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Organisation of a universal newborn hearing screening programme in Flanders

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Abstract. *Organisation of a universal newborn hearing screening programme in Flanders. Objective*: Since 1998 an integrated universal newborn hearing screening programme (UNHSP) based on automated auditory brainstem response (AABR) has been implemented in Flanders. The protocol of the UNHSP is based on guidelines defined by the American Academy of Paediatrics (AAP). The aim of this paper is to report on the screening protocol and to assess its feasibility.
Methodology: Descriptive study based upon an analysis of the screening results in the neonatal non-NICU population of Flanders between 1999 and 2004. The UNHSP, organized by Kind en Gezin (K&G), uses a 2-stage protocol: children with a refer at the first screening test are retested, and those with a refer at the retest are referred to a certified centre. Screening and referral centres communicate their data to a central database at K&G.
Results: From the beginning of 1999 until the end of 2004 a screening was offered to 97.91% of all eligible babies in Flanders; 91.5% of these babies were screened by K&G using the Algo® Portable Newborn Screener. Three-quarters of the referred babies had a confirmed hearing loss. In 57.6% of these babies, hearing loss was bilateral. Some babies had a temporary hearing problem. The false positive rate after two tests was 0.53‰. All ascertained babies started early intervention, most of them before the age of 4 months.
Conclusions: K&G has succeeded in organizing a new, well-structured community-based UNHSP according to the guidelines of the AAP on Neonatal Hearing Screening.

Introduction

Permanent Childhood Hearing Impairment (PCHI) (bilateral and ≥40 dB HL) affects 133 per 100,000 newborns (95% confidence interval 122 to 145).[1] Among these, about 55% have a moderate hearing loss (40-70 dB HL) whereas the remainder have severe (70-94 dB HL) or profound (≥95 dB HL) hearing impairment with an equal distribution between both groups. In Flanders, birth rate averages 60,000 per year and annually 73-87 babies are expected to suffer from PCHI that may interfere with speech and language development. Neonatal Intensive Care Unit (NICU) babies are known to run a 10 to 15 times higher risk for hearing disorders.[2] The incidence of bilateral congenital hearing loss is many times greater than the combined incidence of all commonly screened newborn disorders.[3]

Newborns with hearing loss are deprived of the sensory input that is essential for stimulation of the auditory cortex and speech development. Moreover, hearing loss has pernicious effects on the development of social, emotional, comprehension and motor aspects of personality and on the process of education and parent-child interaction.[4] It has been abundantly documented that early remediation of hearing loss in newborns results in improved development of receptive and expressive language skills.[5] Therefore, early detection of hearing loss is of utmost importance. The Joint Committee on Infant Hearing (JCIH) and the American Academy of Paediatrics (AAP) endorse universal newborn hearing screening (UNHS), evaluation, and family-centred intervention of congenital and childhood hearing loss.[6] The primary objectives, recommended screening parameters, and guidelines for tracking and follow-up of hearing impaired children have been published.[7] Every child with PCHI should be identified before the age of 3 months so that rehabilitation can be started before the age of 6 months. European authorities confirmed this recommendation in the European Position Statement 2000.[8]

In Flanders, a newborn hearing screening programme based on the Ewing distraction test at the age of 9 months was offered by K&G (a governmental institution for child and family health) since 1976. Experience, however, has

Table 1
Guidelines for the requirements of a UNHSP

A minimum of 95% of newborns must be screened.
The methodology should detect, at a minimum, all children with a hearing loss ≥35dB in the better ear.
The false positive rate should be ≤3% and the referral rate for formal audiological testing should not exceed 4%.
The methodology should (ideally) have a false negative rate of zero.
Currently accepted methods for physiologic screening include evoked otoacoustic emissions (EOAE) and auditory brainstem response (ABR), either alone or in combination.
The programme should provide appropriate training and monitoring of performance of the staff responsible for the hearing screening.
The system should guarantee confidentiality and informed consent from the parents must be obtained before screening.
Guidelines must be developed for the responsibility of documenting the screening results and communication of test results with the parents and the child's physician(s).
Collaboration between local, state and nation wide monitoring systems to identify false negatives.
Secure funding for the programme.
Collect critical performance data to ensure that each UNHS programme meets the criteria specified in this statement.

shown that hearing screening with the Ewing test was unreliable. Therefore, K&G, reviewed its hearing screening strategy, and implemented since 1998 a new, well structured UNHSP according to the standards of the AAP and the European Consensus Statement on Neonatal Hearing Screening. The aim of this paper is to describe this UNHSP organized in Flanders and to assess whether it meets the screening guidelines formulated by the AAP (see Table 1).

