Original Article

Audiology 1999; 38:296–302

Neonatal Hearing Screening with Transient Evoked Otoacoustic Emissions: A Learning Curve

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Key Words
Otoacoustic emissions
Neonatal hearing screening
Hearing tests
Hearing-impaired persons
Congenital hearing loss

Abstract
The present paper reports on the implementation of a neonatal hearing screening programme in a private hospital in Belgium. A maternity-based neonatal hearing screening project with transient evoked otoacoustic emissions (TEOAEs) was started in 1993. The cost of the test was not covered by the public health insurance, so the parents had to pay the full cost for screening their child (approximately 30 Euro). Since 1993 the programme strategies have been changed on several occasions to improve the quality and efficacy. A retrospective analysis was performed on: (1) the test pass rate; (2) the coverage; and (3) the number of children who become 'Lost to follow-up' after failing the initial test. The data show a steady learning curve with a time course of several years. They also demonstrate that it is worthwhile and feasible to run a high-quality screening programme in a private establishment.

Introduction

Retrospective epidemiological studies on the prevalence of bilateral congenital permanent hearing impairment (PHI) in Europe have indicated estimates of 1.12 to 2.07 per 1000 live births.[1–3] In these cases auditory deprivation has a serious effect on the speech and language development and thus on the social, emotional and cognitive development of the child. Several authors have demonstrated the benefit of an early identification and intervention (with hearing aids) on the later outcome of language skills in hearing-impaired children.[4–7]

Over the last decade two major new technologies have emerged which make it possible to take objective measures to estimate the likelihood of adequate hearing function in newborn babies: the evoked otoacoustic emissions (EOAEs) and the automated auditory brainstem response (AABR) testing. Furthermore, when compared to the classical infant distraction test which is only possible from around the age of nine months, these new neonatal screening programmes have produced better figures of sensitivity and specificity.[8]

A consensus is growing that neonatal hearing screening is important.[9] Several programmes are being implemented throughout the Western world. In many countries people are still looking for a feasible programme that fits with the national health care system. By necessity most programmes will still be based on existing ones, with slight modifications to cope with the local circumstances. It is essential that these programmes are reported, in order to share the different experiences and to facilitate the organization of new programmes. One such neonatal screening programme, using transient otoacoustic emissions (TEOAEs), was started in St. Augustinus Hospital, Antwerp in 1993.

TEOAEs are low intensity sounds that can be measured by a microphone coupled to the external auditory meatus. They result from energy from the stimulated cochlea passing back into the air contained in the external ear canal. This energy is thought to originate from the outer hair cells whose contractile motions ensure amplification and sharp tuning of the basilar membrane vibration when it is activated by sound. These outer hair cells are the first to be affected by most hearing disorders and ample evidence exists that the presence of EOAEs indicates hearing levels better then 30 dB HL for the frequency range tested.[10,11]

The goal of a hearing screening programme is the early detection and referral of every hearing-impaired child, as defined in the European Consensus Statement on Neonatal Hearing Screening.[9] In order to accomplish this goal, a screening programme needs to include: (1) a high test pass rate; (2) a high coverage; and (3) a stringent follow-up and management.

This paper reports on the evolution of the screening programme and its performance parameters from the start of the programme in 1993 until the end of 1997. It tries to analyse whether the modifications made to the screening protocol actually served the final goal of improving the screening practice. To do that, the test pass rate, the coverage and the number of 'Lost to follow-up' (LTFU) neonates are reviewed at different periods in time.

Received:
November 23, 1998
Accepted:
March 10, 1999

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Part of this paper has been presented under the same title in a poster session at the European Consensus Development Conference on Neonatal Hearing Screening, 15–16 May 1998, Milan.

Material and Method

From 1993 until the end of 1997, 3751 neonates were tested by the University ENT department of St. Augustinus Hospital by means of recording non-linear click evoked TEOAEs. All procedures were made using the ILO88 or ILO288 apparatus (Otodynamics Ltd, England).

The test method was adopted from Bray and Kemp.[12] After insertion and fitting of the neonatal probe with a rubber tip in the outer ear canal, the intensity of the eliciting click resulted in stimuli with a peak sound pressure level in the range of 77–83 dB peSPL. Attention was paid to a low stimulus ringing in the outer ear canal and a flat stimulus frequency spectrum.

The TEOAE recording consisted of a minimum of 20 and a maximum of 260 averaged recordings. The recording was stopped when emissions were sufficiently present to meet the pass criteria, which will be described below. If, after 50 recordings, there was clearly an insufficient response, the test was discontinued and, instead of waiting for 260 recordings, the probe was refitted and the test was restarted.

