A Two-stage Bipodal Screening Model for Universal Neonatal Hearing Screening

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Objective: A model is proposed for universal neonatal hearing screening.
Methods: The screening model is two-staged because it consists of a first test and, in case of failure (1.4% of the subjects), of a retest 3 weeks later. It is bipodal because it involves both the hospital audiologic department and a central Well Baby Organization. The idea is to have a maximal number of newborns tested at the maternity by trained audiologists and to have the Well Baby Organization trace and chase the missing subjects. The model has been evaluated during 1 calendar year (1999) in a maternity with 2,012 newborns.
Result: The result is a coverage of 99.3%. Most newborns (97.3%) were tested at the maternity ward with a total time investment of less than 15 minutes per child. The actual test time is 2 minutes, 12 seconds (median value). The Well Baby Organization keeps track of all the results and has to test no more than 2% of the newborns. Sensitivity and specificity were not the primary outcomes of this evaluation, but they were similar to those of a previous study evaluating the screen procedure on a larger scale, giving a sensitivity of approximately 100% and a false alarm rate of 1/1,000.

Conclusion: These figures demonstrate that universal neonatal hearing screening is feasible within the existing health care structure, with unprecedented coverage, sensitivity, and specificity. **Key Words:** Universal neonatal screening—Sensorineural hearing loss—Neonatal abnormalities.

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The implementation of screening for any disease or impairment requires several prerequisites. Screening and early detection must lead to early intervention with substantial benefit for the patient compared with late intervention (1); the equipment for screening must be available on a large scale and must be affordable (2); the screening procedure must guarantee detection of a majority, if not all, of the subjects with the impairment (3); and the procedure must be feasible (4).

For congenital hearing impairment, it has been shown that early detection is possible and that early intervention leads to significantly better outcome in terms of hearing and speech and language development than in the case of late intervention (5–8), that the equipment to do this is available (otoacoustic emissions or ·automated auditory brainstem response) and relatively cheap when compared with other screening programs (4,5,9), and that the only way to detect almost all children with hearing impairments is by a universal neonatal screening (4,5,10–12). The actual issue in most western countries is to find a

feasible procedure that meets the specific national situation with regard to maternity care. This procedure should combine a minimal cost with a maximal screening efficacy. The cost relates to the equipment used and to the procedure (how to reach newborns and perform a single or multiple tests before referring) (3,11,13,14). The efficacy relates to a maximal coverage and good sensitivity and specificity figures (15,16).

In most western countries, a majority of the babies is born in maternity wards, and it needs no explanation that testing them there is more cost efficient than visiting them at home or inviting them to come to a screening center once they are home. In addition, testing in the maternity ward may have the advantage that professional audiologists may do the screening and that their expertise may serve both the efficacy and the counseling of the parents if needed. Disadvantages of testing in the maternity ward are that it is difficult to reach full coverage either because not all babies are born in maternities or because they leave maternity too early, that a substantial number of babies gets lost to follow-up because maternity wards have no experience in tracing subjects once they have left, and that the data are not centralized for quantitative and qualitative control (16,17).

Because of these considerations, the authors have run

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a pilot project together with the Flemish Well Baby Organization (*Kind en Gezin*) to try to combine the expertise of both a well-functioning centralized controlling organization and a large maternity department (approximately 2,000 births per year) in an hospital that hosts an audiologic department with professional audiologists. The involvement of these two parties is expressed by the term *bipodal*. The term *two-stage screening* is used because the screening consists of an initial test at the maternity ward and a second test 3 weeks later in case of failure. Only a failure on the second test is called a *screen fail* and leads to referral to a specialized audiologic center for further diagnostic workup (16). The aim of this project was primarily to evaluate the feasibility of this type of cooperative screening, the coverage, the number of subjects that get lost to follow-up, and the time investment of the audiologists. The project also included a comparison of two screening devices, both based on the registration of transiently evoked otoacoustic emissions, with one (Echocheck; Otodynamics, Ltd., Hatfield, U.K.) being more user-friendly and more portable (palmtop) than the other (Echoport; Otodynamics, Ltd.), which is a more complicated laptop version.

MATERIAL AND METHODS

During the calendar year 1999, an attempt was made to screen all the newborns in St-Augustinus Hospital of Antwerp, Belgium, both at the maternity ward and the neonatal intensive care unit (NICU).

