
REVIEW

The Hearing Screening Programme for Infants in KK Women's and Children's Hospital — Its Development and Role in Reducing the Burden of Hearing Impairment in Singapore

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ABSTRACT

This paper describes a successful hearing screening programme for infants in KK Women's and Children's Hospital (KKH). Between April 2002 and January 2010, 100,237 (99.8% of all eligible babies) were screened, surpassing the international benchmark of 95%; 0.5% were referred for audiological investigations, significantly below the international benchmark of 4%. The median age of diagnosis in 280 infants diagnosed with Hearing Impairment (HI) [4.8months (1-24)] was lower than in HI children in special schools in Singapore [20.8 months (0-86)]. Hearing aids were fitted for 84 infants at a median age of 7.6 months (2-45) which was higher than the international benchmark of six months, but much lower than in special school children [42.2months (1-120)]. Twenty infants received a cochlear implant at a median age of 20 months (8-38). 69.3% of infants with risk factors for HI were rescreened at three to six months of life. This identified a further 88 infants with HI. Nine were fitted with hearing aids. The incidence of HI in KKH was 3.7 per 1000 infants. The KKH programmes identified 69% of infants with HI born in the restructured hospitals in Singapore, providing the opportunity for early intervention and increased likelihood of a hearing and talking child with HI.

Keywords: Cochlear implants, Deafness, Hearing aids, Newborn

INTRODUCTION

Hearing impairment (HI) is a common disability and a major public health problem. In the United States of America (USA), speech frequency HI was found in 16.1% and 8.5% of adults aged 20 to 69 and 20 to 29 years, respectively¹. 19.5% of adolescents between 12 and 19 years had HI². Deaf adults have higher unemployment rates, are underemployed in the range of occupations and typically earn less than the general population in similar occupations³. Children aged seven to nine with bilateral permanent HI have diminished health status and health-related quality of life⁴. Parents of affected children reported greater financial impact and caretaker burden compared to a control group⁵. This paper describes the establishment of a successful hearing screening programme for infants in KK Women's and Children's Hospital (KKH) and its role in reducing the burden of HI in Singapore.

HEARING IMPAIRMENT IN SINGAPORE

Low has estimated that HI may affect up to 360,000 persons in Singapore and that nearly 3000 young persons below 19 years are born with HI⁶. There is little recent local data. In 1979, 26.5% of armed forces personnel were reported to have HI, defined as a hearing threshold exceeding 25db at 0.5, 1, 2 and 4 Hz⁷. In 1986, Sharpe reported that of 10,745 disabled people aged 0 to 79 years, 27.4% had HI⁸. In 1994, 1.27% of 7778 pilot applicants were rejected for having permanent abnormal hearing thresholds above the minimum standards⁹. In 2002, the prevalence of HI in military conscripts in Singapore was reported to be 36.7 per 1000, of which 4.9 per 1000 were moderate to severe¹⁰. However, the self-administered questionnaire used may have caused an under-reflection of the true prevalence. In a study of school-aged children in Singapore in 1973, 5.4% of 1151 male primary school children aged six to eight failed to pass their hearing test (defined as the failure to hear two or

more of the four frequencies of 500, 1000, 2000 and 4000 cycles per second above a HI of 30 decibels)¹¹.

Students with severe HI received their primary education in the Singapore School for the Deaf (SSD) or the Canossian School for the Deaf (now renamed the Canossian School), then proceeded to mainstream secondary education. In 1979, in a survey of 24 pupils with HI in secondary schools, the majority heard very little of all that was said in class, unless compensatory factors such as visual cues and lip reading were used¹². Yet, three-quarters were above average in mental ability. The most recent study on HI in Singapore was conducted by Low et al in 2001¹³. In the two schools mentioned above, the median ages of diagnosis of HI and intervention were 20.8 months (0-86) and 42.2 months (1-120), respectively. The financial allocation for each child with severe HI was \$11,000 per year, four times that for a child in mainstream education. On average, a student with HI completed primary education in eight years, instead of the usual six and required \$71,500 more than his mainstream peers. They estimated that each year's cohort of affected students would cost an additional \$3 million. Fewer children with HI scored As and Bs in English compared to the national average in the Primary School Leaving Examination. Close to 50% scored Ds, compared to approximately 5% of the national cohort. 15 to 20% scored As in Mathematics and Science compared to 45% of the national cohort. The longer the delay in intervention, the poorer were the academic outcomes.