The opportunity to request an examination was given to the parents of every newborn child by means of a letter, which gave a brief explanation and included a simple registration slip.

In order to anticipate possible parental anxiety in case of test failure, we explained at different stages, both before and after the testing, the relative value of the test result at that stage and what would follow in case of failure.

The initial screening protocol in 1993 was as follows: the neonates were tested in a soundproof cabin or a quiet room at the Audiological Centre of the University ENT department; typically the neonates were brought to the test location by their mother, about 1.5 hours after feeding; testing was done as soon as possible after the parents registered for the test; the pass–fail criterion was a test of qualitative visual scoring based on the Fourier spectrum of the TEOAE wave; in the case of bilateral failure of the first test, the parents were immediately and verbally invited for a re-screen three weeks later; if after six weeks no re-screen had been done, a letter was sent once or twice to the parents to urge them to make an appointment; if a child failed the TEOAE test twice it was referred for a diagnostic ABR test at the age of three months.

Since then, the screening protocol has been changed on several occasions to improve the test pass rate of the first test, the coverage and to reduce the rate of children who become LTFU after failing the first test. These chronological changes are summarized in Figure 1 and divide the total five-year evaluation period (1993–1997) into seven discrete periods which will be referred to in the rest of the report as periods 1 to 7.

Chronologically these changes were the following:

- From May 1993 onwards, the test was done on the last working day before the child left the hospital. This means that, at the earliest, the neonates were tested at the age of three days (period 2).
- From February 1994, the new software (ILO88 V3.94 Quickscreen Test) was available. It allowed numerical assessment of the signal to noise ratio (S/N ratio) in different frequency bands and thus a numerical pass–fail criterion became possible. We concluded that TEOAEs were sufficient if S/N ratio in the frequency bands with centre frequencies 2.4, 3.2 and 4 kHz exceeded 6 dB and S/N ratio in the frequency band with centre frequency 1.6 kHz exceeded 3 dB (period 3). The standard use of a low-frequency cut-off filter was also implemented at that time.
- In June 1994 a consensus was reached with the Neonatal Intensive Care Unit (NICU) to have all their babies screened with TEOAEs. This group consisted of 679 children out of the total of 3751 tested children; 49 per cent of those NICU babies were low or medium care; the other 51 per cent were high care. So, from then on, non-NICU and all NICU neonates were screened (period 4).
- In October 1994 the follow-up strategy was changed. If after four weeks (instead of six) no re-screen had been performed, the parents were urged by letter to do so. If there was still no reaction, then the family doctor and/or the paediatrician was contacted to alert the parents (period 5).
- In December 1995 an information session for general practitioners and paediatricians was organized (period 6).
- In January 1997 the portable ILO288 apparatus became available. From then on neonates could be tested 'on-site' in the maternity ward or intensive care unit. The pass–fail criterion was also changed to an S/N ratio of 6 dB in at least three neighbouring frequency bands drawn from the upper four bands (1.6, 2.4, 3.2 and 4 kHz) and an overall reproducibility exceeding 50 per cent (period 7).

In order to evaluate the efficiency of the test and the whole screening, the 'test pass rate' and 'screen pass rate' will be used

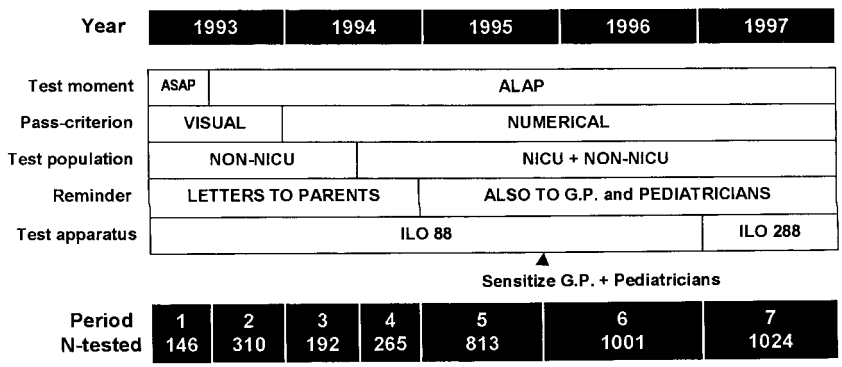

Figure 1. Changes to the screen protocol over the years. ASAP: as soon as possible; ALAP: as late as possible; NICU: neonatal intensive care unit; G.P.: general practitioners.

in the present report. The test pass rate of a hearing screening test is defined as the percentage of children who meet the screening pass criterion at the first test. The screen pass rate of a hearing screening programme, on the other hand, is defined as the percentage of the children who pass at the initial test and the children who, after an initial bilateral failure, pass at the re-screen test which takes place three weeks later.

