Overcoming difficulties in implementing a universal newborn hearing screening program

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The aim of this paper was to present our experience with a universal newborn hearing screening program, including the specific problems and difficulties faced since its beginning, along with the measures used to overcome them and to improve its efficiency. We analyzed data from 22,195 newborns screened by transiently evoked otoacoustic emissions (TEOAE) performed during the first days after birth. 84.8% of the newborns passed on the first test and another 12.15% passed on rescreening before hospital discharge. This produced a “not pass” rate (false-positive and true positive) of 3.05%. The rate of newborns who did not undergo screening and the rate of “lost to follow-up” newborns were reduced in time, due to various applied modifications to the protocol. It may be thus concluded that our protocol proved to be successful in attaining low refer rates for follow-up screening. A major problem that remains unresolved is the absence of effective follow-up.

Key words: hearing, otoacoustic emissions, screening, transiently evoked otoacoustic emissions, universal newborn hearing screening.
on Infant Hearing, which developed the Year 2000 Position Statement\textsuperscript{16}: Principles and Guidelines for Early Hearing Detection and Intervention Programs. Year after year valuable data are accumulating regarding the experience gained throughout the developed countries from these programs, and the vast majority support the idea that the universal neonatal hearing screening program is feasible, cost-effective and positively impacts children’s lives\textsuperscript{17-20}.

Greece was among the first of the European countries to adopt infant hearing screening. Our program, based in a private maternity hospital of Athens, was the first such program in our country, started six years ago at first on a pilot basis and then universally. Until then, newborn hearing testing was available only in a public pediatric hospital in high-risk newborns\textsuperscript{21}. The purpose of this paper was to record in details our experience from the hearing screening program, including the specific problems and difficulties faced since its beginning, along with the measures and means used to overcome them and to improve its efficiency.

**Material and Methods**

**Setting:** Iaso Maternity Hospital is a 286-bed major private unit for obstetrics, gynecology and neonatology, with a full range of perinatal services, providing tertiary care to mothers and infants from Athens and from many other regions of our country. Iaso has functioned since 1995, in a modern, fully equipped eight-floor building, with experienced medical and nursery staff. The Neonatal Intensive Care Unit (NICU) is a 60-bed, fully equipped facility. The institution has an annual delivery rate exceeding 10,000 births, and is providing nursing care to more than 500 females.

**Screening Staff:** A pediatric audiology unit has functioned in Iaso Hospital since 1996, operated by a senior neuro-otologist, an otolaryngologist and a trained nurse. The medical staff was experienced in neonatal hearing screening techniques, whereas the nurse had an educational course, in order to learn the techniques of TEOAE. Screening was performed and supervised by the doctors. Coordinating the daily operation of the screening tests was achieved in cooperation with the nurses, secretaries and head nurses of each separate floor. The medical staff was responsible for the scientific coordination of the program, interpreting the screening results, modifying screening conditions when necessary, keeping and processing screening data and contacting parents and doctors. Tasks, such as offering information sheets to parents, bringing infants from the nurseries to the screening location and arranging administrative issues were performed by the nurses and secretaries.

**Preliminary Measures:** Informing the medical and paramedical staff of the hospital about the necessity of implementing a newborn hearing detection screening program was a prerequisite for the initiation of the program. For that purpose, both lectures and personal communication were used with the obstetricians, the pediatricians and the nursing staff. These efforts aimed towards acquainting all persons involved with newborns with the method of testing, the purpose and the necessity of the screening program, the equipment used, the procedure followed and the possible outcomes. Moreover, cooperation with the nursing staff had to be achieved, in order to provide the optimal conditions for the screening and consequently the optimal results.

**Procedure:** The program was based on a two-stage procedure. The first stage was a pilot study of newborns from the NICU on the basis of high-risk criteria and of selected healthy newborns when requested by the parents or the obstetrician. It was initiated in October 1996 and lasted until December 1999. During this period, all parents were informed about the possibility of hearing screening via a leaflet containing basic information regarding screening and an explanation of the testing procedure, its necessity and the expected benefits. After testing, in cases of failure, the parents were informed the results directly by the medical staff.

