

## A pre-paid newborn hearing screening programme: a community-based study

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**Key-words.** Community-based; newborn; hearing screening; pre-paid; automated auditory brainstem response

**Abstract.** *A pre-paid newborn hearing screening programme: a community-based study.* **Objectives:** To help obstetric hospitals and clinics to implement newborn hearing screening and to test the feasibility of a pre-paid model for screening.

**Patients and methods:** From July 2005 to August 2008, we organised a coordinated newborn hearing screening team with portable automated auditory brainstem response (AABR) to provide in-patient screening after delivery and out-patient re-screening at one month of age in birthing facilities throughout Changhua County, Taiwan. This was a community-based study organised by otolaryngologists at a tertiary referral centre.

**Results:** Ten medical facilities participated in our screening programme. 7,139 out of 12,901 neonates delivered in these facilities during the period were screened for hearing loss. 105 (1.47%) babies who did not pass the in-patient screening were re-screened at one month old. Forty (0.56%) babies referred from the re-screening were sent for diagnostic work-up and six of them failed to show up. The overall follow-up rate was 94.3% (99/105). Eleven babies with bilateral hearing loss and eight babies with unilateral hearing loss were diagnosed. The incidence of bilateral hearing loss in our programme was 1.5/1000. The screening rate descended from medical centre to clinic ( $p < 0.0001$ ). There was no significant difference between the referral rates for different levels of birthing facilities ( $p = 0.5611$ ).

**Conclusions:** Our study demonstrates that a pre-paid model using AABR is feasible at all three levels of medical facilities. Pre-paid community-based screening might be an option for developing countries in the implementation of universal newborn hearing screening.

### Introduction

Congenital hearing loss is one of the most common infant deficits hampering language development and learning. The prevalence rate is estimated to be 1-3/1000.<sup>1,2</sup> Early diagnosis and timely intervention can result in substantial benefits.<sup>3-6</sup> There has been a worldwide effort to promote universal newborn hearing screening. Numerous modifications have been made to protocols in different facilities and in different countries.<sup>1,6-11</sup> Though universal newborn hearing screening is widely implemented in developed countries, its introduction in developing countries may be less straightforward. The healthcare

authorities of these countries have to decide on their own priorities in the light of demands for the management of other life-threatening diseases. The competition for government funding makes pre-paid newborn hearing screening a choice for developing countries.

The first hospital-based screening programme in Taiwan was established in 1998, and the first community-based screening programme in 2002.<sup>12,13</sup> The Health Bureau of Taiwan has already launched several projects to enlighten both associated professionals and the public about the idea of newborn hearing screening. However, as in some developing countries, newborn

hearing screening in Taiwan is not paid for by the National Health Insurance (NHI) system, nor is it mandated by law. Newborns whose parents do not pay for the test or who are delivered in medical facilities that do not offer screening are not tested for hearing loss. The health system in Taiwan is composed of three levels of medical facilities – tertiary referral centres, hospitals and clinics (by order of size and/or integration). Data from the Health Bureau of Taiwan shows that, in 2007, 17%, 52% and 31% of newborns in Taiwan were born in tertiary referral centres, hospitals and clinics, respectively. A national investigation in 2002 revealed that the percentage of medical facili-

ties performing newborn hearing screening was 63% of tertiary referral centres and 17% of hospitals. Most of the birthing facilities running their own newborn hearing screening programmes were either tertiary referral centres or large hospitals. It is of paramount importance to persuade hospitals and clinics to implement screening and to educate parents about the necessity of screening to expand the coverage rate of screening in Taiwan.<sup>13</sup>

We therefore initiated a pre-paid community-based newborn hearing screening programme in Changhua County, Taiwan. Our aim was to help obstetric hospitals and clinics to establish a community-based newborn hearing screening programme and to test the feasibility of a pre-paid screening model.

### Patients and methods

From July 2005 to August 2008, parents of newborns in the well baby nursery (WBN) in Changhua Christian Hospital (CCH) were asked if they would be willing to pay for newborn hearing screening. The reasons for screening were explained beforehand in the regular classes for mothers-to-be in CCH. The pre-paid screening model required the informed consent of the parents to pay for the test. Hearing screening was performed during the hospital stay after birth with an ALGO 3i AABR (automated auditory brainstem response, Natus Medical Inc., San Carlos, CA, USA). This machine produced the screening results "pass" or "refer". The average hospital stay for a healthy newborn is 2 to 3 days in Taiwan. Infants who did not pass the initial screening were re-tested on the

following morning during the admission. Those infants discharged with a "refer" result were scheduled for re-screening at one month of age.