Materials and methods

This paper presents a descriptive study based on a retrospective analysis of screening results collected by K&G between January 1999 and December 2004. Coverage during the implementation period of the UNHSP in 1998 is not included in the analysis.

In the first section, the organisation of the UNHSP in Flanders will be presented followed by a discussion of its performance.

Screening device

The UNHSP in Flanders uses an automated auditory brainstem response (AABR) device: the Algo® Portable Neonatal Hearing Screener (Natus Medical Inc., San Carlos, Ca). The Algo® Portable delivers 100 μs clicks (37.3 clicks/s), with an intensity of 35 dB HL and a frequency content ranging from 700 to 5000 Hz, and records auditory brainstem evoked responses. The Algo® Portable has an internally stored detection template that is based upon the characteristics of evoked waveforms at supra-threshold level of infants with normal hearing. The comparison of the recorded waveforms and an internally stored detection template results in the categorized response of the device: either "pass" or "refer". A "pass" result means that the baby has a 99.98% chance on normal hearing. A "refer" result means that the instrument is not capable of achieving this certainty. Moreover, the Algo® Portable has a dual artefact rejection system that automatically rejects environmental and myogenic noise. The Algo® is designed for screening of babies starting at a gestational age of 34 weeks up to 3 months of age, the upper limit not exceeding 6 months. The test can be performed by only one person who does not need an audiological training. The apparatus is portable, works on batteries, and does not require a special screening environment (e.g. sound proof or electrically shielded room). It can even be performed with the baby lying in the mother's arms. Three disposable surface electrodes are placed: one on the vertex, one on the nape, and one on the top of the shoulder or the cheek. Disposable ear couplers are connected to the transducers and sealed over the infant's ears (see Figure 1).

Results

Organisation of UNHSP in Flanders

K&G is a governmental institution accessible to people living in Flanders (i.e. the Dutch speaking northern part of Belgium) and Brussels. Screening for hearing loss in newborns is one of K&G's tasks as laid down in a decree (Act of the Flemish Community May 29, 1984). The coverage of K&G is very high: almost 88% of young mothers and newborns are seen by a K&G-nurse in the maternity ward, 97% are visited at home, and about 86% visit an infant welfare clinic for further preventive follow-up.