The second stage was a universal newborn hearing screening program. It was initiated in December 1999 and at first covered all newborns of two floors, gradually expanding to include all births in the maternity hospital, and to the present is active on a universal basis. The same procedure was followed, with testing being performed in each separate floor. All newborns were screened with TEOAE, in a quiet room adjacent to the nursery of each floor, away from crying infants, keeping ambient noise as low as possible.
There was an effort to perform as many tests as possible after feeding, so that the neonates would be quiet or asleep. Babies were tested in their cribs and the ear more readily accessible to the tester was screened first. As newborn hearing screening was now more widely known, parents were informed by the pediatricians about testing results and were referred to the Audiology Department only in cases of failure. Leaflets, along with personal communication, were used to inform the parents about the hearing screening program.

**Equipment and Methods:** Testing was performed using the ILO88 Otodynamics analyzer (Otodynamics, London, software version 3.94H) connected to a portable personal computer. The Quickscreen program was employed, with an analysis window 12.5 ms poststimulus. The recording bandwidth was set between 0.75 to 5 kHz and stimulus intensity was approximately 80 dB SPL, nonlinear stimulation mode. Repetition rate was 50 stimuli/sec. We used the standard ILO neonatal probe, with disposable tips. Meatus response monitoring was used to check fitting conditions of the probe and testing began only when a good fit was achieved. The noise rejection level at the probe tip was set to 47 dB. The stability of the stimulus and therefore of the probe fit was observed during the measurement, as a colored light on the screen. A red light indicated the probe should be refitted and the measurement restarted. Depending on the status of the newborns during the test, noise rejection levels might be modified by the examiner. The numbers of responses accepted and rejected by artefact rejection were displayed and updated during averaging. Fifty samples or less were collected during the testing process, if “pass-fail” criteria were met. However, up to 260 or more samples were collected if necessary. In the beginning of our program we used the same pass-fail criteria as were used in the Rhode Island Hearing Assessment Project. A signal-to-noise ratio ≥ 3 dB, across the test frequency bands of 1-2, 2-3 and 3-4 kHz, was considered necessary for a “pass”. Additionally, an overall reproducibility of at least 60% was considered necessary for the final pass.

**Protocol:** A major goal of our program was to minimize the false-positive results as much as possible, without reducing the sensitivity of the test. According to our protocol, the newborns were routinely tested on the third day of life. Although in the majority of babies this was accomplished, we also tested newborns on their second day of life and in some cases on the fourth day (restless babies that had to be retested). Newborns that passed the initial screening bilaterally were discharged from the program. If the test could not be performed, it was repeated later during the same day. If they passed the test they were then discharged. In case of a new failure (either unilateral or bilateral), the procedure was repeated again before discharge from the hospital, which occurred on the fourth day under normal circumstances. In cases where the screening results remained abnormal until the last pre-discharge screening, the newborns were referred for retesting with TEOAE, three to four weeks later. The appointment was arranged following contact with the parents and counseling, in order to explain the meaning of a failed screen. Finally, all babies failing the retest were referred for complete audiological evaluation including acoustic immittance measurements and ABR, after drug-induced sleep. This testing was performed either in Iaso Hospital or in the Audiology Department of a public pediatric hospital with which we cooperated.

**Modifications of the Program:** The routine daily program had to be altered in several instances, so as to incorporate the appropriate timing for the testing of each newborn. Babies were never interrupted from feeding and there was an effort either to test first and delay feeding by delivering babies to their mothers later, or to shorten the duration of the stay in the mother’s room and proceed to testing. Also, newborns were never tested when crying, or when making excessive noise. In that case, testing was postponed until lately that day or the next day.

Another major change was made in our protocol, since all those newborns that failed the initial testing were immediately retested (second screen). Before this, otoscopy and cleaning of the external auditory canal (when necessary) were performed, as well as replacement of the probe tip. Further, during the implementation of the program the initially used “pass-fail” criteria were modified, according to gained experience, data from medical literature, and results from the programs conducted in other countries. More strict criteria were adopted in order to
improve the sensitivity of the program and to minimize the risk of missing hearing impaired newborns. At present, we consider as “pass” a signal-to-noise ratio ≥ 6 dB, across the test frequency bands of 1-2, 2-3 and 3-4 kHz, with an overall reproducibility of at least 70%.