From July 2006 to August 2008, hospitals and clinics in Changhua County were invited to join our community-based newborn hearing screening programme. We offered screening personnel, a portable ALGO 3i AABR, information about screening for medical professionals, fact sheets for parents, and diagnosis and management for referred babies according to the requirements of individual facilities. The screening protocol was essentially the same as that of CCH, except that the initial screening and re-screening took place in each birthing facility. Those who did not pass the re-screening at one month of age were referred to CCH for further evaluation. The diagnostic work-up reimbursed by the NHI, including click auditory brainstem response (ABR), auditory steady state response (ASSR), and 1k Hz tympanometry, were arranged before three months of age. Babies who were diagnosed with bilateral permanent hearing loss were fitted with hearing aids and sent to specialised auditory units for early intervention. Babies with unilateral hearing loss were followed up to monitor sequential development.

### Results

During the study period, one tertiary referral centre (CCH), four hospitals and five clinics joined the newborn hearing screening programme. The duration of participation ranged from 5 months to 37 months. Of the 12,901 WBN newborns born in facilities where

tests were performed during this period, 7,139 (55.3%) were screened for hearing loss. One hundred and five (1.47%) babies did not pass the in-patient screening and were re-screened at one month of age. Forty (0.56%) babies who were referred from the re-screening were sent for diagnostic work-up and six of them failed to show up. Three of the six babies lost to the study were bilateral referrals. The overall follow-up rate for re-screening and diagnostic work-up was 94.3% (99/105). Eleven babies with bilateral hearing loss and eight babies with unilateral hearing loss were diagnosed. The incidence of bilateral hearing loss in our programme was 1.5/1000. After correcting for the missing six babies, the positive predictive rates of referral in our screening programme were 19.2% (19/99) at discharge and 55.9% (19/34) one month later.

Twenty medical facilities provide a maternity service with neonatal care in Changhua County. Four of them are capable of, or already perform, newborn hearing screening. Ten (50%) of the rest were enrolled in our programme and an estimated 70% of infants born in Changhua County gained access to our newborn hearing screening programme. Nearly 40% of the babies born in Changhua County have been screened since 2008. One tertiary referral centre, three hospitals and three clinics that had been performing hearing screening for more than 12 months were chosen to compare the results of newborn hearing screening (Table 1). The screening rates were 60.76%, 56.54% and 47.23% for the tertiary referral centre, hospitals and clinics respectively. There was a

Table 1  
Results of newborn hearing screening in different levels of birthing facilities

	Number of infants screened	Screening rate*	Referral rate at discharge**	Number of infants with hearing loss		Number of infants lost to follow-up
				bilateral	unilateral	
Tertiary referral centre	3881	60.76%	1.6%	7	2	4
Hospital	1721	56.54%	1.28%	3	4	2
Clinic	1391	47.23%	1.44%	1	2	0
Total	6993	56.5%	1.49%	11	8	6

\*  $P < 0.0001$  by the chi-square test and  $P < 0.0001$  by trend test.

\*\*  $P = 0.5611$  by the chi-square test.

significant difference ( $p < 0.0001$ ) in the chi-square test for trend, meaning that the screening rates descended in the order of tertiary referral centre, hospital and clinic. The referral rates at discharge were 1.6%, 1.28% and 1.44% ( $p = 0.5611$ ) respectively. There was no significant difference in referral rates at discharge between different levels of birthing facilities.

## Discussion

Most babies are born in hospitals and clinics in Taiwan. It is therefore definitely not enough in terms of coverage to launch a newborn hearing screening programme without the participation of hospitals and clinics. Often, doctors in charge of the screening are otolaryngologists or paediatricians. Otolaryngologists are familiar with hearing screening and paediatricians are familiar with newborn screening. It would be ideal to combine the two professions in newborn hearing screening, which is usually the case in most tertiary referral centres, as well as in large general hospitals in Taiwan. At CCH, the paediatricians are in charge of the targeted hearing diagnosis of babies in neonatal intensive care units (NICU) and otolaryngologists provide hearing

screening for babies in WBN. Obstetric hospitals and clinics on the other hand might need support with the implementation of screening.<sup>10</sup> The expense of setting up and maintaining single-facility-based programmes and handling the referred infants are problems for these hospitals and clinics. We organised a coordinated screening team to reduce the budget with shared resources, both in personnel and equipment. We also arranged for further evaluation and management for the babies referred. In this way, we helped 50% of hospitals and clinics in Changhua County to link to the chain of screening, diagnosis and intervention of babies with congenital hearing loss to build a community-based screening programme.

