Costs of different strategies for neonatal hearing screening: a modelling approach

H C Boshuizen, G J van der Lem, M A Kauffman-de Boer, G A van Zanten, A M Oudesluys-Murphy, P H Verkerk

Abstract

Objective—To compare the cost effectiveness of various strategies for neonatal hearing screening by estimating the cost per hearing impaired child detected.

Design—Cost analyses with a simulation model, including a multivariate sensitivity analysis. Comparisons of the cost per child detected were made for: screening method (automated auditory brainstem response or otoacoustic emissions); number of stages in the screening process (two or three); target disorder (bilateral hearing loss or both unilateral and bilateral loss); location (at home or at a child health clinic).

Setting—The Netherlands

Target population—All newborn infants not admitted to neonatal intensive care units.

Main outcome measure—Costs per child detected with a hearing loss of 40 dB or more in the better ear.

Results—Costs of a three stage screening process in child health clinics are €39.0 (95% confidence interval 20.0 to 57.0) per child detected with automated auditory brainstem response compared with €25.0 (14.4 to 35.6) per child detected with otoacoustic emissions. A three stage screening process not only reduces the referral rates, but is also likely to cost less than a two stage process because of the lower cost of diagnostic facilities. The extra cost (over and above a screening programme detecting bilateral losses) of detecting one child with unilateral hearing loss is €1500–4000. With the currently available information, no preference can be expressed for a screening location.

Conclusions—Three stage screening with otoacoustic emissions is recommended. Whether screening at home is more cost effective than screening at a child health clinic needs further study.

Methods

We calculated the costs of the screening programme itself, including the costs of all diagnostic testing, but excluding costs (and benefits) of treatment, counselling, and management of the cases detected. Calculations are based on the situation in the Netherlands, where a targeted neonatal hearing screening of all children in neonatal intensive care units, covering about 2% of all neonates, is already in place. Therefore only benefits to the population exclusive of these children are considered. As about a quarter of hearing impaired children are admitted to a neonatal intensive care unit, the costs per child detected would be a quarter lower if screening in the neonatal intensive care unit was not performed. Costs are calculated from the perspective of the health care financiers. Costs to society are partly examined in calculating the average time parents spend participating in the screening
The costs calculated are those of a programme that is already running.

The following elements of the screening programme were varied:

1. The screening method (AABR or OAE). Calculations are based on the use of the most suitable commercially available equipment: the ALGO 1E from Natus Medical Inc (San Carlos, California, USA) (AABR) and the EchoCheck from OtoDynamics Ltd (Hatfield, Herts, UK) (OAE).

2. The number of stages (two or three). Two stage screening procedures are customary, but have high false positive rates, especially when using OAE.

3. The target disorder. The current screening programme in the Netherlands aims to detect both bilateral and unilateral hearing losses. As the benefits of early detection of children with unilateral hearing loss are unclear, we also calculated costs for finding only bilateral hearing loss. In this case, only one ear needs to be screened in most cases; only when this ear fails to respond is the second ear screened.

4. Location—home or the CHC. Screening is more likely to be successful when performed at home than at the CHC because an infant is more likely to be peaceful or sleeping in this environment, and screening can more often be performed at an earlier age when the child is in this state. On the other hand, making home visits exclusively for neonatal hearing screening is time consuming and thus expensive. Therefore we only studied home visits that could be combined with existing home visits from health care workers during the first weeks of life—for example, for sampling of blood for phenylketonuria/congenital hypothyroidism screening or the CHC intake visit. When a combined visit was not possible, we assumed that screening took place during a special home visit or that the parents were invited to a screening session at the CHC (fig 1).

We varied the alternatives, starting from a central variant close to the situation tested in earlier feasibility studies which looked promising at first sight. The central variant is two stage screening at the CHC by the CHC nurse, aimed to detect children with both unilateral and bilateral hearing losses. We then changed the factors listed above one by one.

