Feasibility of implementing a universal neonatal hearing screening programme using distortion product otoacoustic emission detection at a university hospital in Hong Kong

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Objective. To assess the feasibility of implementing a universal neonatal hearing screening programme using distortion product otoacoustic emission detection at a major teaching hospital in Hong Kong.

Setting. Teaching hospital, Hong Kong.

Methods. A total of 1064 infants, together with their mothers, were successfully recruited for the study. The participation rate was 99.3%. A three-stage hearing screening protocol using distortion product otoacoustic emission detection was adopted. Each of the participating infants was screened on three separate occasions (day 1-4, day 5-14, and day 21-30 after birth), irrespective of the test results. A questionnaire was administered to 364 randomly selected mothers to determine whether as consumers of the hearing screening service, mothers would find screening desirable.

Results. Results of the screening demonstrated an incidence of permanent bilateral hearing loss (≥40 dB in the better ear) of 0.28%. The results also showed that 3.5% of the screened infants were referred for subsequent diagnostic audiological assessment, including those suspected with unilateral as well as bilateral hearing loss. Data obtained were comparable to other reported results obtained using multi-stage screening protocols. Taking both the false positive rate and the default rate into consideration, the most appropriate time for screening in this hospital setting appeared to be around day 5 to 14 when infants returned to the hospital’s day centre as out-patients for routine medical follow-up. Since this day centre service is not generally provided by all maternity hospitals in Hong Kong, an alternative time for screening would be around day 21 to 30 when infants could return as out-patients solely for the hearing test. The results of the questionnaire suggested that most mothers thought a neonatal hearing screening would be desirable (91.35%). The majority (81.70%) indicated a preference for screening either within a few days of birth at the maternity ward prior to discharge from the hospital, or between 5 and 30 days when returning to the hospital as an out-patient.

Conclusion. It was concluded that a universal neonatal hearing screening programme could be readily implemented in a maternity hospital setting in Hong Kong.

Key words:
Feasibility studies;
Hearing tests;
Infant, newborn;
Neonatal screening

続け

Objective. 評估於香港一所教學醫院內，推行以畸變產物耳聲發射檢查為新生兒進行聽覺普查計劃之可行性。

設計：描述性研究及問卷調查。

安排：教學醫院，香港。

方法：共有1064名嬰兒及其母親參與研究，參與率為99.3%。採用分三階段進行畸變產物耳聲發射的聽覺普查方案，無論檢查結果如何，每名參與的嬰兒均在三段期間接受檢查，分別是出生後1至4日，5至14日，以及21至30日。並向364
Introduction

It is estimated that approximately 1.5 to 6 in every 1000 newborns suffer from permanent congenital hearing impairment. The great variation in the prevalence of hearing loss reported in studies is thought to relate to variation in the definitions of hearing loss used. When defined as a hearing loss of 40 dB or higher in the better ear, the prevalence ranges from 1 to 3 per 1000 newborns. If unilateral hearing loss is included, the prevalence increases by a further 40%.

The significant negative impact of hearing impairment on the social, emotional, and intellectual development of affected individuals as well as society as a whole, is well documented. The early identification of hearing impairment allows timely intervention to prevent significant speech and language deficits.

In Hong Kong, Maternal and Child Health Centres (MCHC) of the Department of Health used to perform universal hearing screening by using a behavioural (distraction) hearing test at the age of 6-9 months. In keeping with most published studies, hearing impairment was often confirmed only after the first year of age. Recent studies have demonstrated that significantly better language development is associated with the identification of hearing loss and subsequent intervention by the age of 6 months. In an attempt to identify hearing loss at an earlier age, the MCHC have lately revised their practice and have started to perform hearing screening in their centres by using otoacoustic emission detection.

There is a growing consensus in the United States and Europe that all newborns should be screened for hearing impairment within the first few months of life. In 1993, the National Institutes of Health Consensus Development Conference on Early Identification of Hearing Loss in Infants and Young Children recommended that all newborns should be screened for hearing loss within the first 3 months of life, and preferably prior to hospital discharge. In 1994, the Joint Committee on Infant Hearing (JCIH) also recommended that all infants with hearing loss be identified before the age of 3 months, and receive intervention by the age of 6 months. In their 2000 position statement, the principles and guidelines of implementing the screening programme, with appropriate intervention were stressed.