In 2004, the Minister of State of Education reported that there were 460 children with HI in mainstream schools.¹⁴ In the Child Development Programme in Singapore, only 11 preschool children were seen for visual or HI between 2004 and 2006.¹⁵ If there are approximately 3000 persons affected children and adolescents, there may be many unidentified children in mainstream schools.

THE NEED FOR EARLY DETECTION OF HEARING IMPAIRMENT

Late diagnosis of congenital HI can result in significant delays in speech and language development. Without normal stimulation, the central auditory system remains maximally plastic for approximately three and a half years¹⁶. Most infants start to speak at around one year of life after continuous auditory stimulation from birth.

In a large study in the USA prior to the Universal Newborn Hearing Screening (UNHS) programme, the performance standards of 971 severely affected (ie. deaf and near-deaf) students aged 7 to 14 were assessed using the Stanford Achievement Test (9th edition) and were categorised as "advanced" (superior level beyond grade level mastery), "proficient" (solid academic performance, prepared for the next grade), "basic" (partial mastery of the knowledge and skills fundamental for satisfactory work) and "below basic" (less than partial mastery). The median reading comprehension scores for the entire group fell largely in the "below basic" category, with very similar results in reading vocabulary and mathematical procedures¹⁷. At age five, children who were intervened by 11 months of age had significantly better vocabulary and verbal reasoning skills than those intervened later¹⁸. Even with intervention within two months of diagnosis, children with normal cognitive abilities identified before six months of age had an advantage over those diagnosed later¹⁹. In Colorado, odds risk ratio estimates indicated that in hospitals with UNHS programmes, 80% of the affected children with normal cognition had normal language quotients, due to early diagnosis and intervention²⁰. Infants with unilateral HI also need identification as they have worse oral language scores than their siblings with normal hearing²¹.

In 1984, prior to the onset of the UNHS programmes and cochlear implants, Yiap et al found that only 16.7% of 45 affected preschoolers in Singapore showed a good chance of integrating with normal hearing children in Primary One²². More recently, cochlear implantation before the age of two has resulted in a 90% probability of integration into mainstream kindergarten²³. Such early implantation requires a very early diagnosis of HI. It was also reported to reduce the cost of annual education per child by €2,105 (€ 1105 to €5106)²⁴. However, not all hearing-impaired children undergo cochlear implantation. Yoshinaga-Itago et al showed that 40% of children with hearing aids had normal vocabulary scores at 36 months²⁵. The best indication of the success of the UNHS in Singapore was the announcement in The Straits Times on November 5th 2011 that the SSD (which used a combination of sign language, gestures, speech and other media) would close in 2016 due to shrinking enrollment, as "early diagnosis in babies and advances in hearing aids and cochlear implants" have allowed "most hearing-impaired

Table 1. Risk Indicators Associated with Permanent, Congenital, Delayed-onset, or Progressive Hearing Loss in Childhood

1. Caregiver concerns regarding hearing, speech, language or developmental delay
2. Family history of permanent childhood hearing loss
3. Neonatal intensive care of more than 5 days or any of the following regardless of length of stay: ECMO, assisted ventilation, exposure to ototoxic medications (gentamicin and tobramycin) or loop diuretics (furosemide/Lasix) and hyperbilirubinemia that requires exchange transfusion
4. In utero infections such as CMV, herpes, rubella, syphilis and toxoplasmosis
5. Craniofacial anomalies, including those that involve the pinna, ear canal, ear tags, ear pits, and temporal bone anomalies
6. Physical findings, such as white forelock, that are associated with a syndrome known to include a sensorineural or permanent conductive hearing loss.
7. Syndromes associated with hearing loss or progressive or late-onset hearing loss, such as neurofibromatosis, osteopetrosis and Usher syndrome; other frequently identified syndromes include Waardenburg, Alport, Pendred, and Jervell and Lange-Nielson
8. Neurodegenerative disorders such as Hunter syndrome, or sensory motor neuropathies, such as Friedreich ataxia and Charcot-Marie-Tooth syndrome
9. Culture positive postnatal infections associated with sensorineural hearing loss, including confirmed bacterial and viral (especially herpes virus and varicella) meningitis
10. Head trauma, especially basal skull/temporal bone fractures that require hospitalisation
11. Chemotherapy

children to hear well enough to attend mainstream schools", while enrollment in the Canossian School (which does not teach sign language and focuses on the use of speech) has remained stable at 90 children²⁶.

