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Low cost calibrated mechanical noisemaker for hearing screening of neonates in resource constrained settings

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***Background & objectives*: There is a need to develop an affordable and reliable tool for hearing screening of neonates in resource constrained, medically underserved areas of developing nations. This study valuates a strategy of health worker based screening of neonates using a low cost mechanical calibrated noisemaker followed up with parental monitoring of age appropriate auditory milestones for detecting severe-profound hearing impairment in infants by 6 months of age.**

***Methods*: A trained health worker under the supervision of a qualified audiologist screened 425 neonates of whom 20 had confirmed severe-profound hearing impairment. Mechanical calibrated noisemakers of 50, 60, 70 and 80 dB (A) were used to elicit the behavioural responses. The parents of screened neonates were instructed to monitor the normal language and auditory milestones till 6 months of age. This strategy was validated against the reference standard consisting of a battery of tests - namely, auditory brain stem response (ABR), otoacoustic emissions (OAE) and behavioural assessment at 2 years of age. Bayesian prevalence weighted measures of screening were calculated.**

***Results*: The sensitivity and specificity was high with least false positive referrals for 70 and 80 dB (A) noisemakers. All the noisemakers had 100 per cent negative predictive value. 70 and 80 dB (A) noisemakers had high positive likelihood ratios of 19 and 34, respectively. The probability differences for pre- and post- test positive was 43 and 58 for 70 and 80 dB (A) noisemakers, respectively.**

***Interpretation & conclusions*: In a controlled setting, health workers with primary education can be trained to use a mechanical calibrated noisemaker made of locally available material to reliably screen for severe-profound hearing loss in neonates. The monitoring of auditory responses could be done by informed parents. Multi-centre field trials of this strategy need to be carried out to examine the feasibility of community health care workers using it in resource constrained settings of developing nations to implement an effective national neonatal hearing screening programme.**

Key words Auditory response - neonatal hearing screening - noisemakers - sensitivity - specificity

Detecting and rehabilitating neonates with hearing impairment is a healthcare priority. In developed nations legislation exists to enforce rehabilitation latest by 6 months of age[1]. Delay in intervention beyond 6 months results in low educational and employment levels in adulthood[2,3]. Every year in India an estimated 1,50,000 newborns are born with hearing impairment. A majority of these are rehabilitated as late as 5 yr of age[4]. The delay is estimated to be greater in rural areas. Neonatal hearing screening has been in corporated as a component of the National Programme for Prevention and Control of Deafness (NPPCD)[5]. To ensure identification and rehabilitation latest by 6 months, hearing screening will be carried out house to house by behavioural methods using noisemakers. The screening will be performed by anganwadi workers (AWW) and Accredited Social Health Activists (ASHAs) under the supervision of multipurpose health workers. Neonates, who fail the screening test will be referred to the nearest primary health center[6,7].

The NPPCD draft document[5] does not explain the technical specifications of the noisemakers. Locally available bells and rattles which produce high and low frequency sounds are recommended. Since this method is subjective it results in low reliability and hence cannot be used to standardize the testing protocol. Also comparisons across regions are not possible. There is clear evidence about the high sensitivity and specificity of two electrophysiological measures, otoacoustic emissions (OAE) and the auditory brainstem response (ABR)[8]. In the resource constrained settings of developing nations there is a need to examine the relevance of less expensive behavioural methods. Studies using a variety of automated noisemakers such as auditory response cradle, crib-o-gram and behavioural screening research device (noise warblet) and multichannel infant reflex audiometry have demonstrated consistent behavioural responses to 2 - 3 kHz broad band noise at 70 - 90 dB (A). At 50 and 60 dB (A) the responses are not consistent, resulting in poor accuracy[9-11].

There is no published literature on the reliability of health worker based screening of neonates for severe-profound hearing loss using calibrated mechanical noisemakers manufactured using locally available materials in a resource constrained Indian setting. The present study was undertaken to evaluate the strategy of screening neonates using auditory behavioural testing along with monitoring of language and auditory milestones till 6 months of age.