Universal neonatal hearing screening is preferred over screening using a high-risk register, which can usually only identify around 50% or less of infants with hearing loss.

Achieving and maintaining positive parental attitudes is considered essential for any neonatal screening to be sustainable and effective. Negative parental attitudes may be reflected in non-attendance for screening. Reports from the United States and Europe indicate that the majority of mothers considered neonatal hearing screening worthwhile, and that there was a relatively low level of maternal anxiety surrounding neonatal hearing screening compared with other pre- or post-natal screening tests. The current study was undertaken to investigate: (1) the feasibility of implementing a universal neonatal hearing screening programme in a Hong Kong hospital using distortion product otoacoustic emission (DPOAE) detection; (2) when would be the most appropriate time to perform the screening after birth; and (3) if mothers, as consumers of the hearing screening service, would consider screening desirable.

Ethical approval for this study was obtained from the University of Hong Kong Ethics Committee.

Methods

Subjects

The study recruited 1064 infants born in the Tsan Yuk Hospital and their mothers, between 17 May 1999 and 18 October 1999. The total number of births in the hospital during this period was 1076. Tsan Yuk Hospital is one of the major university teaching hospitals in Hong Kong, with a delivery rate of about 5000 births annually, which is typical for a local district hospital in this region. The hospital provides obstetric and neonatal intensive care and follow-up services for women and their newborn infants.

All the mothers were well-informed about the screening procedures involved and the reasons for screening. Those who agreed to participate in the study completed and returned a consent form prior to the testing. In addition, 364 mothers were randomly selected and asked to fill in a questionnaire (Appendices 1 and 2). The questionnaire comprised five questions. The aim of having a short and simple questionnaire was to achieve a higher response rate.
Distortion product otoacoustic emission tests were recorded using the Otodynamics ILO 292 Echoport System (Otodynamics Ltd, Hertfordshire, UK). A calibration check was performed daily to ensure satisfactory transduction of the test stimuli, and thus accuracy of the test results. Testing was performed by a trained nurse for 8 hours per day, 6 days each week. Testing was not performed on Sundays and public holidays. Testing was carried out either in the maternity ward or in the day room of the hospital, neither of which had undergone special acoustic treatment.

A three-stage hearing screening protocol was adopted. Each of the 1064 participating infants was screened on three separate occasions, irrespective of test results (Fig). For babies that appeared healthy after delivery, the first screening took place prior to hospital discharge (day 1-4 after birth). The second screening took place when they returned to the hospital’s day centre as out-patients for routine medical follow-up (day 5-14). The third screening was scheduled for day 21-30 after birth, with the mothers and babies returning to the hospital solely for the hearing test. A similar protocol was adopted for infants admitted to the Neonatal Intensive Care Unit (NICU) with the screening intervals determined from the date of discharge from the NICU rather than the date of birth.

At the time of this study, no universally accepted con-
sensus with regard to testing methodology for DPOAE existed. The screening pass criteria for this study were defined as the detection of DPOAE responses for at least 7 out of 9 points for primary tones of 70-70 dB sound pressure level in the frequency region between 1 kHz and 6 kHz bilaterally. A true response was one in which the amplitude of DPOAE obtained was at least two standard deviations above that of the noise floor, collected in a minimum of three sweep cycles. The aim of these set criteria was to detect hearing loss of 40 dB or more.

Babies that failed the last screening (scheduled for day 21-30 after birth) were referred to an audiologist for a full audiological assessment, which included diagnostic auditory brainstem response (ABR) measurement using the Nicolet Spirit evoked potential system. Upon confirmation of hearing loss, a referral would be made to the ear, nose and throat, and paediatric specialists for consultation, followed by auditory habilitation.

Infants who passed the last screening test were seen by the MCHC at the age of 6 to 9 months for a behavioural hearing test. This was followed by telephone interviews with the parents on two separate occasions when the child was aged 18 months and 36 months. The aim of the interviews was to determine whether the child had passed the MCHC hearing test and whether speech development was normal.

Results

Coverage
The screening results are summarised in Table 1. The participation rate of the 1076 mothers enrolled in the study whose babies were born in the hospital during the study period was 99.3% (n=1068), from which a total of 1064 (98.9%) infants were eventually screened. Four infants were not screened—one died soon after birth, while the other three were transferred to another hospital immediately after discharge from the NICU.