UNHS data from KKH, Singapore General Hospital (SGH) and National University Hospital (NUH) has shown that the incidence of severe to profound congenital HI was 1.7 per 1000 newborn infants²⁷. It is thus the most common congenital problem detectable by newborn screening in Singapore, more common than congenital hypothyroidism, found in 1 in 3000 newborn babies²⁸. The UNHS is thus a second level prevention of disability, as it is the early intervention of identified infants that prevents or reduces the later disability.

THE SITUATION BEFORE THE UNIVERSAL NEWBORN HEARING SCREENING PROGRAMME IN KKH

KKH practised targeted screening of high risk (HR) infants at six months of age²⁹ which could only identify 50% of infants with congenital sensorineural HI³⁰. Recall of infants for hearing

screening was left to individual doctors, without tracking or surveillance. The rest of the hearing screening was carried out in the community, which Low et al found to be inconsistent¹³. The polyclinic programme consisted of a parental questionnaire and simple testing at periodic intervals between three months and four years of life. These often missed even severe to profound HI. Private sector screening was even more subjective and not even attempted in some infants. Four percent of Singapore children were not immunised, so they were only screened by the School Health Service in primary one.

THE DEVELOPMENT OF THE HEARING SCREENING PROGRAMME IN KKH

In 2000, the Ministry of Health (MOH) formed a committee to study the early detection and intervention of HI in children in Singapore, which recommended that a national UNHS programme was highly feasible and that a software programme for tracking was necessary¹³. In 2002, a Health Service Development Programme (HSDP) grant by the MOH funded the UNHS programme in KKH for

18 months, covering the cost of the equipment, staff and software. The target was to screen 95% of all infants by one month of life and to diagnose and provide intervention for HI by six months of life, in keeping with the recommendations by the 2000 Joint Committee of Infant Hearing (JCIH) of the American Academy of Pediatrics³¹. It became a fully paid service in 2003. The JCIH had also identified children who were at high risk (HR) of HI (Table 1). The UNHS team systematically recalled them for hearing screening at three to six months of life to identify late-onset HI. This High Risk Hearing Screening (HRHS) programme is unique to KKH as no other hospital systematically attempts to diagnose late onset HI in HR infants.

Guidelines on the establishment of a UNHS programme were reported by Lim and Daniel in 2008³². The following factors were taken into consideration in the development of the KKH programme:

1. The newborn population in KKH and the healthcare system. As outpatient postnatal care was provided by a wide range of facilities, it was difficult to ensure that all infants were screened for hearing by one month of age. Both NUH³³ and SGH¹³ had shown that pre-discharge screening was feasible. Furthermore, in 2003, it was noted that 59% of 2331 parents of unscreened newborn infants refused the hearing screening at four months for various reasons, including "my baby can hear" (49.5%), "no family history of hearing loss" (12.6%), "parents can monitor" (13.5%), "the test is too costly" (10.8%), "wait till the baby is older" (7.2%) and "no time" (5.4%)²⁷.

SGH achieved a pre-discharge screening rate of 77.2% with weekday screening¹³. Because of defaulters in the group recalled for outpatient screening, 4.2% of the newborn population was not screened. In 2002, KKH had approximately 14,000 deliveries per year. A similar strategy would require the recall of 3200 newborn infants (62 per week or 12 per working day) and miss approximately 600 infants. The HI incidence in SGH, which had high risk Neonatal Intensive Care Unit (NICU) and Special Care Nursery populations, was 10 per 1000, higher than the published literature. With a similar patient profile in KKH, six infants with HI might be missed per year. It was decided that daily screening by dedicated screeners was needed to screen the approximately 40 newborn infants per day, as well

as those who needed repeat screens.

