. Financial support guarantees programme monitoring, resulting in an optimal tracking of the referred newborn. This should finally result in early habilitation and the improvement of the hearing impaired child.

7.6 Appendix

The members of the Dutch NICU neonatal hearing screening working group are: Henrica L.M. van Straaten, (co-ordinator; Isala Clinics, Zwolle); Paul H. Verkerk MD, Elysee T.M. Hille (TNO-PG Leiden), Wim Baerts, MD, Carin M. Bunkers, Enna W.A. Smink (Isala Clinics, Zwolle), Ruurd M. van Elburg, MD (Vrije Universiteit Medical Centre, Amsterdam) Martin J.K. de Kleine (St Joseph Hospital, Veldhoven), Joke H. Kok, Prof, MD; Adrie Ilsen (Academic Medical Centre, Amsterdam), A.P.G.F. Visser (University Hospital of Nijmegen), Linda S. de Vries, Prof, MD (Wilhelmina Children’s Hospital, Utrecht), Nienke Weisglas-Kuperus (Sophia Children’s Hospital, Rotterdam).

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7.8 References