The program initially only functioned during work days. Newborns scheduled to be discharged during the weekend or on Monday were examined on the second or the fourth day, resulting in reduced efficiency of the program. With testing on the second day, there was greater chance of abnormal results. With testing on the fourth day, although there were less false-positive results, repeat testing in cases of abnormal findings was not possible the next day because the newborn was discharged. The problem was finally resolved by implementing seven-day screening, after engaging additional staff. Initially, the staff conducting the program, aside from the supervising senior neuro-otologist, consisted of one doctor and one nurse. One more doctor and two nurses were gradually added, in order to conduct screening on a daily and universal basis. Of the medical staff, one doctor supervised the nurses during test performance; the second doctor was responsible for the follow-up of babies that failed the initial screening, and also replaced the other doctor in case of holidays or in other necessary instances. The senior neuro-otologist was responsible for the scientific coordination and modification of the program, interpreting the screening results and contacting parents and doctors, if necessary. The new nurses were appropriately educated by the doctors in performing the screening test and were informed in detail about the specific issues involved in conducting a universal hearing screening program in newborns.

The increasing number of newborns examined daily, the extension of the program to include weekends, and the testing of newborns separately in each floor, each an independent functional unit, thus required continuous transfer of the portable equipment and the testing staff and rendered additional equipment necessary in order to perform the tests efficiently. One more portable TEOAE device was supplied, which was used for follow-up and as a reserve in case of failure of the device used on the floors. From our experience, we estimate that at least one piece of equipment per 500 births/month is needed to operate the screening program efficiently.

Another issue that we had to confront was how to minimize the number of newborns who failed to enter the screening program. This was due to errors of scheduling and poor cooperation between the program’s nurses and the nurses and secretaries of the hospital. Negative attitude on behalf of several parents and from the medical staff of the hospital also contributed to these misses. However, after the universal application of the program, the scientific community and parents became better acquainted with the program’s procedures; information was given in every case with lectures, booklets and on an individual basis; and better scheduling and standardization of the program was achieved. Another problem was the initially high rate of newborns that failed to follow the re-test process. This was addressed with better follow-up scheduling and with written and oral information provided to the parents.

Results

Cumulative results from all the newborns tested so far have not yet been processed. However, a significant sample of newborns tested in a three-year period is herein presented, when hearing screening was universally applied after the initial pilot stage of the program, in order to show preliminary results and to extract useful conclusions about the effects of the applied modifications.

In this period, 22,195 newborns were tested (Table I); 84.8% of these babies (18,820) were found normal, passing the initial test, whereas 15.2% (3,375 babies) failed the test. Finally, 2,696 (12.15%) of these babies were discharged from the program after passing the repeat test during their hospital stay, whereas 679 babies (3.05%) were asked to return one month later for re-testing, as they failed all tests performed before discharge from the maternity hospital. This resulted in a high “pass” rate of 96.95%, a value that approaches the specificity rate, since it is known that the prevalence of congenital hearing loss is quite small\textsuperscript{10}, and that most of the newborns with positive results are probably false-positive.
Table I. Preliminary Results in a Large Sample of Newborns Tested Over Three Years

<table>
<thead>
<tr>
<th></th>
<th>Total</th>
<th>Normal</th>
<th>Abnormal</th>
</tr>
</thead>
<tbody>
<tr>
<td>First examination</td>
<td>22195</td>
<td>18820</td>
<td>3375</td>
</tr>
<tr>
<td>Predischarge examination</td>
<td>3375</td>
<td>2696</td>
<td>679</td>
</tr>
<tr>
<td>Follow-up</td>
<td>189 (0.85%)</td>
<td>144 (0.65%)</td>
<td>22 (0.12%)</td>
</tr>
</tbody>
</table>

and not truly hearing impaired. It is not yet possible to estimate the sensitivity due to the incomplete follow-up of all the newborns.