The infants who did not pass the UNHS needed a referral to the Otolaryngology Department for audiological assessment, which had high costs borne by parents. Although the JCIH had recommended that the referral rate be less than four percent, SGH reduced it to 2.7% with a positive predictive value (PPV) of 51%¹³. For KKH, this would have resulted in approximately 400 yearly referrals or seven weekly referrals, of which 49% would have normal hearing. Defaults, unsuccessful first attempts and other delays would inflate this number. Thus, it was necessary to reduce the referral rate, as well as increase the PPV.

Mothers who delivered vaginally were usually discharged by 24 hours, either at 12 noon or at 5pm. Screening started at 7am to reduce the disruption to the doctors' work flow.

Some infants readmitted for phototherapy needed to be rescreened. Those transferred to the Children's Intensive Care Unit or the Department of Pediatric Surgery for continuation of care also needed to be screened before discharge.

2. The screening test. The Auditory Brainstem Response (ABR) was chosen for several reasons: the recall rate after an inpatient OAE at the time was 6.4%³⁴. For KKH, approximately 900 infants would have to be recalled for outpatient re-screening per year, or 17 per week, which not only would have been difficult to accommodate, but would also result in a large number of defaults. SGH had shown that 48.4% did not return for their outpatient screen¹³. Two-step ABR programmes had reported a referral rate of 3.2% to Otolaryngology³⁴ and a false positive rate of 0.2-0.9%³⁵.

With the largest NICU in Singapore, these graduates were at a higher risk of developing auditory neuropathy/asynchrony than the well baby population³⁶. This would be missed by the OAE, which does not test the hearing pathway beyond the cochlea, as the ABR does.

It was also the screening test used in the first year for the HR screening programme. However, screening a three to six month old using the ABR proved to be difficult in a busy outpatient clinic. The HR screen currently in use is the OAE, which is easier and quicker in a child who is awake. Attempts to identify

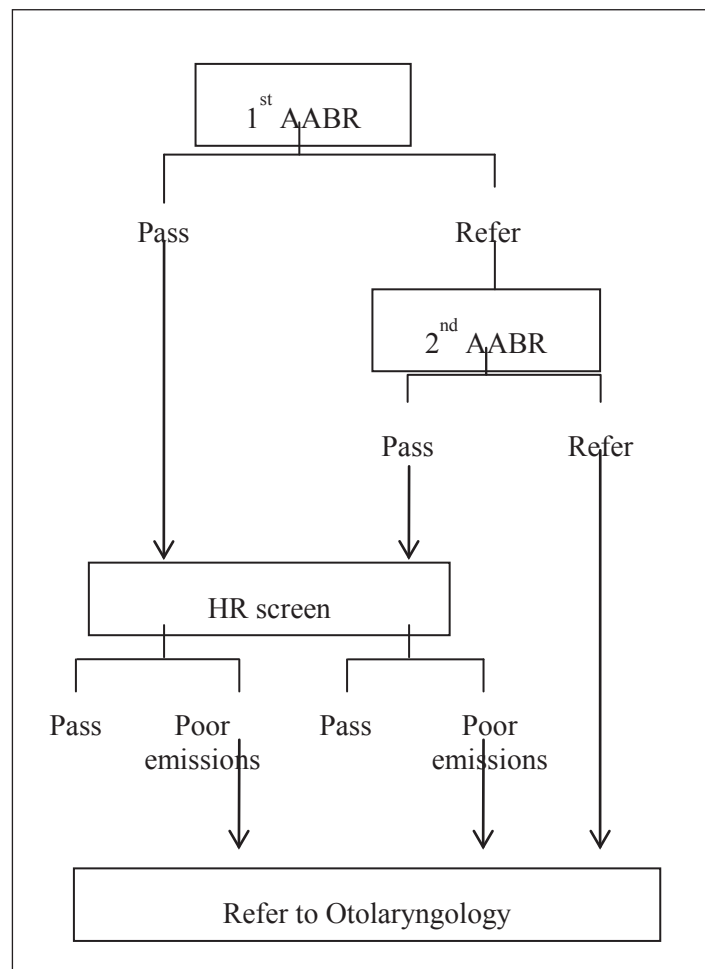


Fig. 1. The universal and high risk hearing screening protocol.

