

Feasibility and implementation of neonatal hearing screening in the Netherlands

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Introduction

In the Netherlands about 60 regionally organised preventive health care organisations are taking care of the legally mandated immunisation programs for babies and toddlers, as well as the screening programs for metabolic and sense organ dysfunctions. These organisations run a fine maze system of Well Baby Clinics (WBCs), one is in every baby's neighbourhood. Babies go there regularly, their developments to be monitored by nurses and youth health physicians. This structure of preventive health care is possible and affordable thanks to the high population density in the Netherlands.

Since the mid sixties of the last century hearing screening is done in the WBCs for babies aged 9-11 months. This is a three-stage screen by distraction testing. The refer rate of this screen is about 7 %, which is high. Originally this screen was installed for finding the babies with a permanent hearing impairment; in practice nowadays most of the referred babies have otitis media with effusion. The need to treat this condition was strongly in dispute in the Netherlands over the last decade. These referred babies with non-permanent hearing loss were and still are polluting the screen that was installed to pick up the babies with permanent hearing impairment. This degrades the quality of the screen and the compliance of parents and doctors with referral for diagnostic assessment after an alarming outcome of the screen. The degrading quality of the present screen and the availability of methods to screen the ear function of neonates were the motives for me and a lot of others to carry out and push for feasibility studies on neonatal hearing screening.

A hospital based neonatal hearing screening program is not feasible in the Netherlands, because the coverage of the program would be too low for a population screen. In the Netherlands 30 to 35 % of the babies are born at home. In most of the remaining 65-70 % of cases the mother and baby are discharged from the hospital within 12 to 18 hours. A hospital-based screen would probably cover 50 % of the neonates at maximum. Pregnancies are guided and controlled from 3 months through one week postpartum by certified midwives, general practitioners, or a gynaecologist for a small proportion of the women. At some point during pregnancy the pregnant woman decides whether the delivery will be at home or in the hospital, whatever is judged to be safe and practical. In case of a home delivery a last minute transfer to a hospital is always an option as almost everybody lives within a 30 minute drive from a hospital. Direct postnatal health care for the mother and the baby is given at home by a bedside nurse and the midwife or the general practitioner during about 1 week post partum.

Given this structure of perinatal health care, in the Netherlands neonatal hearing screening has to be done either at home or in a WBC.

Feasibility trials

During our own studies on OtoAcoustic Emissions (OAEs) in neonates we got convinced, somewhere in 1992, that it was technically possible and affordable to do a universal neonatal screening. A nationwide screening program cannot be managed from a university hospital, but a continuous cooperation with the organisations responsible for the existing hearing screening program was possible. Our advice was to combine an OAE-screening with the PKU-screen, which is also done at home.

At about the same time two paediatricians were initiating hearing screening trials by Automated Auditory Brainstem Response (AABR) recording, both in Neonatal Intensive Care Unit

(NICU) babies and in healthy babies in the WBC. Since 1995 results of feasibility trial were published (mainly in Dutch language papers and reports). These studies are summarised below.

Trial 1 was carried out by van Zanten & van der Lem (1998). A total of 487 neonates were tested both at home and at the WBC. The aim was to find out which of the two was the better test site. A screen-pass was defined as 'OAEs proven to be present in both ears'. Testing was done by audiotically skilled personnel and with the ILO288 device (OtoDynamics) with expert judgement of the recorded OAEs. Recordings were broadband (0.7 to 6.25 kHz) and without the Real Time Low Frequency cut-off. The results are given in Table 1. It is clear that the WBC condition, with an overall refer rate of 5.8%, is the worse of the two, compared to 1.6% in the home condition. But it is also clear that repeat screening is better. If the screening was repeated once the overall refer rate dropped to 0.6%.

Site of testing	Home	WBC	Both
N-included neonates	545	487	487
%-Bilateral technically successfully tested	98.7	94.3	98.8
%-Bilateral technical fails	0.4	3.1	0.2
%-Bilateral proven OAEs	92.1	86.4	97.3
%-Unilateral proven OAEs	6.1	7.8	2.1
%-Refer due to uni- or -bilateral technical fail or OAE-absence	1.6	5.8	0.6

Table 1: Results of a two repeat OAE-screening trial in 1995. It was attempted to screen each neonate twice, once at home and a second time at the WBC.