Of the 679 newborns whose parents were requested to return with the infants for follow-up, 189 children presented and 490 did not. One hundred and forty-four of them had normal emissions, whereas 45 babies continued to have abnormal TEOAE, 22 unilaterally and 23 bilaterally. These babies were referred for further complete clinical and audiological evaluation with ABR and acoustic immittance measurements and although final results are not yet available for all of them, to date eight babies have been proven to suffer from a congenital hearing loss.

Considering the results of the examined sample in total, it may be observed that abnormal results after the initial test decreased significantly over time. However, the observed decrease in the final abnormal results, when the newborns were discharged from the hospital, was not as impressive (Fig. 1). It may thus be concluded that although the “pass” rate of the test (and probably the specificity) had minor improvement (Fig. 1). It may thus be concluded that although the “pass” rate of the test (and probably the specificity) had minor improvement during this period, which alone is important, the main result was that fewer retests were needed. Therefore, a significant saving of human and financial resources was obtained.

A similar effect may be observed in the miss rate and “lost to follow-up” rate of newborns during this period. The miss rate, which was quite high during the initial stage of the program, was due to various reasons, such as parental refusal to accept testing of their babies, negative attitude from several obstetricians, errors in scheduling and deficient cooperation between nurses and secretaries, unavailability of seven-day testing, or even referral to public pediatric hospitals. After coping with all these obstacles, a significant decrease was observed in the miss rate (Fig. 2). The same trend occurred in the rate of “lost to follow-up” newborns, which was also initially significant but improved over the three years due to the several measures taken (Fig. 3). Nevertheless, to date this rate remains too high.

Although several improvements were applied continuously during this stage of the screening program, four major modifications of the protocol were studied separately, because it was thought that they would contribute significantly to a better efficacy of the program. These included: (1) Appropriate time of testing for each newborn and postponement of testing in case of crying or excessive noise; (2) Immediate rescreen, in case of a ‘fail’ result; (3) Seven-day screening with additional staff and equipment;
Fig. 2. Miss rate in a sample of 22,195 newborns, screened during a period of three years and divided in six-month intervals.

Fig. 3. Percentage of “lost to follow-up” newborns in the sample of 22,195 newborns, divided in six-month intervals.

and (4) Adoption of a more strict ‘pass’ criteria. Figure 4 illustrates the various points in time that these modifications were applied. The effects of these modifications on the ‘fail’ rate, the ‘miss’ rate and the ‘lost to follow-up’ rate are shown in Table II. In this Table, comparisons between these rates were performed between the newborns screened one month prior to and one month after the modification, in order to avoid other possible conflicting factors, in case samples of longer periods would be compared. It appears from these comparisons that the last modification did not result in any improvement in the above rates. However, the remaining three major modifications improved the ‘fail’ rate of the program. The seven-day screening also improved the ‘miss’ rate, whereas the ‘lost to follow-up’ rate was not improved via any of the applied modifications, but its decrease was continuous and may have been due to the various measures taken for informing the parents and to better scheduling of the follow-up process.

Discussion
The development and application in clinical practice of automated ABR and of otoacoustic emissions marked the beginning of a new era in the field of screening for congenital hearing loss. These new techniques allowed for an objective evaluation of hearing almost as soon as the babies were born, filling the two-year gap needed from the traditional behavioral methods.
of evaluating hearing to establish a diagnosis and intervention\textsuperscript{5}. In particular, TEOAE seemed to provide an optimal solution to this problem, as testing was simple, easy to perform (even by technicians and paramedical staff), non-expensive and faster than ABR\textsuperscript{26}. Several investigators\textsuperscript{27} have questioned TEOAE’s validity in hearing screening programs, as there have been many protocols, with different pass-fail criteria, influencing the sensitivity and specificity of this method. Additionally, TEOAE does not provide information for the auditory pathway beyond the organ of Corti, allowing for some false-negative test results. Nevertheless, it appears that testing with TEOAE is at the moment a valuable tool available for the early detection of hearing loss\textsuperscript{28} and many universal hearing screening programs in practice apply them as the method of choice, followed by ABR in cases of failure.