AABR: Automated Auditory Brainstem Response; HR: High risk

suitable outpatient ABR screening equipment have been unsuccessful.

However, compared to the OAE, ABR is a more expensive newborn screening test, takes a longer time to complete and may miss mild hearing loss.

3. The screening and follow-up protocol. Figure 1 shows the two-step ABR UNHS and the HRHS protocols. As inpatient re-screening of an infant with a “refer” result had been shown to reduce false positives³⁷, screening began as early as six hours of age, allowing for rescreening some infants later in their hospital stay.

Although the outpatient UNHS screen was initially performed by the audiologist or audiology technician in the Otolaryngology outpatient clinic, this step was eventually taken over by the UNHS team as obtaining appointments within the

required three to six week period proved to be difficult.

HR factors (shown in Table 1) were identified from the infant’s case notes or from parental history obtained by the screener. These infants were given a screening appointment at three to six months of age, communicated a month earlier through a letter.

Infants who defaulted the repeat screen were contacted by letter with a repeat appointment given. If they defaulted again, the UNHS office staff personally contacted the parents.

4. The screening equipment. Different brands of ABR were tested before the Natus Algo3 (Automated ABR, AABR) was selected. The programme started with four portable units, three for the inpatient and one for the outpatient UNHS. Two units were

replaced in 2011 with the Algo5. The HR screen is performed with the OAE equipment in the Otolaryngology Department.

5. Staff. The UNHS is directed by a neonatologist, as are the programmes in NUH and SGH, as the newborn population is managed by the neonatal department.

The coordinator is a staff nurse who trains and supervises the screeners, manages the logistics, screening equipment and consumables, ensures the smooth and accurate daily screening process and facilitates follow-up.

The data manager is responsible for the HITRACK database. Five screeners receive one-to-one practical hands-on training, are empowered to answer common queries from parents and carry out the daily inpatient and outpatient screens.

Infants who do not pass the UNHS or HRHS are referred to the Otolaryngology Department for audiological assessment. The HRHS is performed by the audiologist or audiology technician in the Otolaryngology Department.

All infants who need audiological assessments are also referred to the neonatal outpatient clinic so that appropriate developmental follow-up for children with HI can be provided.

The screeners faced some challenges in the nursery. The main difficulty was access to the infant, who were either being breastfed, examined by doctors, or were having their serum bilirubin or metabolic screening performed. This was reduced by starting hearing screening at 7am before the ward round began. High myogenic activity of some infants required some tests to be repeated or prolonged. This problem has been reduced considerably with the use of the Algo5. In addition, as more infants remain with the mother after delivery for breastfeeding, screening now requires the infant to be transferred from the mother's bedside to a quiet room for screening, thus lengthening the screening process. The short hospital stay for infants born by normal vaginal delivery does not always allow repetition of the test before hospital discharge. It has been difficult to screen long-staying infants in the NICU, as the ambient noise from the environment and the equipment prevent a successful screen.

6. Communication. Every parent is given a brochure on the UNHS after the delivery of the infant. The newborn screener informs the parents of the hearing screen; verbal assent is taken. The screening result is communicated to parents verbally and recorded in the infant's health booklet. The newborn screener, nurse coordinator, the nurse who discharges the parent and the doctor who reviews the newborn infant all participate in the communication of the need for a repeat screen. The appointment for the repeat screen is given before discharge or communicated by letter from the UNHS office.

7. Financing the programme. For the first 18 months, the programme was funded by the HSDP grant, but became a fully paid service in late 2003. There is now a one-time inpatient payment for the UNHS, which includes all the repeat screens necessary for the newborn infant, including inpatient repeats for failed screens, inaccurate results, repeats after a new risk factor (eg. exchange transfusion) and one outpatient UNHS screen. While the two-step ABR UNHS programme is more expensive than the two-step OAE (US\$32.81 versus US\$26.89 per test) the costs per identified child is similar³⁴. The high risk hearing screen is paid for at the time of the test.