Although there is still no evidence that detecting unilateral hearing impairment at a very young age is beneficial, both ears were routinely tested. However, in the case of a unilateral fail, we did not encourage the parents to make a new appointment for a re-screen but, if they insisted, they could do so. In the report a 'pass' means that the screening criterion was met unilaterally or bilaterally.

Analysis of the Results

Table 1 and Figure 2 summarize the test pass rate of the initial test with TEOAEs for the NICU, the non-NICU and the total population. For the first three periods there are no pass rate figures available for the NICU population because the systematic screening in the NICU was not then operational.

For the total population, the differences in pass rates from one period to the next are visible but not statistically significant. Figure 3, on the other hand, shows the linear regression statistics for the test pass rate evaluated on a monthly basis over the whole period of five years. The regression line has a slope value of 0.085 per cent per month which significantly differs from zero ($p<0.001$). This means that over a period of five years, the annual increase in pass rate was approximately 1 per cent. During the last 24 months, test pass rates, except on one occasion were always over 95 per cent with an average of 97.3 per cent.

The test pass rate in the non-NICU population was always slightly higher than in the NICU population. The average difference in pass rate for periods 5, 6 and 7 is 1.2 per cent but this difference is not statistically significant (Chi-square, $p>0.05$).

The screen pass rate is the overall pass rate of screen+re-screen and is summarized in Table 2. This pass rate was always high: ≥99.4 per cent, with an overall average of 99.6 per cent.

Figure 4 shows the evolution of the coverage of the screening for NICU and non-NICU populations. At the start of the programme, the coverage was only 18.3 per cent of all live births in the hospital. This number steadily grew to 49.5 per cent for the last evaluation period. The impact of the systematic screening in the NICU is clearly visible on this figure. Its introduction (period 4) added some 7 to 9 per cent to the coverage.

The results for the number of children who were lost to follow-up (LTFU) after failing the initial test are shown in Figure 5. At the start of the programme (periods 1 and 2) the percentage of LTFU was unacceptably high at 50 per cent. From period 3 fewer children were lost and in period 7 this occurred in only 11 per cent of the neonates who failed the initial test bilaterally.

Table 1. Test pass rate (per cent) in the different evaluation periods for the NICU, non-NICU and total population. A 'pass' means that the screening criterion was met unilaterally or bilaterally.

	Period 1	*Period 2*	*Period 3*	*Period 4*	*Period 5*	*Period 6*	*Period 7*
NICU	N.A.	N.A.	N.A.	92.0	95.7	96.4	96.3
Non-NICU	93.2	94.8	92.2	95.3	97.0	97.5	97.6
Total	93.2	94.8	92.2	94.3	96.7	97.3	97.4

N.A.=not available

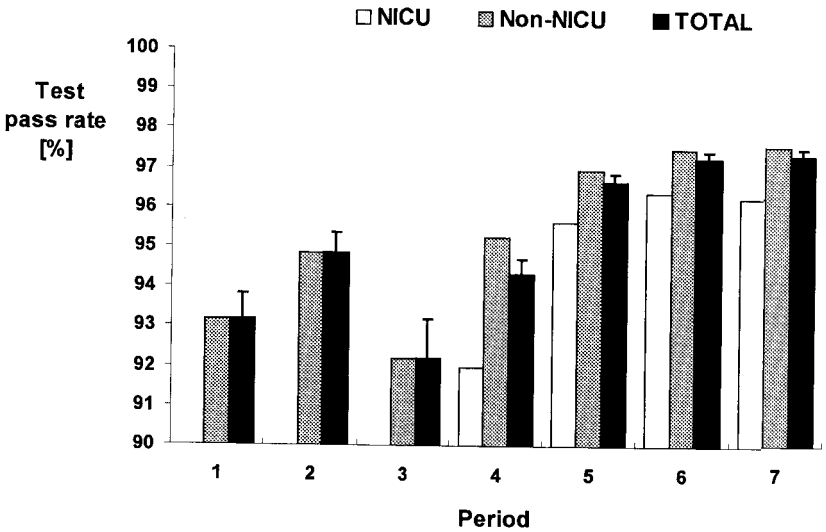

Figure 2. Evolution of the test pass rate for the initial TEOAE test in the NICU, non-NICU and total population. The standard error of the mean is shown for the total population. A 'pass' means that the screening criterion was met unilaterally or bilaterally. NICU: neonatal intensive care unit.