The technique, equipment, procedure, and decision criteria have already been described elsewhere (16). Briefly, non-linear click-evoked transient otoacoustic emissions were recorded with either the Echoport or the Echocheck devices. Both devices were alternated on a weekly basis. The Echocheck is a fully automatic device giving a pass or a fail based on a fixed algorithm. The Echoport yields a large number of numerical and visual data on the basis of which the examiner has to score the tested ear. Our criteria have been described and evaluated before (16–18) and can be summarized as a signal to noise ratio of 6 dB in at least 3 neighboring frequency bands drawn from the upper 4 bands (1.6, 2.4, 3.2, and 4 kHz) and an overall reproducibility exceeding 50%. An audiologist or a supervised audiologist in training did all tests. The subjects were screened as late as possible, which was on the last working day before the child was supposed to leave the hospital, which was typically at postnatal day 3–5. A fail was defined as a bilateral fail. In such case a retest was scheduled 3 weeks later. If the child failed this test as well, an auditory brainstem response (ABR) with air and bone conduction was scheduled at the age of 3 months. In case of proven hearing loss, the child was referred for hearing aid fitting with the aim to have the hearing aids operational by the age of 6 months.

To assess the total time involved in this screening, 3 time registrations were done at the maternity ward: 1) the total time that the audiologist spent daily for the screening, which includes the collecting of the list of newborns, putting these data in a database, performing all the tests at the maternity wards or NICU, informing and counseling the parents, distributing preprinted reports for the pediatrician or family doctor, putting all the results in the database, and establishing the weekly contacts with the Well Baby Organization; 2) the room time for each

child, which is the time that the audiologist stayed in the room of each newborn; and 3) the test time, which is the time for testing both ears. A Mann-Whitney test is used to compare test times between the Echoport and the Echocheck, and a chi-square test with Yates correction is used to compare pass rates between the 2 devices.

The Flemish Well Baby Organization (*Kind en Gezin*) coordinated and sponsored the project (approximately $11.08 or 12.4 euros/screen). All results were reported to this organization on a weekly basis. For this, two lists were faxed, one with the names of the children who passed the test and one with the names and coordinates of the children whom we missed and should be "chased" by the Well Baby Organization. The latter situation could arise for different reasons: The child was missed for the first test because he or she left the hospital earlier than planned or because of administrative problems, the parents of the child may have refused the screening, or the child failed the first test and was not brought to the second test scheduled for 3 weeks later. These children were actively traced and chased by the Well Baby Organization, and they were tested by means of an automated ABR (ALGO; Natus Medical, Inc., San Carlos, CA, U.S.A.) at the Well Baby centers or at home if necessary. The Flemish Well Baby Organization provided us with feedback on all the children whom they had to chase to complete our database and to allow full analysis.

RESULTS

During calendar year 1999, a total of 2,012 children were born in the St. Augustinus Hospital, 1,781 in the maternity ward and 231 in the NICU. Approximately 60% of infants born in the NICU were high care, and approximately 40% were low and medium care. Four children born in the NICU died while still in the hospital. They were not tested and were excluded from the analysis. Coverage data are shown in Table 1.

Of the 1,954 newborns tested in the hospital, 49.8% were tested by means of the Echoport and 50.2% by means of the Echocheck. The test results of the first screen are shown in Table 2. All 41 children tested by the Well Baby Organization passed the test (40 bilaterally and 1 child from the NICU unilaterally). Pass rates did not differ between the Echoport and the Echocheck (the bilateral pass rates were 95.4 and 94.4%, respectively; $p > 0.05$).

From Table 2 it can be inferred that 28 children needed a retest. Twelve (43%) were brought for this retest at St. Augustinus Hospital; the others (57%) had to

TABLE 1. *Coverage of the neonatal hearing screening**

	Total	Maternity ward	NICU
Number of newborns	2008	1781	227
Tested in the hospital	97.3%	1742 (97.8%)	212 (93.4%)
Tested by Well Baby Organization	2%	30 (1.7%)	11 (4.8%)
Not tested	0.7%	9 (0.5%)	4 (1.8%)
Refused the test		4	2
Lost to follow-up		5	2

*Numbers and percentages of newborns tested in either the maternity ward or the neonatal intensive care unit (NICU).

TABLE 2. *Pass and fail rate of the first test**

	Total	Maternity ward	NICU
Number tested	1995	1772	223
Pass	98.6%	1745 (98.5%)	222 (99.6%)
Bilateral		1679	213
Unilateral		66	9
Fail	1.4%	27 (1.5%)	1 (0.4%)

*Numbers and percentages of newborns that passed or failed the first test. NICU, neonatal intensive care unit.

be chased by the Well Baby Organization. All were found and retested. In case of failure, an ABR was performed at the age of 3 months and approximate audiometric hearing levels were deduced from the ABR thresholds. The results are summarized in Table 3. Figure 1 shows the extrapolated results for the two-stage bipodal screening as calculated for 1,000 newborns. The results of the time registration at the maternity ward are shown in Figure 2 and Table 4.