Trial 2 was done by Oudesluys-Murphy & Harlaar (1995). AABR screening at home was done with one repeat trial after 1 week. The ALGO-1 (Natus) device was used. WBC nurses did the screening. A screen pass was defined as 'AABR recordable in at least one ear'. The results are summarised in Table 2.

N-included neonates	284	Incomplete screen in 7
N-neonates with a complete screen	277	100 %
N-unilateral or bilateral pass	266	96 %
N Bilateral refer	11	4 %

Table 2: result from a two repeat AABR trial by Oudesluys-Murphy & Harlaar (1995).

Trial 3 was a comparison of OAE versus AABR screening (Lem et al, 1998). The question to be answered was which of the two methods was most reliable and successful as carried out by WBC nurses. Each neonate included was screened in the WBC with either the ILO288 (OtoDynamics) device as OAE-screener (again broadband recording, with low-frequency real time cut-off and automatic scoring as to pass or refer) or the ALGO-1E (Natus) device as AABR-screener. A screen pass was defined as 'ABR or OAE proven to be present in both ears'. In case of refer a second test was done 1 week later, again in the WBC. The OAE-screening equipment (PC and software) gave significantly more technical problems and concomitant screen fails than the AABR equipment. During the trial the decision was made to stop the OAE-part until these problems could be diminished. As a consequence also the training of nurses to carry out the screening was revised. The technical problems with the OAE-screening method are reflected in the results, given in Table 3, by a lower number of OAE-tested neonates and a higher OAE- than AABR-refer rate. It was also clear that recruiting non audiotically-skilled screeners had a negative effect on the overall refer rate. A comparison with the result of trial 2 showed that using the ALGO-1E in stead of the ALGO-1 as AABR screener gave a better overall refer rate.

	AABR	OAE	Total
N-included neonates	633	355	984
N-referred	6	10	16
%-Referred	1	2.7	1.6

Table 3: Results from a study by Lem et al (1998). WBC nurses screened neonates at the WBC by either OAE or AABR recording.

Another part of this trial was the design and testing of automated administrative procedures of a screening program. Automated administrative procedures are needed to keep down the costs and to improve the program quality. It is virtually impossible to control the track of the invited and referred neonates by hand. The risk of losing track of one in very many neonates can be minimised with automation.

As another part of this trial a model study was done on costs of neonatal hearing screening and diagnosis by Boshuizen et al and published in Dutch also in 1998 and internationally in 2001. Very concisely summarised the result is that the difference in costs per diagnosed hearing impaired neonate is due to the difference in costs of the disposables of OAE versus AABR testing. OAE screen stages have a higher refer rate than AABR screen stages, but cause much less costs per test. Furthermore it was concluded that it was more cost effective to do a three stage rather than a two stage OAE-screen, due to the high costs of the diagnostic follow-up. So a three stage OAE-screen was advised for a larger and final trial.

Trial 4 was a study on various implementation forms of a three stage OAE screen and the merits of the automated administrative system. The merits of various forms of implementation had to be studied, because the WBC-regions are differentially organised. A uniform implementation is not possible. In some regions the PKU/CHT screen is carried out by the WBC-nurse, in other regions by the midwife or the general practitioner. The sheer number of professionals that would have to be necessarily trained and equipped excluded the latter two professions as candidate hearing screeners. In the WBCs in total a much lower number of professionals (nurses) with screening as a special task could be appointed.

In three differentially organised regions a year cohort of neonates were screened by a three stage OAE-screen. The EchoCheck (OtoDynamics) device was used, designed for Automatic-OAE screening. This type of equipment automates a number of tasks of a screener, like checking whether the ear-probe fit is good enough for recording OAEs. Notebook PCs and a central computer were used for the administration.