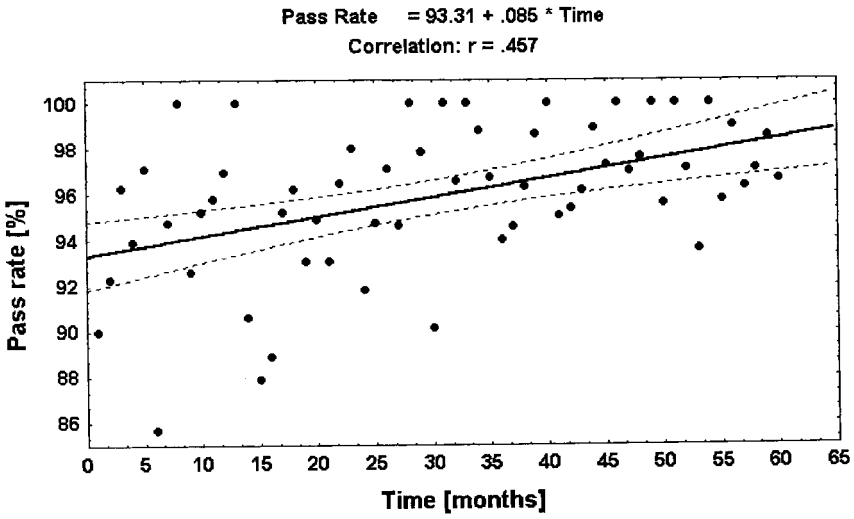

Figure 3. Linear regression statistics on the test pass rate evaluated on a monthly base for the whole evaluation period (five years). A 'pass' means that the screening criterion was met unilaterally or bilaterally.

Table 2. Screen pass rate (per cent) (test+retest) for the total population for the different evaluation periods. A 'pass' means that the screening criterion was met unilaterally or bilaterally

	Period 1	*Period 2*	*Period 3*	*Period 4*	*Period 5*	*Period 6*	*Period 7*
TOTAL	100.0	100.0	100.0	99.6	99.4	99.6	99.4

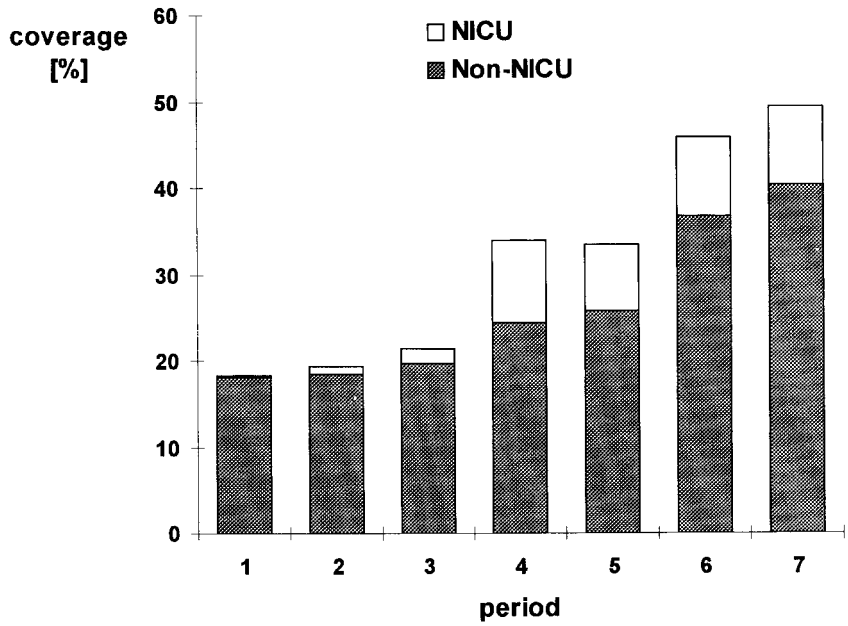

Figure 4. Screening coverage for the different evaluation periods. Black bars represent the coverage attributable to the non-universal screening in the non-NICU ward; the white bars represent the coverage attributable to the universal screening in the NICU ward. As mentioned in the text, all NICU children were screened from period 4 onwards. (NICU: neonatal intensive care unit).