The outcome measures calculated were:
- cost per child screened;
- cost per child detected with a hearing loss of 40 dB or more in the better ear;
- percentage of children referred for further diagnostic testing;
- positive predictive value of a positive screening result (at the end of the two or three stage screening programme) for hearing loss of 40 dB or more in the better ear;
- the average time parents spent attending the screening programme and further diagnostic testing (assuming only one parent attends).

Attendance rates were estimated from experience with the phenylketonuria/congenital hypothyroidism screening, current infant hearing screening, and previous feasibility studies in the Netherlands. Costs of equipment and consumables were based on quotes from the importers, assuming that 350 devices would be purchased. We assumed that equipment would be replaced on average every five years, which is relatively short because of continuing technical developments in this field. Hourly rates for the CHC nurses were based on national norms, previous research into the costs of child health care, the collective labour agreement for the CHC, and the calculation method for costs of home care used by the National Health Tariffs Authority.

Figure 1 Flow through diagram of the screening programme as used in the model. The upper stream represents cases in which the hearing screening could be combined with a regular home visit. The lower stream represents activities carried out only because of the hearing screening. In some variants only the lower stream was used. NICU, Neonatal intensive care unit; CHC, child health clinic.
The elements printed in bold differ from the central variant. Values are mean (SEM).

Results

Table 1 shows that the costs of screening with AABR are appreciably higher than screening with OAE, a conclusion upheld by the sensitivity analysis. Table 2 shows that this is mainly due to the costs of the consumables (disposable earcoupler and electrodes) used with the ALGO 1E. It has been suggested that it is possible to economise by reusing these consumables, but even then AABR would be more expensive because of the higher costs of the equipment despite its higher specificity, which reduces the costs of further diagnostic testing. When the time parents have to invest is considered, however, the extra time involved in using AABR is easily outweighed by the reduction in time necessary for further diagnostic testing. When the total equipment related costs (depreciation and consumables) are left out, OAE screening costs €18.2 per infant screened and AABR screening €17.2. Three stage screening has lower costs than two stage screening, as a third stage only slightly increases the costs of the screening itself, but greatly reduces the costs of further diagnostic testing (table 2).

Scoring for bilateral hearing loss reduces both the costs per infant screened and the number of referrals for further diagnostic testing. Such screening, however, will not detect most children with unilateral hearing loss. On the basis of limited data available,18–21 we estimate that the number of newborn infants with unilateral hearing loss is up to twice the number with bilateral hearing loss. Thus the cost per child detected with unilateral or bilateral hearing loss will be up to three times lower than the cost given in tables 1 and 3, and the extra cost of detecting a child with unilateral hearing loss (over and above the cost of a screening programme aimed at detecting only bilateral losses) is roughly €1500–4000.

The sensitivity analyses showed that the cost of a two stage OAE screening programme taking place at the CHC does not significantly differ from the cost when such screening takes place during combined home visits when possible, and at the CHC otherwise. However, the percentage of children referred for further diagnostic testing is lower and the time parents have to spend is less in the variant with home visits. These differences are confirmed by the sensitivity analysis.

As three stage screening is probably preferable to two stage screening, table 3 presents all possible variants with three stage screening. Again screening using OAE costs less than screening using AABR, but has a lower positive