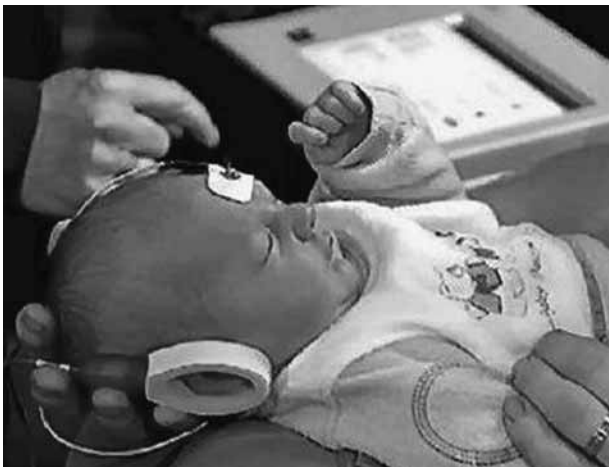

Figure 1
Algo hearing screening in a 4-weeks old baby

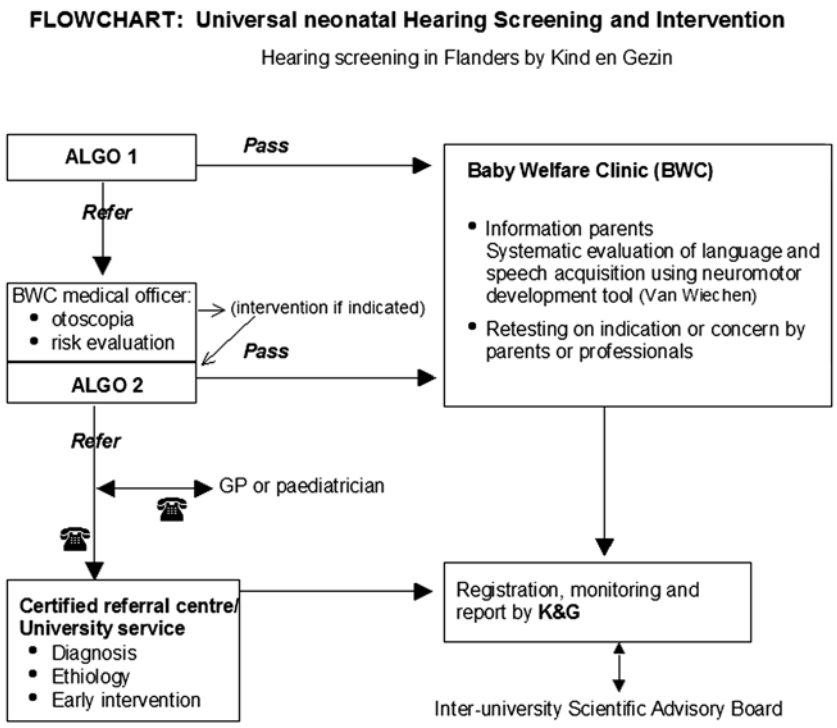

Figure 2
Algo procedure for UNHS in Flanders

During the visit at the maternity ward, every new mother is informed about hearing screening: extensive information and a folder about the Algo® test are given. At about the age of four weeks the district nurse carries out the hearing screening. K&G keeps records of all tested infants and of the child's evolution at each visit to ensure a long-term follow-up and to avoid dropouts. The UNHSP has a 2-stage screening protocol (Figure 2).

Babies with a "pass" result for both ears are assumed to have normal hearing. They are further monitored at visits in the welfare baby clinic with regard to their speech and language development, in order to detect secondary hearing impairment, "late onset" hearing impairment, or progressive hearing loss.

If the first test result is a "refer", a second test is carried out within 48 hours in the presence of the medical officer of the baby welfare clinic. The latter examines the ears and draws up a specific case history based on the risk list of the Joint Committee on Infant Hearing Screening.[4] If the second test is again a "refer", the family doctor or paediatrician is immediately contacted, and in mutual consultation, the baby is immediately referred to a certified specialized referral centre or university service.

This UNHSP protocol implies strict collaboration of different players in the health care system, and has been approved by the Flemish health minister. It guarantees optimal follow-up without losing referred children.

The 23 specialized referral centres for early monitoring, diagnosis, and integral rehabilitation, and the university ENT-departments in Flanders are supposed to report their evaluation within two weeks after referral. In case of confirmed hearing loss, additional tests for a comprehensive diagnosis are carried out during a second phase. A detailed description of the audiometric and etiological work-up was recently published by Declau *et al.*[9] At the same time, a multidisciplinary approach begins immediately, with integral rehabilitation and home support (audiology, speech therapy, physiotherapy, teaching and psychological support, and contact groups).

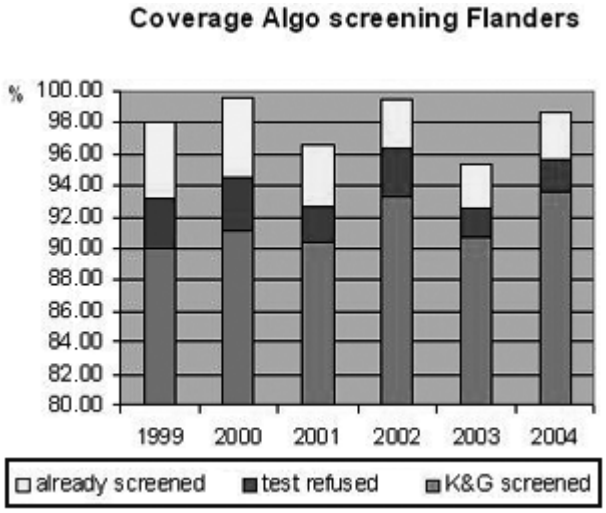

Figure 3
Screening coverage of non-NICU babies in Flanders during the period 1999-2004.