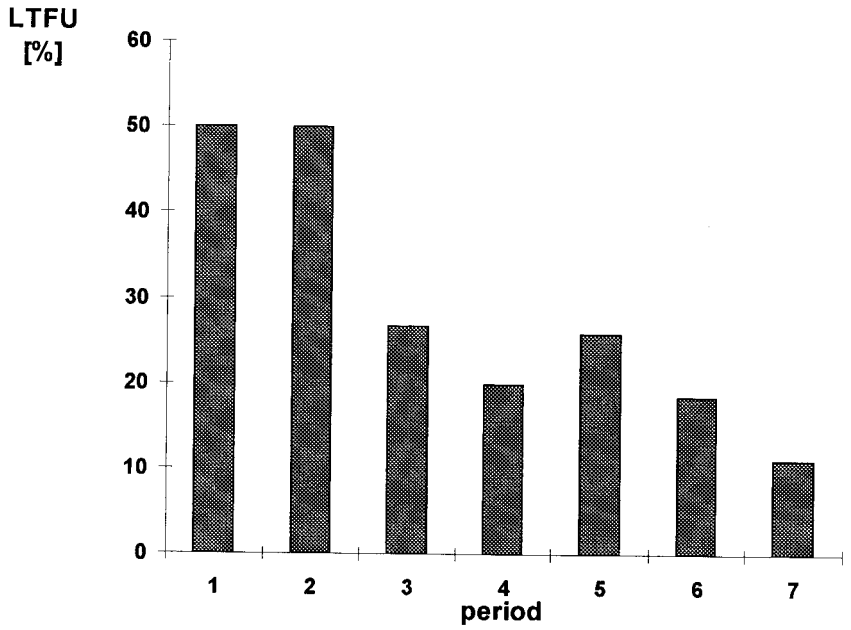

Figure 5. The percentage of neonates who became LTFU after failing the initial TEOAE test for the different evaluation periods. (LTFU: lost to follow-up).

Discussion

The present paper evaluates the implementation of a neonatal hearing screening programme with TEOAEs in a private maternity hospital. It reports on the successive modifications that have been implemented throughout the years and on the learning curve in terms of: (1) pass rate; (2) coverage; and (3) number of children lost to follow-up.

Test Pass Rate

The test pass rate is an important parameter to assess the efficiency of a screening test. To improve this test pass rate, the test moment was delayed from testing as soon as possible after birth to testing as late as possible, i.e. on the last working day before the child leaves the hospital which was typically at about day 4. This is probably the main factor to explain the test pass rate increase from 93.2 to 94.8 per cent for the total population. Other authors have confirmed this finding.[13–15] Debris and vernix are thought to obliterate the external meatus and middle ear in some cases during the first one to three days of life.

After changing from a visual to a more rigid numerical pass criterion a decrease of test pass rate (from 94.8 to 92.2 per cent) was observed. From periods 3 to 6 the pass rate grew to around 97 per cent, although no relevant changes took place during these periods. This increase is thought to be due to a learning effect of the testers. In particular, testers had to learn by experience how the test probe is optimally fitted in the external meatus of the neonate's ear. According to Culpepper, this probe-fit is the single most important factor to maintain low refer rates.[16] Recommendations on the insertion of the probe into the ear canal include: the use of the largest probe tip that can be inserted; the pulling of the pinna outward and upward; and the use of a slight twisting motion.

The test pass rate in the NICU group was always slightly lower then in the non-NICU group. This may be because these neonates were tested at an older age which is associated with the production of more internal noise. Other studies indicate lower test pass rate figures for the NICU population.

Compared with other studies, these test pass rate figures (NICU and non-NICU) are very high. A possible explanation for this may be that in Belgium, in contrast to most other countries, neonates typically reside for five days in the maternity ward and thus the test could be performed as late as day 4 or 5. By that time, the prevalence of obliteration of the external meatus and middle ear becomes extremely low. The fact that all testers were dedicated and trained audiologists may be a second factor to explain these high figures when compared with other studies.

The screen pass rate, which evaluates the efficacy of the total two-stage screening programme, was always high (>99 per cent) with a total mean of 99.6 per cent. This means that only 4 per 1000 screened needed to undergo further diagnostic ABR testing. Half of those were found to be hearing impaired, so only 2 per 1000 screened neonates had a false positive screening result.

Coverage

For a screening programme which requires parents to pay to have their child screened, the coverage is obviously an important factor.