Material & Methods

A prospective cross-sectional single-blinded study was conducted in a tertiary care hospital (St. John Medical College & Hospital, Bangalore) in India. The period of data collection was from January 2006 - December 2008. A battery of tests consisting of ABR, OAE and behavioural observational audiometry using a digital audiometer is the standard protocol for neonatal hearing screening. A total of 425 neonates were selected by stratified convenience sampling from a population of 3235 neonates (age range: 1 - 30 days) who enrolled in the ENT (Ear, nose, throat) department of a 1200 bedded multispeciality charitable hospital in south India for hearing screening. Absence of OAE and no waveforms up to 70 and 90 dB (A) stimulus in ABR was defined as severe and profound hearing loss, respectively. Twenty neonates with confirmed severe-profound hearing loss were included. The sample consisted of 231 males and 194 females. Stratified selection was done to ensure that 10 per cent of selected neonates had a risk factor for development of hearing impairment. This was done to ensure similarity to a community setting where 10 per cent are expected to be at risk[7]. All the 20 hearing impaired neonates were rehabilitated by hearing aids/total communication. None of them could afford cochlear implants. Nine of these 20 neonates had known risk factors (3 : family history of hearing impairment, 2 : infections, 2 : hyperbilirubinaemia, 1 : low birth weight, 1 : external ear deformity). The remaining 11 had no risk factors. Socio-economic status was evaluated with the modified Kuppuswamy classification[12] based on education, occupation and income of the head of the family. The socio-economic distribution was: 10 per cent - lower, 12 per cent - upper lower, 52 per cent - lower middle, 16 per cent - upper middle and 10 per cent - upper. The study was approved by the St John's National Academy of Health Sciences Institutional Ethics Review Board. Informed written consent was obtained from the parents before including the neonates in the study. For illiterate parents the consent was obtained after verbal explanation in the presence of a third party witness who also signed the form.

Designing the calibrated noisemaker: Four noisemakers were designed in the form of hollow cylinders made of particle wood. These were selected from a set of educational sound boxes manufactured by a local carpenter and modified in the laboratory of Indian Institute of Science, Bangalore. Plastic and metallic solid spheres were placed inside these cylinders. The intensity and spectrum of the noise produced was recorded and analyzed using a digital sound recorder and analyzer (Micro Track II, Avid Technologies Inc., US and Matlab spectral analysis module). A digital sound recorder was used to measure noisemaker intensity at the level of the actual neonate's pinna at a distance of 5 cm from the external auditory meatus. 96,000 sound samples were captured per second for 60 sec (5,760,000 samples). Fourier analysis algorithms were used for spectral and noise averaging. The dimensions of each cylinder were - height : 8.3 cm, radius: 2 cm, thickness: 3 mm and density: 0.435 g/ml. The nature and number of solid spheres in each of the calibrated noisemakers were as follows : 50 dB (A) (4 plastic spheres of density 1.414 g/ml) , 60 dB(A) (1 metal sphere of density 8.276 g/ml), 70 dB(A) (5 metal spheres of density 8.276 g/ml) and 80 dB (A) (4 metal spheres of density 19.557 g/ml). Holding the curved sides and briskly moving the rattle up and down generated the noise. No electrical power supply was required. The cost of manufacturing was ₹ 500 for a set of four cylinders. All the noisemakers had their fundamental frequency at 2 kHz (range: 1900 - 2200 Hz). The sound intensity of the noisemakers ranged between 2 dB (A) above and below the specified intensity. All measuring instruments were calibrated with ISO 9001 standards to ensure internal validity[13].

Behavioural response to auditory stimulus from the noisemakers by the health worker: Six health workers were trained up to the advanced level of the Primary Ear Care Manual published by World Health Organization[14]. All the workers have completed primary school (up to 5th grade). Thirty hours of training over 3 months was administered to the health workers in the rural health centre by the ENT specialists, audiologists and social workers. These health workers studied the behavioural responses of the neonates using the calibrated noisemakers. The protocol for the testing was as follows.