For these reasons, we have based our newborn hearing screening program in Iaso Hospital on TEOAE. This institution was an ideal setting for the application of such a program, as it presents a very high birth rate. As soon as the administration of the hospital was informed by the Department of Audiology about the feasibility and usefulness of a hearing screening program, they were very positive and have made efforts to assist in its implementation in the best possible way. However, various problems emerged during the application of the program, which had to be addressed in order to obtain optimal efficiency. In the pilot stage, testing was performed only in babies from the NICU and on a voluntary basis in healthy babies. The numbers of newborns tested in this preliminary stage was rather low, as compared to the number of newborns tested during the universal second stage, but it proved of major importance in defining and dealing with the everyday problems that had to be resolved, since the same problems would be met during the universal screening that would follow, but on a larger scale.

Since November 1999 when the program was applied on a universal basis, the number of babies tested has exceeded 1,000 per month. A major problem we faced was to minimize the number of newborns that failed to enter the screening program due to various reasons, including negative attitude on the part of some of the medical staff of the hospital, parental refusal, and poor cooperation between nurses and secretaries. We confronted the latter problem by providing better scheduling and standardization of the program, which resulted in a reduced miss rate, improving the overall performance of the program. Our efforts to induce a positive attitude in the medical staff of the hospital towards hearing screening has met with slower progress, but persistent

| Years | Months | 1\textsuperscript{st} | 2\textsuperscript{nd} | 3\textsupers

<table>
<thead>
<tr>
<th>Time of testing</th>
<th>1-6</th>
<th>7-12</th>
<th>13-18</th>
<th>19-24</th>
<th>25-30</th>
<th>31-36</th>
</tr>
</thead>
<tbody>
<tr>
<td>Testing anytime</td>
<td></td>
<td></td>
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<table>
<thead>
<tr>
<th>Rescreen</th>
<th>No immediate rescreen</th>
<th>Immediate rescreen</th>
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<tr>
<th>Days of screening</th>
<th>5-day screening</th>
<th>7-day screening</th>
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<tr>
<td>3-dB in 3 frequency zones</td>
<td>6-dB in 3 zones</td>
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<tr>
<th>‘Pass-fail’ criteria</th>
<th>3-dB in 3 frequency zones</th>
<th>6-dB in 3 zones</th>
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Fig. 4. Major modifications to the protocol over the years.
and continuous provision of information has gradually led to a change in attitude. As Iaso was one of the pioneers in neonatal hearing screening in our country, little to no information was available to the obstetricians and the pediatricians at that time. Thus, our goal was to not only inform the medical staff about the relatively new technique of TEOAE, but also to provide sufficient, convincing and valid data that would support the necessity of the program’s implementation. Details about the method, the procedure followed, the cost and the possible benefits and implications from the screening had to be discussed with all doctors involved and a communication for the future had to be established. We proceeded with a detailed but easily understandable presentation of the various issues of the program to the doctors, stressing the importance of early detection of hearing loss, the simplicity and non-invasiveness of the test and the positive attitude of physicians worldwide, following a similar procedure as carried out in other programs elsewhere.

Apart from informing the scientific and paramedical staff of the hospital, informing the parents of the newborns was an even more difficult and crucial issue. Information about the existence of the screening program, when and how testing was performed, and about test results and their meaning had to be occasionally provided. We dealt with this issue by preparing leaflets on hearing screening, its importance, and on the value of early diagnosis of hearing loss, which were distributed to all parents. Parents were welcomed to contact the Audiology Department for any additional information and help. That was also the practice in many programs implemented worldwide. However, the overprotective behavior of Greek parents towards their babies presented a severe obstacle to the implementation of the program. The lack of information on the subject through other official sectors caused a certain insecurity regarding the need and usefulness of the screening, which made them reluctant to participate. Our continuous efforts to provide information, advice and recommendations in every case, and moreover, the successful performance of the screening program, resulted in the end almost eliminating the occasional negative parental attitude.