The initial coverage of about 20 per cent rose to around 50 per cent in the last period. The starting of the systematic screening in the NICU in period 4 added some 10 per cent to the total coverage. The organization of an information session to sensitize general practitioners and paediatricians probably caused an increase in coverage of another 10 per cent (period 5 to 6). General awareness of the public about the test and for the ease of testing was probably responsible for the continuous slight increase in coverage. Although a coverage of 50 per cent is not bad in view of the fact that the screening is not free of charge, it is still low from an epidemiological point of view and

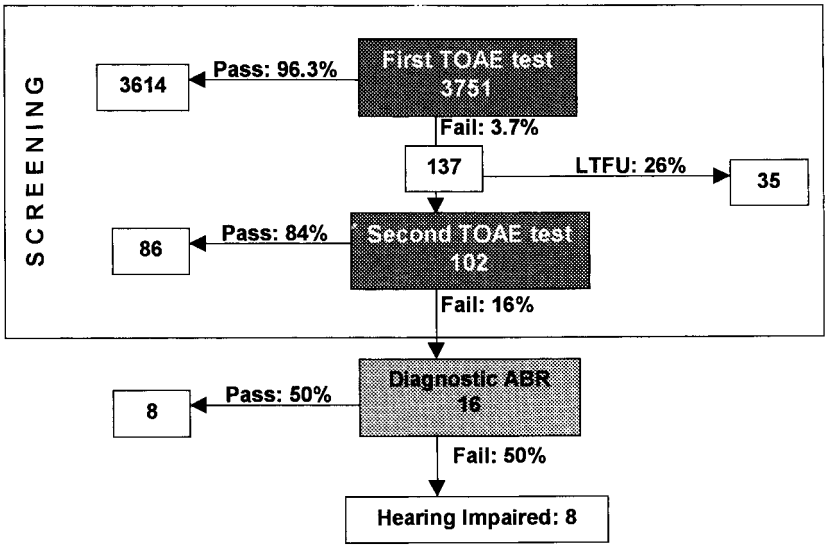

Figure 6. The two-stage screening protocol and diagnostic ABR testing. The pass, fail and LTFU rates are shown for the total of all neonates tested. (LTFU: lost to follow-up).

is considered to be insufficient for a screening programme. The fact that the cost of the test was not covered by the general health insurance and thus had to be charged to the parents is considered to be the main reason for this low coverage.

Lost to Follow-up (LTFU)

The number of children who became LTFU after failing the first test was the major problem in the beginning. One of the children who became LTFU in period 2 was later identified as bilaterally deaf. After identification of this problem (period 2) a more strict follow-up strategy was instigated and this resulted in a drop of LTFU from 50 per cent in periods 1 and 2 to about 25 per cent for periods 3 and 4. Contacting the family doctor or paediatrician to sensitize the parents in period 5 did not immediately result in a decrease in LTFU. The effect of these two changes became visible only in the last period when the LTFU became as low as 11 per cent.

The global evaluation of the screening programme for the whole period, from 1993 until 1997, is shown in Figure 6. Up to the present, 16 of the 3751 screened neonates were referred for diagnostic ABR testing. Half failed this test and were identified as being bilaterally hearing impaired. Of those eight children two were found to have a profound hearing impairment (>95 dB HL), one was severely impaired (loss of 70 dB HL) and five had a moderate hearing impairment (loss of 40–50 dB HL). Three out of these eight neonates (38 per cent) came from the NICU population, the other five from the non-NICU population. All were referred for early rehabilitation and intervention. The other eight children (0.2 per cent) underwent ABR and proved to have normal hearing. One might argue that, in these eight children, unnecessary morbidity could have been caused. This would, however, be limited to complications of ABR and general anaesthesia, which are known to be extremely rare. In our 16 children no such complications occurred.

Conclusions

It is our experience that starting a neonatal hearing screening programme requires permanent quality control and daily efforts to improve the outcome. We believe that our results clearly show that the case is worth the investment and the enthusiasm. A steep learning curve is the result of continuous attention being given to even the smallest details. High quality figures can be reached and reasonable coverage is feasible in a private establishment. The authors believe that this should encourage all clinicians who are in charge of maternity units not to wait for the health care system to implement nationwide programmes, but to start immediately in their own unit. The hearing-impaired child can only benefit from it.

Acknowledgments

The authors wish to thank the paediatricians and nursing staff of the maternity and neonatology departments of St Augustinus Hospital, the audiology students from the Katholieke Vlaamse Hogeschool of Antwerp for their co-operation and Prof. K White (NCHAM) for his support, especially with the management software.

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