Coverage of the UNSHP

From the beginning of 1999 to the end of 2004, 364,506 non-NICU babies were eligible for Algo® screening in Flanders. An invitation for hearing screening was given to 97.91% of them; 2.09% could not be contacted. 91.50% of the eligible babies were screened by K&G (n = 333,503), 2.6% of the babies had parents who refused the test, and 3.81% of the babies were screened in the maternity ward (Figure 3).

Data from the follow-up database indicate that 1,109 children from the screened non-NICU population had twice a "refer", and were referred to a certified centre, resulting in a referral rate of 3.3‰. Of the referred babies, 178 had normal hearing, 44 had no confirmatory report (some of them deceased before hearing could be tested), 47 had a temporary hearing loss, and 840 had a confirmed hearing loss. Of the babies with confirmed hearing loss, 484 (57.6%) had bilateral hearing loss, 356 (42.4%) had unilateral hearing loss. Based on these numbers, one can calculate that 1.45‰ of the screened babies had a PCHI, and 0.53‰ of the parents were falsely alarmed after two "refers".

The programme should provide appropriate training and should monitor the performance of the staff responsible for the screening programme

During the implementation period of the UNHSP, the nurses received in large groups a theoretical training on AABR of 3 hours and a practical training of 2 hours. Three devices for each district were available (for about 1,000 newborns per year).

One programme coordinator at the central office of K&G is working on a fulltime base for day-by-day support of the programme, for a helpline, for contacts with the referral centres, and for maintenance of the database and analysis.

Screening results must be documented and communicated to the parents and child's physicians

The specialized referral centres have a commitment to keep both K&G and the general practitioner informed of the hearing tests results and rehabilitation data via a standardized report. The processing and interpretation of these data permits accurate evaluation of the programme and supportive control of the protocol. At the same time the database gives a clear overview of congenital hearing impairment in Flanders and is an important source for epidemiological research.

Funding

Because the test is fully integrated into the existing K&G-programme of basic preventive child and family care, very little additional time or staff needs to be provided for the UNHSP, the target group can easily be approached, and the test can be offered free of charge. Funding is provided by the Flemish Government.

Discussion

The four pillars of a successful UNHSP are (1) a suitable screening instrument, (2) maximum coverage, (3) an effective follow-up and (4) cost-effectiveness.

The Algo® Portable Newborn Hearing Screener is generally recognized as a suitable and effective instrument for objective hearing screening, with a reported sensitivity of 100% and specificity of 96% in term high risk neonates.[10] Van Straaten *et al.*[2] found a 100% sensitivity in NICU babies with a 94% specificity after the first screening and a 100% specificity after a second screen at

term.[11] Because screening with the Algo® is based on AABR, function of the middle ear, cochlea, auditory nerve, and brainstem auditory pathways is tested. In addition to identify children with severe or profound bilateral hearing loss, the Algo® also identifies infants with unilateral hearing losses. Parents' acceptance of the test is very high, which is another parameter for a good test instrument. By selecting a stimulus intensity of 35 dB HL, the sensitivity-specificity trade-off was biased toward referring infants with even a mild hearing loss. When children with such hearing losses are identified early, systematic intervention programmes can prevent the sequelae of mild bilateral, unilateral or fluctuating conductive hearing loss. The device is easy to use and the immediate "pass-refer" results don't require costly professional interpretation. The screening is not time-consuming: the median test time is about 2-4 minutes if the baby is quiet or sleeping.

Based on the figures of the period 1999 to 2004, the entire K&G screening programme ensures coverage of 91.50%, which is very high. Only 2.6% of the parents refused the test proposed by K&G. Their refusal was based on personal, religious, or cultural reasons. Another 3.81% of newborns were tested before discharge. These screenings are based on independent, autonomous initiatives, and cannot be considered systematic.