The median times show that with the Echocheck the test takes 2 minutes, 12 seconds, and that the audiologist stays 5 minutes, 41 seconds, in the room with the newborn. With the Echoport, this takes more than 1 minute extra. This difference is statistically significant (p < 0.001).

The average total time spent per child, including the administrative work, is 17 minutes 36 seconds. After finishing this study, total time registration has been continued for 150 newborns to eliminate the additional time spent for study-specific tasks, such as timing the different steps. For these 150 newborns, the average total time spent per child is 14 minutes, 38 seconds.

DISCUSSION

The development of a device to measure otoacoustic emissions (ILO; Otodynamics, Ltd., Hatfield, U.K.) some 20 years ago triggered a new wave of interest in universal neonatal hearing screening. Since then, different types of equipment have been developed that are

TABLE 3. *Pass and fail rate of the retest**

	Total	Maternity ward	NICU
Number retested	28	27	1
Pass	79%	21 (78%)	1 (100%)
bilateral		21	0
unilateral		0	1
Fail	21%	6 (22%)	0 (0%)
Mild (<40 dB)		2	
Moderate (40–60 dB)		2	
Moderately severe to profound (>60 dB)		2	

*Numbers and percentages of newborns that passed or failed the retest that was typically performed 3 weeks after failing the first test. NICU, neonatal intensive care unit.

essentially based on the principle of either otoacoustic emissions (19–23) or automated ABR (24,25). This has made it possible to easily test the hearing of newborns. It had been speculated before that early detection and early intervention would substantially improve the fate of the child with congenital hearing loss. Soon after the introduction of hearing screening and early intervention programs, it could be demonstrated that the communicative skills and speech-language development of children with hearing impairments indeed changed dramatically (5,23–25). It was equally clear that only universal screening programs would be able to detect all or most of the children with congenital hearing impairments. Attempts to limit the screening to a targeted subpopulation by the use of checklists of indicators of hearing impairment failed because only about half of the hearing-impaired children seemed to have one of these indicators. The Rhode Island project showed that it was feasible to establish such a universal neonatal hearing screening program with the use of the ILO otoacoustic emission analyzer (4,5). A coverage of 95% with a high sensitivity (almost 100%) and a low false alarm rate (less than 5%) could be obtained. These figures proved for the first time that it is possible to organize a hearing screening program for very young children with good screening parameters. This prompted several international authorities to strongly advocate the implementation of universal neonatal hearing screening (5,26,27).

Other countries since then have considered implementing some type of neonatal hearing screening, and they are facing problems to fit this in existing structures like the well baby organizations and maternity wards. A universal screening program seems to be the natural responsibility of a nationwide well baby organization. Such organizations have expertise in keeping large databases, in tracing newborns, and in controlling screening programs. However, they lack expertise in hearing evaluation and, in consequence, in counseling parents properly, especially in cases of hearing problems. In addition, they experience problems in establishing sufficient coverage (28,29). In contrast, maternity wards seem to be the natural place to look for newborns, and even if not all of them are born in a maternity ward (in Belgium, more than 99% are born in maternity wards), testing those who are at the maternity ward should save a lot of money and effort. It seems reasonable to expect a high coverage.

The pilot study that has been reported on in this paper has evaluated a bipodal screening program. The different steps of the screening procedure with the decision criteria to define pass and fail were established and optimized over the years and were published separately (16,18). The cooperation between the two parties starts with the audiologists trying to test as many newborns as possible at the maternity ward. They give full weekly reports to the Well Baby Organization, including the data of the babies that passed the test and of those that were not tested for any reason. The Well Baby Organization thus keeps track of all newborns having passed the test and of those that still have to be tested by their own structure. In
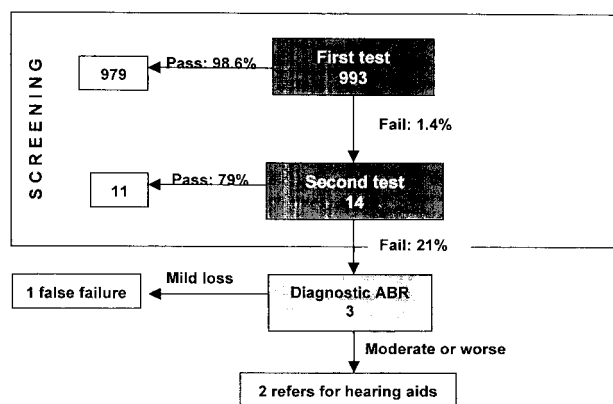
FIG. 1. Flowchart and results of a typical screen calculated for 1,000 newborns. Because 7 are missed (see text), only 993 undergo the first test. When the screening is defined as the combination of the first test and the retest, the screen pass rate is 99.7%. *ABR*, auditory brainstem response.