<table>
<thead>
<tr>
<th>Method</th>
<th>Number of stages</th>
<th>Aim</th>
<th>Place of screening</th>
<th>CHC</th>
<th>CHC</th>
<th>CHC</th>
<th>Home visits only</th>
<th>Home visit + CHC</th>
</tr>
</thead>
<tbody>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of both unilateral and bilateral losses (screening of both ears)</td>
<td>20.9 (2.3)</td>
<td>29.4 (3.0)</td>
<td>18.1 (1.7)</td>
<td>17.0 (1.9)</td>
<td>23.7 (2.9)</td>
<td>20.1 (2.1)</td>
</tr>
<tr>
<td>AABR</td>
<td>2</td>
<td>Detection of both unilateral and bilateral losses (screening of both ears)</td>
<td>29.0 (6.4)</td>
<td>40.7 (9.4)</td>
<td>25.0 (5.4)</td>
<td>23.5 (5.9)</td>
<td>32.1 (8.2)</td>
<td>27.4 (6.0)</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>3.6 (1.0)</td>
<td>2.3 (0.6)</td>
<td>1.8 (0.6)</td>
<td>2.2 (0.8)</td>
<td>2.0 (0.8)</td>
<td>2.4 (0.9)</td>
</tr>
<tr>
<td>OAE</td>
<td>3</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>2.2 (0.7)</td>
<td>3.5 (1.1)</td>
<td>4.5 (1.6)</td>
<td>3.6 (1.4)</td>
<td>3.9 (1.7)</td>
<td>3.3 (1.4)</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>1.2 (0.1)</td>
<td>1.2 (0.1)</td>
<td>1.2 (0.1)</td>
<td>1.1 (0.1)</td>
<td>0.9 (0.5)</td>
<td>0.4 (0.07)</td>
</tr>
</tbody>
</table>

The elements printed in bold differ from the central variant. Values are mean (SEM).

OAE, Otoacoustic emissions; AABR, automated auditory brainstem response; CHC, child health clinic; NICU, neonatal intensive care unit.

Table 2 Breakdown of the costs per child screened for the variants in table 1 (in €)

<table>
<thead>
<tr>
<th>Method</th>
<th>Number of stages</th>
<th>Aim</th>
<th>Place of screening</th>
<th>CHC</th>
<th>CHC</th>
<th>CHC</th>
<th>Home visits only</th>
<th>Home visit + CHC</th>
</tr>
</thead>
<tbody>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of both unilateral and bilateral losses (screening of both ears)</td>
<td>7.3</td>
<td>8.9</td>
<td>7.5</td>
<td>5.9</td>
<td>11.5</td>
<td>8.0</td>
</tr>
<tr>
<td>AABR</td>
<td>2</td>
<td>Detection of both unilateral and bilateral losses (screening of both ears)</td>
<td>2.3</td>
<td>4.3</td>
<td>2.3</td>
<td>2.3</td>
<td>3.0</td>
<td>3.0</td>
</tr>
<tr>
<td>OAE</td>
<td>3</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>0.5</td>
<td>7.9</td>
<td>0.5</td>
<td>0.5</td>
<td>0.5</td>
<td>0.5</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of unilateral hearing loss (screening of both ears)</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.7</td>
<td>0.0</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>2.2</td>
<td>2.0</td>
<td>2.2</td>
<td>2.0</td>
<td>1.6</td>
<td>1.6</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>1.5</td>
<td>1.3</td>
<td>1.5</td>
<td>1.5</td>
<td>2.0</td>
<td>2.0</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>1.1</td>
<td>1.1</td>
<td>1.1</td>
<td>1.1</td>
<td>1.1</td>
<td>1.1</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>3.1</td>
<td>3.8</td>
<td>2.9</td>
<td>3.7</td>
<td>3.4</td>
<td>3.9</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>Detection of bilateral losses only (screening of both ears)</td>
<td>20.9</td>
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<td>23.7</td>
<td>20.1</td>
</tr>
</tbody>
</table>

The elements printed in bold differ from the central variant.

OAE, Otoacoustic emissions; AABR, automated auditory brainstem response; CHC, child health clinic.

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predictive value. Screening at home has a higher positive predictive value than screening in the CHC. Screening with OAE aimed at detection of both unilateral and bilateral hearing loss yields rather low positive predictive values, even in the most favourable cases. Some of the false positives, however, will be children with mild hearing loss (less than 40 dB in the better ear).