A primary concern in follow-up is dropout. A 2-stage protocol as used by the K&G UNHSP reduces dropout. Such a protocol is only feasible when the screening technology has a low false-positive rate. Using the AABR- technology, this requirement is met. Besides reducing dropout, the efficient protocol greatly decreases parental anxiety by providing an answer in a short period of time: only 1 in 2000 parents are falsely alarmed and that at the most for a few weeks. Support by a programme coordinator and a phone-helpline are essential for guaranteed quality screening and follow-up.

1.45 per thousand of the screened babies had PCHI, which is in agreement with other studies.[1] The false-positive rate significantly determines the total cost of a screening programme. Due to the high specificity of the Algo®-AABR, very few babies will be referred for superfluous follow-up testing. In this study the referral rate was 3.3‰, and only 0.53‰ of the screened babies had a false positive result. These figures are much lower than those mentioned in the UNHSP guidelines (Table 1).

Fitting the Algo® universal hearing screening programme into the normal preventive programme of K&G was feasible at no extra personnel costs. The natural skills of the experienced nurses in handling babies are also an important determinant for UNHSP performance.

The timing of the hearing screening is another important determinant for UNHSP performance. Pushing the screening timing to the earliest possible moment after birth is likely to increase coverage, but will increase the amount of false positive results and hence the number of stressed parents and personnel. Part of the referrals in this scenario can be attributed to conductive problems (debris in the external ear canal, amnion fluid in the middle ear) and to delayed maturation of the central nervous system. Recent publications show an increase of specificity (decrease of false positive results) for those test carried out later.[12,13] "False alarms" will shake confidence in parents and professional healthcare takers, which will influence the trigger effect of the screening.

According to medical models, remediation of hearing loss during the first weeks after birth will not result in stimulation of the auditory cortex and hence rehabilitation programs will only be started from the 2nd or 3rd month of live onwards. Therefore, hearing screening immediately after birth is not necessary.

From a socio-pedagogical point of view, the first weeks of life are most crucial for the identification process of parents and their baby. Any concern regarding a congenital anomaly can badly affect parental bonding, the so called "attachment". In order to achieve highest screening coverage, however, it seems most appropriate to screen babies prior to dismissal from the maternal ward. Therefore, most countries planning the organization of a UNHSP consider a hospital based screening shortly after birth. The question is whether this approach does not have a high relational cost. K&G has chosen to offer hearing screening at 4 or 6 weeks after birth which could be fully integrated in their already existing preventive care programme.

An important programme such as *universal* hearing screening, can probably not succeed when based on the voluntary collaboration of screeners or institutions. Even with strong commitments and agreements, good working long-term collaboration is the

weak link. To be successful, a universal programme has to be organized on a governmental level, being part of a project of nationwide health care. The existence of a Flemish public organisation for preventive care, covering the total population of newborns and keeping a database of all children, was the opportunity to organise the screening programme on a community based (population oriented) model.

Screening without reliable follow-up was not an option. It is just a first step. A swift retest procedure for those who failed the first test, a controlled referral to a highly qualified and experienced centre, offering accurate auditory and paediatric assessment, early home intervention, integrated rehabilitation, and a system of data collection and evaluation are as essential as the UNHSP. K&G aimed to integrate all these components in one screening programme. In order to guarantee follow-up of the referred babies, K&G organizes direct referral of babies to certified centres, always in agreement with the general practitioner or paediatrician. This quick referral procedure also saves time and money for the parents and for the community.

Conclusions

Flanders was the first region in the world with a UNHSP freely accessible to the whole population. Between January 1999 and December 2004, 333,503 babies have been screened for hearing loss by the district nurses of K&G using the Algo® Portable AABR device. The UNHSP also includes an efficient registration system and follow-up strategy: almost no children were lost at follow-up. The data confirm the feasibility of a UNHSP meeting all standards and requirements formulated by the American Academy of Paediatrics (and the European Consensus group).

Now that a wide range of screening instruments are available, and early medical intervention is possible, it is no longer acceptable that newborn hearing screening is not a general practice. Every year, thousands of babies, born with a hearing impairment are unable to develop speech and oral communication, because the medical world and the public health policy makers are not aware enough or give less importance to the consequences of this handicap. Early universal hearing screening of newborns should be a public health priority all over the world and most certainly in all well developed countries.

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