Incorporating the procedure of the screening in the everyday routine of the maternity hospital was another problem encountered. The collaboration of the nurses was crucial in this field, as they had to reschedule their program, mainly with regard to the feeding time of the babies, in order to make testing easier. Although at the beginning of the program there were some difficulties, a good level of cooperation was reached in a short time and at present we face only some minor dysfunction in this field. Another practical issue was the need for additional equipment and staff. However, since no government or other institutional funds were available and the program was self-funded by charging the parents of newborns for the examinations performed, it was necessary to function initially on a limited basis. As soon as the necessary funds to accomplish the needs for conducting universal screening with success were obtained, these problems were immediately resolved.

An important factor in our program was to achieve a high pass rate, thus minimizing the need for re-testing and the false-positive results due to inappropriate test conditions or other environmental factors. Based on the experience from other programs, where low referral rates were obtained after appropriate measures taken, we aimed at reducing this rate substantially. For that purpose we applied a protocol that allowed for as many testing sessions as possible before discharge. While the majority of newborns were tested on the third day, we tried to test some of them even on the second day, keeping in mind the possibility for re-testing. As most mothers and healthy babies stayed at least four days in the hospital, and occasionally five or even more days due to unpredictable reasons, we had the opportunity to retest them two or even three times. While this did increase the work load, we avoided the need for referral in a substantial number of cases, and we believe this was a time-effective approach. Thus, we have obtained quite a low referral rate of approximately 3.05%. We strongly believe that testing repetition both immediately after an initial “fail” result and before discharge is the answer to the problem of high referral rates, since even a minimal displacement of the probe or even change in the status of the newborn may have a crucial effect on the results of the test.

Within the three-year period during which our sample was drawn, 679 newborns failed initial testing and were asked to return for a re-
evaluation. However, 490 (72.1%) did not show up, resulting in a high “lost to follow-up” rate. There were many reasons for this, such as the absence of a schedule to follow these children and contact their families when appointments were not kept; occasional referral to other audiological centers or hospitals; indifference on behalf of the parents due to incomplete information about the goals and importance of the screening program; and negative attitude of a small part of the medical community. Also, it should be mentioned that Iaso Maternity Hospital provides tertiary care to mothers and infants not only from Athens, but from many other regions of the country. Thus, follow-up is difficult in these cases, and either the newborn is completely lost from the program, or is referred and further followed-up in local facilities. Several measures had to be taken to address this problem, such as written documentation in the infant’s records indicating that hearing screening had been performed in the maternity hospital, along with the results. Personal contact with the parents was also undertaken, and they were given explicit information regarding the meaning of a “fail” result and were asked to return for further evaluation one month later. It should be noted that soon after the universal application of the program, negative attitude on behalf of the parents seldom occurred, as the medical community and the parents became more acquainted with the program’s requirements, goals and possible shortcomings.

Finally, two additional issues that caused initial skepticism towards the screening program should be mentioned. The first was the fear that possible hearing deficits revealed by the testing would have an adverse effect on the obstetricians, negatively affecting their relation with the parents. However, after implementing screening on a universal basis, and after the initial successful application of the program, the parents themselves suggested to the occasionally hesitant obstetrician that their babies should participate in the program. The second reason was financial, because in the private setting of the hospital parents were obligated to cover the cost of the screening. This was addressed by reducing the cost of the examination by approximately half the original cost.

A significant problem which remains unresolved is the absence of effective follow-up, diagnosis and intervention in cases with hearing impairment. When the screening program was first implemented, all these procedures were scheduled to be conducted via local facilities. However, although the Audiology Department of the hospital was fully equipped to perform detailed diagnostic procedures, various problems arose, such as ineffective cooperation with anesthetists, as well as referral to other public pediatric hospitals, since a maternity hospital is not generally considered suitable to provide such facilities. Also, a complete diagnostic work-up in a private unit is quite expensive, and many parents were reluctant to pay for this expense. In response to these problems, we referred infants with abnormal results to public pediatric hospitals, but even this was not entirely successful because many cases failed to follow our instructions. At present we are still pursuing a solution to the issue by following the models of other functioning programs31, by establishing a close contact with a specific public pediatric unit with immediate referral for a complete diagnostic procedure and intervention when needed, in an effort to reduce to the extent possible the rate of those “lost to follow-up”.

REFERENCES