TABLE 4. *Time registration for the first test in the maternity ward**

	Echoport		Echocheck	
	Room time	Test time	Room time	Test time
Upper extreme	850	540	676	418
Upper quartile	559	306	437	224
Median	439	208	341	132
Lower quartile	365	150	278	94
Lower extreme	197	63	132	33

*Time (seconds) spent in the maternity room (room time) or just for testing (test time) with the Echoport® and the Echocheck³. For both systems approximately 7%–9% outliers exist beyond the upper extremes.

case of fail at first test, a retest is immediately scheduled 3 weeks later. If the parents fail to show up for this retest, the data of the child are immediately added to the weekly report so that the Well Baby Organization knows that this child should be actively chased.

The results show that a high coverage of 99.3% is obtained by this bipodal system. Half of the remaining 0.7% are missed because the parents refuse screening. Such coverage has never been reported before for a universal screening program (17). In addition, 97.3% of the newborns could be tested at the maternity ward, taking no more than 15 minutes per child when the Echocheck was used, which has become the standard in our department. So, the Well Baby Organization can focus all of its energy on the remaining 2.7% of neonates, resulting in an additional 2% coverage. In this particular setting, the intervention of the Well Baby Organization was required
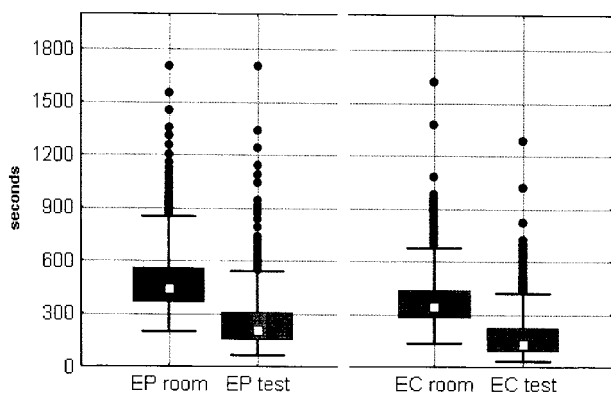
to complete a minimal fraction of the first stage (41 newborns, or 2% of the cohort) and a larger fraction of the second stage (16 babies, or 57% of the ones who failed the first test). Of the children that failed the first test, no one was lost to follow-up. Although this study was too small to evaluate test sensitivity and specificity, a hit rate of more than 2 hearing-impaired newborns per 1,000 and a false alarm rate of less than 1 per 1,000 are in line with a previous study on a larger population. These figures seem also exceptionally good and the authors believe that this is at least partially because of different procedural factors such as the moment of testing (3–5 days after birth), the decision criteria (unilateral fail = pass), and the fact that trained audiologists perform the test. Trained audiologists are not more expensive than nurses in Belgium. The authors feel that their expertise in handling hearing-impaired people adds to the quality of the screening and certainly to the quality of counseling the parents in case of a fail. This results in an important reduction of the parental anxiety in comparison to counseling by others.

It is obvious that a high-quality screening program alone is not sufficient but should rather be followed by a well-structured and widely available diagnostic follow-up and an early intervention program. In Belgium, a limited number of diagnostic centers are recognized by the Well Baby Organization for the diagnostic workup after referral. This diagnostic workup includes full audiologic assessment with bone and air conduction ABR, connexin-26 analysis, ophthalmologic examination, electrocardiogram, and medical imaging. By the time the child has reached the age of 3 months, the diagnostic assessment is to be completed, and the child is referred to a specialized center for hearing aid fitting and early educational and developmental intervention. By the age of 10–12 months, the auditory performance with hearing aids is assessed by means of audiometry and phoneme discrimination tests. On the basis of these results, it is decided whether the child continues with hearing aids or is referred for cochlear implantation. Thanks to this tight scheme, the age of implantation has shifted from 2–3 years to below 2 years.

In conclusion, the present bipodal model is the result of multiple modifications to existing techniques and of

FIG. 2. Time spent in the room and time spent for the actual testing for the Echoport (*EP room, EP test*) and the Echocheck (*EC room, EC test*). In the box- and whiskerplots, the central dot represents the median time, the box represents the upper and lower quartiles (P25 and P75), and the whiskers represent the lower and upper extremes. The outliers are depicted as individual dots above each plot. The exact values can be read in *Table 4*.

an attempt to combine the specific competencies of two involved parties. It is applicable in many situations and can fit in most local situations. It may result in universal screening with better coverage, sensitivity, and specificity than those reported in other studies (4,13,15,30–34).

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