Of the 1064 infants who were screened, 38 (3.6%) demonstrated at-risk factors. The at-risk factors, which were similar to those adopted by the JCIH, were defined in this study as: a positive family history, congenital infection, craniofacial anomalies, very low birth weight (≤1500 g), neonatal jaundice, septicemia (or use of antibiotics), asphyxia, and being admitted to the NICU.

Screening outcomes
The screening resulted in 37 (3.5%) infants being referred for diagnostic audiological assessment, including infants with suspected unilateral as well as those with suspected bilateral hearing loss (Table 1). Three infants were subsequently diagnosed with permanent bilateral hearing loss (≥40 dB in the better ear), and another three with permanent unilateral hearing loss. The former represented 0.28% of the total births (n=1076). Table 2 summarises the types of hearing loss identified.

Of the remainder referred for diagnostic audiological assessment, 8 (21.6%) infants were diagnosed with transient hearing loss in one or both ears, and 21 (56.8%) infants were found to have bilateral normal hearing. The hearing status of two infants could not be determined, as they had left Hong Kong shortly after the referral was made.

Table 1. Overall screening results

<table>
<thead>
<tr>
<th>Category</th>
<th>No. of newborn infants</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total births</td>
<td>1076</td>
</tr>
<tr>
<td>Total participating infants</td>
<td>1068 (99.9%)</td>
</tr>
<tr>
<td>Total infants screened</td>
<td>1064 (98.9%)</td>
</tr>
<tr>
<td>Outcome of screening</td>
<td></td>
</tr>
<tr>
<td>Pass</td>
<td>1027 (96.5% of total infants screened)</td>
</tr>
<tr>
<td>Referral</td>
<td>37 (3.5% of total infants screened)</td>
</tr>
<tr>
<td>Outcome of referral for diagnostic audiological assessment</td>
<td></td>
</tr>
<tr>
<td>Permanent hearing loss</td>
<td></td>
</tr>
<tr>
<td>Unilateral</td>
<td>3 (8.1% of total referrals), 1 of the 3 presented with at-risk factors</td>
</tr>
<tr>
<td>Bilateral</td>
<td>3 (8.1% of total referrals), 1 of the 3 presented with at-risk factors</td>
</tr>
<tr>
<td>Transient hearing loss (e.g. otitis media)</td>
<td>8 (21.6% of total referrals)</td>
</tr>
<tr>
<td>Normal hearing</td>
<td>21 (56.8% of total referrals)</td>
</tr>
<tr>
<td>Defaulted</td>
<td>2 (5.4% of total referrals)</td>
</tr>
</tbody>
</table>

Table 2. Types of permanent hearing loss identified (n=6)

<table>
<thead>
<tr>
<th>Infant No.</th>
<th>Hearing loss</th>
<th>Presence of risk factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Bilateral moderate</td>
<td>No</td>
</tr>
<tr>
<td>2</td>
<td>Bilateral moderate</td>
<td>No</td>
</tr>
<tr>
<td>3</td>
<td>Unilateral moderate</td>
<td>No</td>
</tr>
<tr>
<td>4</td>
<td>Unilateral severe</td>
<td>No</td>
</tr>
<tr>
<td>5</td>
<td>Bilateral moderate</td>
<td>Yes, cleft lip and palate</td>
</tr>
<tr>
<td>6</td>
<td>Unilateral moderate</td>
<td>Yes, septicemia/antibiotics, Neonatal Intensive Care Unit stay</td>
</tr>
</tbody>
</table>

Table 3 gives the profile of the screening programme results, complete with data on pass, fail, and default rates. The pass rates for the first (day 1-4), second (day 5-14) and third (day 21-30) screenings were 40.9%, 78.9%, and 95.8%, respectively. The default rates for the first, second, and third screenings were 24.2%, 14.2%, and 18.0%, respectively.