8. Data management, follow-up and surveillance. A commercial database (HITRACK) was used from the start of the programme. The data manager downloads the screening data from the Natus screening equipment on a weekly basis and performs the manual entry of HR factors, as well as outpatient UNHS/HRHS and audiological assessment results (including information on intervention whenever possible). Frequent audits of both the UNHS and HRHS programmes are carried out, as well as adjustments of the protocol whenever necessary. The data manager also audits the daily screening process by reviewing the screening data. Surveillance is the responsibility of the UNHS staff, who ensure that all newborn infants are screened, follow-up appointments matched with medical visits, defaulters contacted and repeat appointments given.

9. Support for the programme. The fact that the UNHS in KKH started with a HSDP grant is a reflection of the support of the MOH. It has

continued to receive the full support of the hospital's administration, as is the case in NUH and SGH. Hence, unlike the USA, the UNHS in Singapore did not need legislation for its implementation. It is important to note that it was established with the cooperation of the Otolaryngology Department, as it would follow that a large number of very young infants would be referred for audiological assessment.

THE OUTCOME OF THE UNIVERSAL NEWBORN AND HIGH RISK SCREENING PROGRAMMES IN KKH

Between April 2002 and January 2010, the UNHS in KKH screened 100,237 newborn infants (99.84% of all eligible infants), thus surpassing the JCIH benchmark of 95%³⁸. The incidence of HI of any type or severity was 2.8 per 1000 newborn infants; 1.8 per 1000 infants had severe-profound HI. The referral to the Otolaryngology Department was 0.5%, which was far below the JCIH benchmark of 4%. Two hundred and eighty infants have been diagnosed to have HI of which 61% were sensorineural, 8.9% mixed and 57% bilateral. The median age at diagnosis was 4.8 months (1-24 months)³⁹. The positive predictive value for the AABR was 81.4%. An accurate negative predictive value could not be calculated as there was no mechanism for identifying children with HI diagnosed later. Hearing aids were fitted for 84 infants at a median age of 7.6 months (2-45 months), which was slightly above the six month benchmark set by the JCIH. Twenty infants with bilateral severe-profound HI with inadequate benefit from hearing aids and auditory-verbal therapy had a cochlear implant at a median age of 20 months (8-38 months). Forty-seven infants were medically treated for conductive HI at a median age of five months (2-24 months). Fifty-two infants were also surgically treated at a median age of 10 months (3-56 months).

Of 3927 infants (69.3% of eligible infants) who underwent the HRHS in KKH between 2002 and 2010, another 88 infants were diagnosed to have HI⁴⁰. The majority (74%) had conductive HI, of which 11(16.7%) were medically treated and 31 (47%) were surgically treated. Nine infants required hearing aids. One infant was referred to the Canossian School. The HRHS programme has shown that the true incidence of HI in infancy was 3.7 per 1000 and not 2.8 per 1000 as suggested by the UNHS programme. It has provided better clarity

and earlier diagnosis of HI in high risk groups. It was initially reported that very low birth weight infants (<1500g) in KKH had a HI incidence of 12.3 per 1000⁴¹. With the UNHS and the refinement of the HRHS, it has been shown to be 34 per 1000⁴². The age of diagnosis of HI in children with Down Syndrome has fallen from later than two years to 6.6 ± 3.3 months⁴³. The combined data of the three restructured hospitals in 2002-2003 has shown that 78% of the births in the restructured hospitals were in KKH, where 69% of newborn infants with severe-profound congenital HI were identified²⁷. Thus, it has made a significant contribution in the early identification of HI in Singapore, creating a window of opportunity for early intervention and increasing the likelihood of a hearing and talking child with HI.

CONCLUSION

The hearing screening programme in KKH has not only allowed the diagnosis and early intervention of congenital HI in a significant number of infants, thus giving them an opportunity for the development of normal speech and language, but has also shown the value of repeat screening in HR infants. It has shown that the true incidence of HI in infancy is 3.7 per 1000 and not 2.8 per 1000 as suggested by the UNHS programme. While it is clear that there are still challenges to be addressed, the reduction of the median age of diagnosis from 20.8 to 4.8 months and of intervention from 42.2 to 7.6 months (before normal infants start to speak) will surely have a lasting impact for the affected children born in this cohort in KKH.

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