About 55% of parents who were informed of the necessity of newborn hearing screening through our programme paid for the tests. The screening rate was not satisfactory, but is comparable with other pay-for-test models in Taiwan.<sup>13</sup> The pre-paid model allowed our screening programme to work independently of government funding or private donations at the expense of equality for all newborns. The epidemiological data and experience learned from

this pre-paid model could also be of help for nationwide universal screening in the future. It is a compromise and a temporary way of providing screening in Taiwan until we have a better alternative. It might also be applicable to other developing countries.<sup>14-16</sup> The screening rates in our programme were in accordance with the levels of medical facilities, with the highest being in tertiary referral centre and the lowest in clinics. We presumed that this was because the parents who chose the upper levels of medical services were more prepared to pay extra money for additional services not covered by the NHI. Since fewer than 20% of the infants were born in tertiary referral centres and the screening rate was lowest in clinics, hospitals should be the main target of action for increasing the coverage rate of our model.

The screening tool chosen – otoacoustic emission (OAE) or AABR – depends on the needs of each programme. OAE is cheaper and less time-consuming but it has a higher false-positive rate, and the possibility of false-negative results by missing auditory neuropathy/dys-synchrony has caused concern.<sup>12,13,17-19</sup> Screening with OAE should also take place at least 36 hours after birth to reduce

the referral rate.<sup>12</sup> We chose AABR as our screening tool since the short hospital stay of newborns in Taiwan makes efficient screening with OAE difficult. This is especially true in community-based screening where the screenings are not done every day because of the sharing of resources between different medical facilities. Screeners are often forced to screen babies at less than optimal times. In our programme, initial screenings were usually conducted less than 24 hours after delivery, and the referral rate at discharge was less than 2% in all 3 levels of birthing facilities. The overall referral rate was 1.47% at discharge and 0.56% one month later, which concurs with other studies using AABR and the recommendations of the American Academy of Pediatrics.<sup>7,20-22</sup> The cost of confirming hearing loss in screening programmes with OAE alone, OAE followed by AABR, or AABR alone varies in different programmes.<sup>21</sup> However, parents have to pay more for AABR than OAE in a pre-paid model. This is a financial burden for parents and explains at least part of our unsatisfactory screening rates.

It is not an easy task to get parents to bring their babies back for the follow-up visit. Six out of 40 babies in need of diagnostic work-up were lost to follow-up. Three of the six were lost early in our programme when the audiologists, rather than the screeners, contacted the families for follow-up. Our screeners are now responsible for each baby they screen, from screening to explaining the results and scheduling re-screening one month later. Knowing that the screeners will be involved in the whole process, the parents put more trust in them. When an

infant is referred, mutual contact between the birthing facility and diagnostic centre is highly valued. Double checking from both sides to make sure the infant is properly followed up is vital to prevent loss to follow-up. However, it is the screener's responsibility to encourage the parents to attend as scheduled. Minimising the need for follow-up by using AABR to reduce the numbers of babies referred is also fundamental to fewer cases being lost to follow-up. It was estimated that at least 1 of our 6 losses might have had bilateral hearing loss requiring early intervention. The loss of each baby in need of intervention is a major loss in any hearing screening programme. We passed the data of the cases that were lost to follow-up to the Early Intervention Plan of the Health Promotion Section of Changhua County Public Health Bureau for further monitoring. However, it is a Taiwanese tradition for a pregnant woman to return to the house of her biological parents after delivery and to stay there for one month for a "healthful rest". The baby often returns home with the mother at about one month of age, which makes follow-up even more challenging. A national surveillance and tracking data system could be a solution to this problem.

### Conclusions

Newborn hearing screening is important in giving babies with hearing loss opportunities for better development. Our programme demonstrates that a pre-paid model using AABR is feasible at all levels of medical facilities in Changhua County, Taiwan. Pre-paid community-based screening

might be an option for developing countries in the implementation of universal newborn hearing screening.