Setting the stage for testing **-** Test was performed in a quiet room [ambient noise ~ 40 - 50 dB (A)]. Neonates were in a state of light sleep and were placed in their parent's arm. The state of light sleep was confirmed by flipping the eyelid slightly or touching the eyelid lightly with a finger. If there was a quiver of eyelids, it was considered as light sleep. The parent was instructed not to respond to the stimulus. The observer and neonate were separated by a glass screen. The observer was blinded to the stimulus. This was ensured by masking through ear phones and keeping the stimulus producer out of the visual field. A silence of 1 minute was ensured between the stimuli to set the stage to achieve maximum response.

Stimulus - The tester presented the stimulus at a distance of 1 meter from the testing ear and away from the visual field of the observer and neonate. Four stimuli were applied at 50, 60, 70, and 80 dB (A). The stimulus was applied suddenly and maintained at the same intensity for a duration of 2 sec. The tester noted the time at which the stimulus was presented. Stimuli were presented 3 times with a 1 min interval of silence. The observer noted the response and the time at which it was observed. The observer was not aware when the stimulus was presented. The observer looked for one of the following responses: eye widening/eye blink, stirring or arousal from sleep and startle. A qualified audiologist observed if the health worker correctly noted the behavioural response.

Interpretation - The times at which the stimulus was presented by the tester and response noted by the observer were compared. If response occurred within 2 sec of presentation of stimulus, it was taken as a valid response. The presence of at least 2 valid responses was taken as a pass, for that stimulus intensity level.

Checklist for parents to monitor auditory and language milestones up to 6 months: The health worker educated the parent to look for the following milestones till 6 months of age and report immediately if these were not attained. The auditory milestones were: startling or turning head to a loud sound and child getting aroused from light sleep by an "sshhh" sound. The language milestones were: producing differential cries to hunger and happiness, repeating the same sounds (cooing/ gooing). The checklist was printed on a small card (10x10 cm) in English and Kannada (the predominant local language) and was given to the parent. The health worker explained the milestones for illiterate parents.

In case of any delay in the milestone the parents were asked to call the audiologist whose telephone number was printed on the follow up card and take an appointment. Also a health worker was appointed exclusively to ensure regular follow up at 3.5, 6, 9, 12 and 24 months. In case of absenteeism a phone call was given to the contact numbers which were recorded in a register. If, in spite of this, there was further absenteeism a home visit was performed.

Quantifying the screening capabilities of the noise maker: The neonates screened by the health worker were subjected to OAE and ABR by the audiologist. All the neonates were followed up for a period of two years to exclude neuromuscular deficits and global developmental delays. A 2x2 contingency table was constructed, where the rows represented the results of the noisemaker and the columns represented the reference standard results. OAE, ABR and behavioural evaluation at 2 yr of age is the reference standard to confirm hearing impairment. The measures used to evaluate noisemaker screening capability were sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), likelihood ratio of test positive/negative, accuracy of likelihood ratios, pre- and post-test probabilities of positive and negative test. Normograms were constructed to evaluate the shift in probabilities before and after the test[15].

Statistical analysis: Fisher's exact test was used to estimate the statistical significance of calculations derived from the 2x2 contingency tables; 95 per cent confidence intervals were calculated for all the measures of screening. $P<0.05$ was taken as significant. A sample consisting of 405 normal infants and 20 hearing impaired neonates has 90 per cent power to detect an anticipated positive likelihood ratio of 10 or more with 5 per cent type 1 error. A likelihood ratio of 10 or more extrapolates to a significant shift in the pre- and post-test probability. The likelihood ratios were weighted for prevalence to enable generalizations across various regions of the country. Statistical Package for Social Sciences 16 (SPSS .16) was used to perform the statistical analysis. Power Analysis Statistical System (PASS) was used to calculate power.