Telephone interviews
Interviews were successfully conducted with 983 parents...
(95.7% of participants whose infants passed the DPOAE screening). It was noted that 966 babies had passed the subsequent MCHC hearing test at the age of 6 to 9 months and 17 babies had failed the test (Table 4). The 17 babies that failed had then been referred to Child Assessment Centres (CAC) for further diagnostic tests. All of these babies eventually passed the tests and were found to have bilateral normal hearing. The exact reasons why the infants initially failed the hearing test at the MCHC (eg transient hearing loss caused by otitis media) could not be determined through the telephone interviews. It was also noted that the MCHC had referred another 24 babies to the CAC for assessment of suspected speech and language delay. However, all these babies passed the subsequent hearing tests performed as part of the assessment battery.

Forty-four (4.3%) parents remained unable to be contacted at the end of the study (Table 4). Most were thought to have moved house or to have left Hong Kong. Since the incidence of permanent bilateral hearing loss reported in the literature is very low (0.1%-0.3%), the missing information from these 44 babies precludes accurate calculation of the sensitivity, specificity, and false negative rate for the screening programme.

**Questionnaires**

A total of 347 questionnaires were collected prior to the testing (Appendices 1 and 2). The return rate was high (95.3%). Results of the questionnaires are shown in Table 5.

The majority of mothers (88.1%) who responded were aware of the significance of a hearing impairment for their baby’s acquisition of speech and language (Question 1). However, they scored relatively poorly on the questions regarding their knowledge of hearing developmental milestones (Question 2).

Most mothers (91.4%) thought that neonatal hearing screening was desirable (Question 3). The majority (81.7%) indicated that they would prefer their baby to be screened either: (1) within a few days after birth at the maternity ward prior to discharge from the hospital; or (2) between the ages...
of 5 and 30 days when returning to the hospital for outpatient care (Question 4). Most of the mothers (8.6%) who did not express definite approval of the screening process (Question 3) preferred screening to be at or after the age of 3 months at their local MCHC (Question 5).

Discussion

The observed referral rate for infants for subsequent diagnostic audiological assessment of 3.5% is similar to that reported in most published studies and is below the 4% recommended by the JCIH following screening. The observed 0.28% incidence rate of permanent bilateral hearing loss (≥40 dB in the better ear) of all newborns is also close to figures reported in the literature (0.1%-0.3%). The findings of the hearing assessments, together with the results of the telephone interviews, suggest that the screening protocol used had good sensitivity and specificity, although this could not be accurately calculated.

The data on pass, fail, and default rates summarised in Table 3 suggest optimum times to conduct screening. The high default rate for first screening conducted on day 1 to 4 (24.2%) may be due to the manpower restrictions in this study, which deployed only one research nurse. This meant that infants who were born on Saturdays, Sundays, or public holidays may have gone home before screening could be provided. The default rate (18.0%) was also high for the third screening performed on day 21 to 30. This may reflect the fact that some infants left Hong Kong after hospital discharge but may also suggest that some mothers do not wish to return to hospital solely for a hearing test.

The pass rates for the first, second, and third screening tests were 40.9%, 78.9%, and 95.8% respectively, suggesting that the false positive rate would decrease significantly if the screening was performed later. Scheduling screening around day 21 to 30, as opposed to day 5 to 14, would minimise the false positive rate, and thus unnecessary referrals for subsequent full audiological assessment. At this age (approximately 1 month), artifacts of DPOAE recording that can be caused by incomplete clearance of normal foetal middle ear fluid after birth will be minimal, while the baby will still be relatively inactive for ease of testing. However, testing at this time requires the mother to bring the baby back to the hospital solely for the hearing test, and thus this is likely to cause the default rate to rise. It appears that the prospect of decreasing the false positive rate has to be weighed against the benefit of minimising the default rate. This can be a difficult judgement.

When both the false positive rate and the default rate are taken into consideration, the most appropriate time for screening in our hospital setting appears to be around day 5 to 14 when infants return to the hospital’s day centre as out-patients for routine medical follow-up. Since this day centre service is not generally provided by all maternity hospitals in Hong Kong, an alternative to consider would be around day 21 to 30 when infants can return to the hospital as out-patients solely for the hearing test.

The design of the questionnaire aimed to determine whether mothers, as consumers of the hearing screening service, would find the idea of screening desirable. It is interesting to note that most mothers were aware of the significance of a hearing impairment on their baby’s acquisition of speech and language. However, they demonstrated less knowledge of hearing developmental milestones. Therefore, it appears that mothers might overlook hearing problems and that relying on parental identification of hearing problems is inadequate.