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### References

1. Mehl AL, Thomson V. The Colorado newborn hearing screening project. 1992-1999: on the threshold of effective population-based universal newborn hearing screening. *Pediatrics*. 2002;109(1):E7.
2. Dauman R, Daubech Q, Gavilan I, Colmet L, Delaroche M, Michas N, Baldet F, Saget F, Diallo A, Duriez F, Olegaray F, Soriano V, Debruge E. Long-term outcome of childhood hearing deficiency. *Acta Otolaryngol*. 2000;120(2):205-208.
3. Yoshinaga-Itano C, Coulter D, Thomson V. The Colorado Newborn Hearing Screening Project: effects on speech and language development for children with hearing loss. *J Perinatol*. 2000;20(8 Pt 2):S132-S137.
4. Kennedy CR, McCann DC, Campbell MJ, Law CM, Mullee M, Petrou S, Watkin P, Worsfold S, Yuen HM, Stevenson J. Language ability after early detection of permanent childhood hearing impairment. *N Engl J Med*. 2006;354(20):2131-2141.
5. Declau F, Doyen A, Robillard T, de Varebeke SJ. Universal newborn hearing screening. *B-ENT*. 2005; Suppl 1: 16-21.



6. Verhaert N, Willems M, Van Kerschaver E, Desloovere C. Impact of early hearing screening and treatment on language development and education level: evaluation of 6 years of universal newborn hearing screening (ALGO) in Flanders, Belgium. *Int J Pediatr Otorhinolaryngol.* 2008; 72(5):599-608.
7. Iwasaki S, Hayashi Y, Seki A, Nagura M, Hashimoto Y, Oshima G, Hoshino T. A model of two-stage newborn hearing screening with automated auditory brainstem response. *Int J Pediatr Otorhinolaryngol.* 2003; 67(10):1099-1104.
8. Davis A, Hind S. The newborn hearing screening programme in England. *Int J Pediatr Otorhinolaryngol* 2003; 67 Suppl 1:S193-S196.
9. Swanepoel de W, Hugo R, Louw B. Infant hearing screening at immunization clinics in South Africa. *Int J Pediatr Otorhinolaryngol.* 2006; 70(7):1241-1249.
10. De Capua B, Costantini D, Martufi C, Latini G, Gentile M, De Felice C. Universal neonatal hearing screening: The Siena (Italy) experience on 19,700 newborns. *Early Hum Dev.* 2007;83(9):601-606.
11. Van Kerschaver E, Boudewyns AN, Stappaerts L, Wuyts FL, Van de Heyning PH. Organisation of a universal newborn hearing screening programme in Flanders. *B-ENT.* 2007; 3(4):185-190.
12. Lin HC, Shu MT, Chang KC, Bruna SM. A universal newborn hearing screening program in Taiwan. *Int J Pediatr Otorhinolaryngol.* 2002; 63(3):209-218.
13. Lin CY, Huang CY, Lin CY, Lin YH, Wu JL. Community-based newborn hearing screening program in Taiwan. *Int J Pediatr Otorhinolaryngol.* 2004; 68(2):185-189.
14. Habib HS, Abdelgaffar H. Neonatal hearing screening with transient evoked otoacoustic emissions in Western Saudi Arabia. *Int J Pediatr Otorhinolaryngol.* 2005;69(6):839-842.
15. Low WK, Pang KY, Ho LY, Lim SB, Joseph R. Universal newborn hearing screening in Singapore: the need, implementation, and challenges. *Ann Acad Med Singapore.* 2005;34(4): 301-306.
16. Chapchap MJ, Segre CM. Universal newborn hearing screening and transient evoked otoacoustic emission: new concepts in Brazil. *Scand Audiol Suppl.* 2001;(53):33-36.
17. Kennedy C, McCann D, Campbell MJ, Kimm L, Thornton R. Universal newborn screening for permanent childhood hearing impairment: an 8-year follow-up of a controlled trial. *Lancet.* 2005;366(9486): 660-662.
18. Ngo RY, Tan HK, Balakrishnan A, Lim SB, Lazaroo DT. Auditory neuropathy/auditory dys-synchrony detected by universal hearing screening. *Int J Pediatr Otorhinolaryngol.* 2006; 70(7):1299-1306.
19. Kirkim G, Serbetcioglu B, Erdag TK, Ceryan K. The frequency of auditory neuropathy detected by universal newborn hearing screening program. *Int J Pediatr Otorhinolaryngol.* 2008;72(10):1461-1469.
20. Erenberg A, Lemons J, Sia C, Trunkel D, Ziring P. Newborn and infant hearing loss: detection and intervention. American Academy of Pediatrics. Task Force on Newborn and Infant Hearing, 1998-1999. *Pediatrics.* 1999;103(2):527-530.
21. Lin HC, Shu MT, Lee KS, Lin HY, Lin G. Reducing false positives in newborn hearing screening program: how and why. *Otol Neurotol.* 2007;28(6):788-792.
22. Mason JA, Herrmann KR. Universal infant hearing screening by automated auditory brainstem response measurement. *Pediatrics.* 1998;101(2):221-228.

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