Results

Efficacy of noisemakers as a tool for neonatal hearing screening: All the noisemakers had a sensitivity of 100 per cent. The specificity was high only for 60-80 dB (A) noisemakers. Though the negative predictive value of all noisemakers was high, the positive predictive value was high only for 70 and 80 dB (A) (Table I). The use of 50 and 60 dB (A) noisemakers as screening tools will increase the number of false positive referrals for severe-profound hearing loss. The 70 - 80 dB (A) noisemakers will screen for severe and profound hearing loss with less false positive referrals.

Figs 1-4 show the pre-test/prior and post-test/posterior probabilities of the noisemakers to detect a case of severe-profound hearing impairment. The likelihood ratio of a positive test was highest for 70 and 80 dB (A) noisemakers. This also reflects the significant shift in the pre- and post-test probabilities. These Bayesian techniques further reinforce the efficacy of the 70 and 80 dB (A) noisemakers as screening tools for neonates with severe and profound hearing loss. All these results were statistically significant $P<0.001$.

Environment for screening of neonates: In our study the 70 and 80 dB (A) noisemakers elicited consistent behavioural responses when the infant was in light sleep in a room with an ambient noise of 40 - 50 dB (A). If the neonate failed to respond to 70 and 80 dB (A) stimuli the likelihood of severe or profound hearing impairment was 19 and 33, respectively. This strong response is attributed to the strength of the stimulus above the ambient noise floor. The low likelihood ratio for 50 and 60 dB (A) was probably due to the poor signal to noise ratio. At an ambient room noise of 40 - 50 dB (A) , a 5/10 dB rise in the signal was not strong enough to elicit a clear behavioural response. Also neonates are not developmentally ready to respond at these levels.

Table. Screening characteristics of the noisemakers

	Noisemaker			
	50 dB (A)	60 dB (A)	70 dB (A)	80 dB (A)
Sensitivity (%)	100 (79.9-100)	100 (79.9-100)	100 (79.9-100)	100 (79.9-100)
Specificity (%)	69.4 (64.6-73.8)	86.9 (83.1-89.9)	94.8 (92-96.6)	97 (94.7-98.3)
PPV(%)	13.9 (9.1-20.4)	27.4 (18.5-38.6)	48.8 (34.2-63.5)	62.5 (45.2-77)
NPV (%)	100 (98.6-100)	100 (98.9-100)	100 (99.1-100)	100 (99.1-100)
NPV accuracy (%)	70.8	87.52	95.05	97.17
Likelihood ratio +	3.3 (2.8-3.7)	7.6 (5.9-9.8)	19.2 (12.7-29.2)	33.75 (19.32-58.9)
Pre-test probability + (%)	4.9	4.9	4.9	4.9
Post-test probability + (%)	13 (8 -21)	27 (18-39)	48 (33-64)	63 (43-78)
Fisher's exact test	$P = 0.001$	$P = 0.001$	$P = 0.001$	$P = 0.001$

95 % confidence intervals; PPV, positive predictive value; NPV, negative predictive value; LR, likelihood ratio

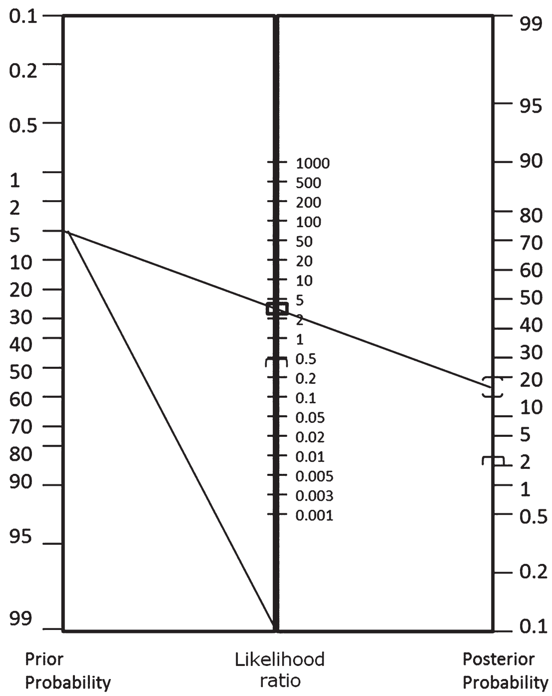

Fig. 1. Normogram for 50 dB (A) noisemaker. Upper line and lower lines indicate the likelihoods for test positive and negative, respectively.