Most mothers thought that neonatal hearing screening was desirable. This is consistent with the observed overall participation rate of 99.3%. Of the 1076 babies born in the hospital during the study period, only eight mothers declined consent for their child to participate in the study, indicating they would only consent to well-established and proven hearing testing protocols.

Most mothers preferred hearing to be screened within a few days after birth at the maternity ward prior to discharge from the hospital, or between the ages of 5 and 30 days when returning to the hospital as an out-patient. However, approximately 18% expressed a preference for the screening to be undertaken at their local MCHC, which are usually more easily accessed than hospitals.

Conclusion

This study demonstrates the effective implementation of a universal neonatal hearing screening programme in a maternity hospital in Hong Kong using DPOAE measurement. The observed incidence of hearing loss and referral rate of infants for subsequent diagnostic audiological assessment were similar to those in most published studies using multi-stage screening protocols. The study also demonstrated that mothers believe a neonatal hearing screening to be beneficial for their babies. Taking both the false positive rate and the default rate into consideration, the most appropriate time for screening in this hospital setting appeared to be around day 5 to 14 when infants returned to the hospital’s day centre as out-patients for routine medical follow-up. Since this day centre service is not generally provided by all maternity hospitals in Hong Kong, an alternative time for screening suggested would be around day 21 to 30 after birth, with infants returning to the hospital as out-patients solely for the hearing test.

To determine the ideal universal neonatal hearing screening protocol to be adopted in Hong Kong, many other issues should be considered which have not been addressed by this study. These include:

1. Estimation of screening cost, such as the cost of equipment, disposables, personnel, administration, and follow-up testing;

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Further research on these areas is indicated.

References

Appendix 1. Questionnaire

1. How likely do you think hearing impairment may affect a baby’s normal acquisition of speech and language?
   - Very much likely
   - Fairly likely
   - Not quite likely
   - Not at all
   - Don’t know

2. By what age do you think a baby should be doing the following (please indicate, e.g. 6 months)?
   - Quiets and listens to familiar voice
   - Starts to move eyes or head toward sounds
   - Responds to simple words, such as his or her name, “bye-bye” and “no”

3. Do you think a technically feasible neonatal hearing screening is desirable?
   - Yes (go to Question 4)
   - No (go to Question 5)
   - Don’t know (go to Question 5)

4. If answering “Yes” to Question 3, when and where should the screening be administered?
   - Within a few days after birth at the maternity ward prior to discharge from the hospital
   - Between the ages of 5 and 30 days when the baby is brought back to the hospital as an outpatient
   - Between the ages of 5 and 30 days and at your local Maternal and Child Health Centre
   - Others (please specify):

5. If answering “No” or “Don’t know” to Question 3, would you wish your baby to be screened at all?
   - Yes, and the screening to be administered at or after the age of 3 months at your local maternal and child health centre when attending for general health check-up and vaccination.
   - Yes, and the screening to be administered at the age of about 3 years at nursery schools.
   - Yes, and the screening to be administered at the age of about 6 years at primary schools.
   - Yes, and the screening to be administered at the age of about ______ at (please specify location) ______
   - No

—End—

Appendix 2. 家長問卷

一 你認為聽覺受損會不會影響嬰兒的正常語言發展？
   - 完全不可能
   - 部分不可能
   - 可能
   - 確有機會
   - 不清楚

二 你認為嬰兒應在何時開始接受以下反應？（舉例：六個月）
   - 聽到熟悉的人的聲音會靜下來聆聽
   - 聽到聲音會望著或把頭轉向聲音來源
   - 對單字作出反應，如“他／她的名字、「拜拜」、「唔好」

三 你認為為初生嬰兒作聽覺測驗是否恰當？
   - 是（請跳到第四題）
   - 否（請跳到第五題）
   - 不清楚（請跳到第五題）

四 若你認為「是」，那麼應在何時及何地作測驗較為合適？
   - 嬰兒出生前於醫院產科病房
   - 嬰兒出生後五至三十天內於親子中心
   - 嬰兒出生後五至三十天內於家庭
   - 其他（請註明）

五 若你認為「否」或「不清楚」，那麼你是否希望子女接受聽覺測驗？
   - 是，於出生後三個月至兩歲內於親子健康中心
   - 是，於兩歲至三歲內於學校
   - 否

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