**Discussion**

In this study we compared the cost of finding a child with bilateral hearing loss using different screening programmes (cost comparison), but we did not carry out a full cost effectiveness study of neonatal hearing screening. After all, the implications in terms of costs and benefits are identical for each child detected, regardless of the method, and therefore it is not necessary to calculate full cost effectiveness of each programme to decide on the most cost effective programme. However, this reasoning rests on two assumptions that may be disputed. The first is that the number of cases missed by the programmes (which could be at considerable human and social cost) is comparable in each programme. Cases missed can be due to non-compliance (of parents or doctors) with the screening programme, or to false negative test results. The non-compliance was estimated (based on compliance in other neonatal screening programmes) to vary between 1.1% and 3.5% in the different screening programmes, and was mainly determined by whether the screening included home visits or not. The number of false negatives may depend on the method used (OAE or AABR). To date, however, data on the false negative rates of these methods are almost absent, as such data require follow up experience of tens of thousands of screened children. For children admitted to neonatal intensive care units, AABR is more sensitive, both theoretically and based on case reports, as it also detects hearing loss generated in the pathway between the cochlea and the brainstem. However, this type of hearing loss is probably extremely rare in children who have not been admitted to neonatal intensive care units. The second assumption is that all programmes detect the children early enough. Introducing a third screening stage or postponing screening until the child is old enough to visit a CHC would cause some delay. However, when the screening is well organised, these delays should not prevent detection before the age of 3 months.

Our approach is mainly financial, and therefore aspects such as the emotional cost to parents of false positive results or the detrimental consequences of false reassurance in the case of a false negative are not included. However, such arguments are important in deciding which programme should be implemented.

The calculations presented here are based on the situation in the Netherlands, which in many respects (price of equipment and consumables, price of labour) is similar to that in the rest of Western Europe. The most relevant aspect is probably the high population density of the Netherlands, making travelling distances relatively short. Nevertheless, in most countries there are large urbanised regions that are as densely populated as the Netherlands.

Screening with AABR with current equipment (ALGO 1E, requiring costly consumables) is more expensive than screening with automated OAE, but has lower referral rates. Therefore, at present, AABR can only be recommended when the benefit of lower referral rates is judged to be very important or when AABR is judged to have a significantly higher sensitivity.

The cost of screening for bilateral hearing loss only is lower than the cost of screening for unilateral loss also, but such screening will miss most infants with unilateral hearing loss. The value of detecting children with unilateral hearing loss is controversial, as there is no compelling evidence that early detection leads to a relevant gain in health and development. We showed that the extra cost of detecting a child with unilateral hearing loss (over and above the cost of a screening programme aimed at detecting only bilateral loss) is roughly €1500–4000. It is a matter of opinion whether it is worth paying this price. In the Netherlands it was decided to include screening on
unilateral losses in further implementation studies.

Screening at home during the first two weeks of life results in lower referral rates than screening at a CHC three weeks later. Whether screening at home is also financially opportune remains to be seen. Further field experiments will have to decide this issue.

In England a cost of £13.9 (about €23) per child screened was calculated for a universal programme of neonatal hearing screening,\(^1\)\(^2\) which is within the range we calculated for a two stage OAE based screening programme, but lower than a two stage ABR based screening programme. In the United States, the cost of a screening programme using OAE in the first stage and AABR in the second stage was estimated to be $26.05 (about €30) per child screened,\(^3\)\(^4\) and that of a screening programme using AABR to be $17 (about €20).\(^5\)\(^6\) Our estimates for two stage screening programmes lie between these two findings. So, despite the fact that in our community based screening programme more equipment is needed (about one screening device per 500 children born annually), screening can be performed at costs similar to those calculated for these programmes in the United Kingdom and United States. We speculate that, in the CHC model, better use of equipment is needed (about one equipment for two stage screening programmes).

In conclusion, we recommend three stage screening with OAE. Whether screening at a CHC three weeks later. Whether screening at home is also financially opportune remains to be seen. Further field experiments will have to decide this issue.

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