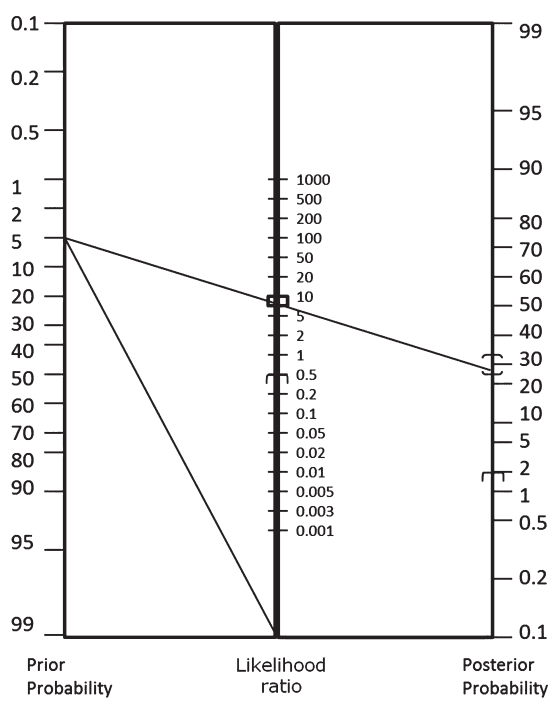

Fig. 2. Normogram for 60 dB (A) noisemaker. Upper line and lower lines indicate the likelihoods for test positive and negative, respectively.

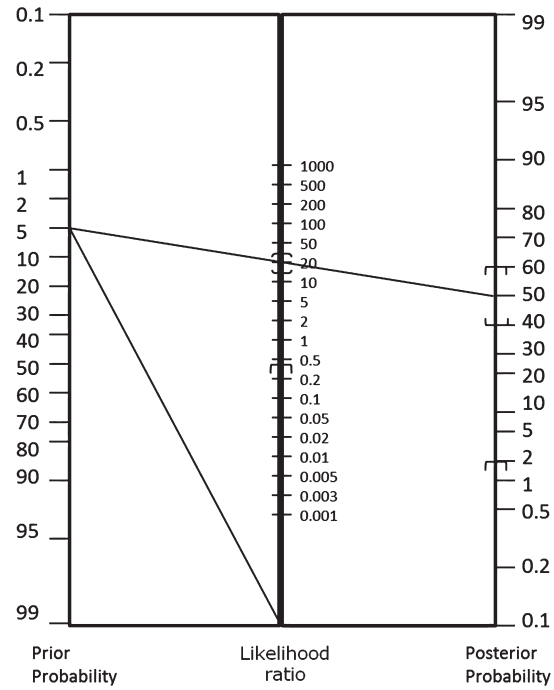

Fig. 3. Normogram for 70 dB (A) noisemaker. Upper line and lower lines indicate the likelihoods for test positive and negative, respectively.

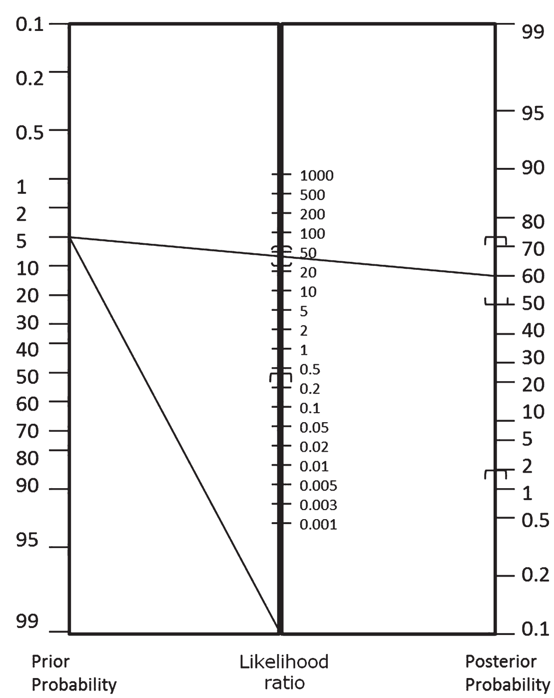

Fig. 4. Normogram for 80 dB (A) noisemaker. Upper line and lower lines indicate the likelihoods for test positive and negative, respectively.