In two regions about 1600 neonates had their screening stage(s) at home. In one region (Alkmaar) the first stage was combined with the PKU/CHT screen at the age of 4-8 days. In the other region (Rivierenland) the first stage was done at the age of 2-4 wks, the regular 'first contact' between WBC and baby. In the third region (Brabant NE) in about 1600 neonates, all three stages were done at the WBC. The results of this screening program are given in Table 4, only as to the mere screen figures. Many other aspects of the program have been evaluated. For instance, also the parents' opinion about the program and the anxiety caused by the results has been assessed.

Stage	Region	Alkmaar	Brabant NE	Rivierenland	Total	%	%
0	Population	808	1579	949	3336		100
1	Included	770	1482	885	3137	100	
	Lost	38	97	64	199		6.0
	Fails	46	118	73	237	7.6	
2	Included	46	106	70	222	100	
	Lost	0	12	3	15		0.5
	Fails	16	57	34	107	48.2	
3	Included	16	51	33	99	100	
	Lost	1	6	1	8		0.2
	Fails	11	34	24	69	69.7	
Total	Completed	769	1464	881	3114	100	
	Lost	39	115	68	222		6.7
	Referred	11	34	24	69	2.2	
Coverage (%) of population		95.2	92.7	92.8	93.3		
Referred (%) of completed		1.4	2.3	2.7	2.2		

Table 4: Figures describing the coverage and yield of the 3-stage A-EOAE-screen

It is clear from this table that large differences between regions do exist, which are probably due to differences in implementation.

Trial 5 was started because the refer rate in trial 4 was still not as low as can be theoretically with current automated methods. After trial 3 it was concluded that adding a 3rd screen stage, by A-OAE-testing, was cost effective. In trial 5 it was evaluated what the effect of a 3rd stage by AABR-testing would be. This was a smaller scale trial only carried out in the region of Alkmaar; so all tests were done at the baby's home. The results are shown in Table 5.

Population	808		765	
Stage	Program 1	Fail rate (%)	Program 2	Fail rate (%)
1	A-OAE	6.0	A-OAE	5.1
2	A-OAE	2.0	A-OAE	1.3
3	A-OAE	1.4	AABR	0.4

Table 5: Effect on program refer rate of AABR as 3rd screen stage.

First, the results show that indeed exchanging the A-OAE for an AABR test as a 3rd stage in this region reduced the overall refer rate significantly from 1.4 to 0.4% , which is about as good as can be expected. Secondly, these result show that the screeners in this region did in the second cohort a better job, the 1st stage fail-rate reduced from 6.0 to 5.1 % and the 2nd stage fail-rate from 2.0 to 1.3 %. This is the effect of learning.

Discussion

Thinking about the yield of a universal neonatal hearing screening program in the non-NICU group of neonates, one must take into account that the NICU-group of neonates will yield between 30 and 40 % of the hearing impaired neonates. Even a perfect screening program in only the non-NICU group will only identify 60-70% of the target group. The estimated prevalence of bilateral neonatal permanent hearing impairment in childhood in the Netherlands is 0.14 %. A hearing screening program for non-Nicu group of neonates can at best identify about 0.1 % of this

population as 'probably hearing impaired'. In the Netherlands a screening program for the NICU-group is already running (van Straaten, 2001). And the structure of perinatal health care only allows for a community-based type of universal neonatal hearing screening program for the non-NICU group.

It was shown in the trials described above that it is quite possible to do neonatal hearing screening at the baby's home and that surprisingly this condition gives the best overall screening program results. This statement holds irrespective of the type of screener, being an audiotically skilled or unskilled professional. It was shown too that automated hearing screening devices enable a high quality screening program to be carried out by nurses of the Dutch Well Baby Clinics.

Probably the most cost-effective program for the Netherlands is a three-stage screen, starting with two stages of automated OAE-testing and with automated ABR-testing as final stage. Convinced by the results of the trials, the Dutch government has decided to finance the transgression from the existing universal hearing screening at the age of 9-11 months to a neonatal screening. The start of the region-by-region exchange was July 1, 2002. The first results from the regions that made the change already are at least as good and probably even better than those of the trials.

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