In a community based setting, higher ambient noise is expected. The effectiveness of 50 and 60 dB (A) noisemakers would be further reduced.

Reliability of behavioural observation by the health worker: All the observations made by the health worker were verified by a qualified audiologist. All behavioural responses noted by the health worker were valid. There was no incorrect response noted. None of the families migrated during the study period neither were there any mortalities. Hence follow up was complete.

Discussion

In our study 70 and 80 dB (A) noisemakers could effectively screen infants with severe and profound hearing impairment. Also, in comparison to 50 and 60 dB (A) noisemakers the false positive referral rates were low. Mechanical toys have been employed in conjunction with noisemakers as a screening tool at 9 months of age using the distraction method. At 9 months these tests have shown a positive predictive value of 35-80 per cent to screen mild-moderate hearing loss[16-18]. Studies using a variety of automated noisemakers such as the auditory response cradle, crib-o-gram and behavioural screening research device (noise warblet) and multi channel infant reflex audiometry have demonstrated consistent behavioural response to a 2-3 kHz broad band noise at 70-90 dB (A). At 50 and 60 dB (A) noisemakers have poor sensitivity and specificity. The poor response is explained by the natural developmental delay in neonatal response to these intensities[19]. The state of the infant could be another confounding factor. Though a state of light sleep was ensured before testing, there was a possibility of the neonate going to deep sleep during the testing.

The 70 and 80 dB (A) noisemakers were very sensitive in screening for severe and profound hearing impairment. The specificity was also high for these noisemakers. There were no neonates with mild or moderate hearing loss in our test population and hence the usefulness of this device to screen for mild - moderate hearing loss could not be assessed. The Joint Committee for Infant Hearing Screening Position Statement (1994)[7] has stated that behavioural measures, including automated behavioural techniques, cannot validly and reliably detect the criterion hearing loss of 30 dB (A) hearing level in infants less than 6 months of age. This conclusion was based on studies that have reported that 35 - 80 per cent false positive results and up to 38 per cent false negative results can occur with behavioural screening. The high sensitivity and specificity of 70 and 80 dB (A) noisemakers observed in our study could be attributed to the following factors. The stimuli used were intense and it was ensured that the test was performed in a quiet environment with the newborn in light sleep. The criteria for referral were severe to profound hearing loss unlike other studies where even mild to moderate hearing loss were considered. In addition, blinding the observer to the stimulus reduced the bias of over- reporting.

There was a significant shift in pre- and post-test probabilities for test positive (screen pass) for 70 and 80 dB (A). Previous studies have not employed and reported these results for the screening of newborn hearing. Use of prevalence weighted Bayesian methods allows for the results to be used in neonatal populations with different prevalence rates.

At 2 yr follow up none of the infants who had passed the initial screening had delay in age appropriate auditory or language milestones. Delayed onset hearing loss has been reported in infants with cytomegalovirus infection, those requiring extracorporeal membrane oxygenation and even in infants with no risk factor for developing hearing loss[20,21]. It has been reported that at 6 months of age behavioural response to auditory stimulus is an effective method to evaluate hearing[18].

In conclusion, hearing screening using calibrated mechanical noisemakers of 70 and 80 dB (A) with follow up monitoring of auditory and language milestones was found to be an effective strategy to identify severe and profound hearing loss in neonates by 6 months of age in a controlled setting. Parents can also be educated by these trained health workers to identify age appropriate auditory responses in their children by 6 months of age. Multi-centre field trials need to be carried out to examine the feasibility of community health workers and teachers using this strategy in resource constrained settings of developing nations to implement an effective initial national neonatal hearing screening programme

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