

Neonatal hearing screening programme and challenges faced by the developing country: A tertiary care hospital experience

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Abstract

Objective: To screen new-borns to diagnose any hearing impairment early.

Method: The prospective, cross-sectional study was conducted at the Department of Otorhinolaryngology, Head and Neck Surgery Liaquat National Hospital, Karachi, from November 1, 2020, to April 30, 2021, and new-borns of either gender aged >12h born via spontaneous vaginal delivery, induced labour, and Caesarean section. A predesigned questionnaire was used to collect detailed case history, including gestational age, duration of labour, and other prenatal, natal, and postnatal risk factors. Otoacoustic emission test was performed, and infants referred twice were scheduled for complete diagnostic evaluation and brainstem evoked response audiometry. Data was analysed using SPSS 23.

Results: Of the 267 neonates, 249(93.3%) passed the first screening. Of the remaining 18(6.7%) neonates, 8(44.4%) passed the second screening, while 10(55.5%) were asked to come for a follow-up after three weeks. Of them, 3(30%) returned for check-up, while 7(70%) did not show up.

Conclusion: Neonatal risk factors associated with hearing loss need to be identified, and a comprehensive hearing screening programme is required for neonates.

Keywords: Hearing, Otoacoustic emission, High risk, Neonatal screening. (JPMA 73: 1788; 2023)

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Introduction

Screening neonates for hearing impairment is the standard care in many developed countries. Approximately 1 to 3 infants per 1,000 have a hearing impairment¹ with a 10-20-fold increase in high-risk children.² It was estimated in 2018 that 466 million people, approximately 6.1% of the world's population, live with disabling hearing loss and 34 million (7%) of these are children. This frequency may rise to 630 million by 2030 and reach more than 900 million by 2050.³

Hearing loss is the second most prevalent disability in the global trend of developmental disabilities⁴ in which the frequency of hearing loss in Pakistan is 13 per 1,000 live births compared to 4 per 1,000 live births worldwide.⁵ Universal neonatal screening protocol is well established in the developed countries.⁶ According to a World Health Organisation (WHO) report of new born and infant hearing screening in 2009 updates, there is no organised nationwide hearing screening programme in South Asian countries.⁷

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Busse et al. have described the existing neonatal hearing screening programmes, their outcomes, and factors influencing the neonatal hearing screening policy and performance across 47 countries and regions.⁸ The reason behind missing patients in low- and middle-income countries (LMICs) are logistical constraints, cost issues, poor infrastructure, and lack of proper patient data management and tracking systems. Limited access to healthcare units, time constraints, and other co-morbidities cause dropouts.⁴

The current study was planned to screen neonates to diagnose any hearing impairment as part of a programme launched at health facility in Pakistan.

Patients and Methods

The prospective, cross-sectional study was conducted at the Department of Otorhinolaryngology, Head and Neck Surgery Liaquat National Hospital, Karachi, from November 1, 2020, to April 30, 2021. After approval from the institutional ethics review committee, the sample size was calculated using the WHO calculator while assuming the prevalence to be 6.6%,⁹ margin of error (d) 3%, and alpha (α) 95% confidence level. All children born in the hospital during the stipulated time were assessed. Those included were neonates of either gender aged >12h who were born via spontaneous vaginal delivery (SCD, induced labour or Caesarean section (CS). Those born with external ear

deformities (microtia/anotia/meatal atresia) and those whose parents refused to give consent for the screening test were excluded.

After taking informed consent from the participating parents, the new-borns were studied, and data was recorded to include mode of delivery, gestational age, duration of labour, Appearance-Pulse-Grimace-Activity-Respiration (APGAR) score, Hammersmith neonatal neurological examination (HNNE) score, low birth weight (LBW), family history, radiation exposure, addiction, ototoxic drug use, birth trauma, in-utero infection, intrauterine growth retardation (IUGR), craniofacial anomalies, mechanical ventilation, neonatal intensive care unit (NICU) stay, hyperbilirubinaemia, neonatal meningitis, electrocardiogram (ECG) for long corrected QT interval (QTc), parental concern (Annexure).

After pre-screening parental counselling, the neonates were subjected to otoacoustic emission (OAE) testing (Messers Grason Stadler Instrument; GSI, Corti, USA).

Bilateral testing during natural sleep was done.¹⁰ The screening was reported as 'Refer', meaning hearing problem detected, or 'Pass', meaning hearing problem not detected, in one or both ears, and the results were handed over to the child's parents. One copy was kept for hospital records and future correspondence.

Children who passed the test were not followed up, whereas those who did not pass the OAE test in the first attempt and had risk factors associated with congenital hearing loss were given a follow-up appointment for further evaluation within three weeks of birth. They were clinically examined with an otoscope and were advised for brainstem evoked response audiometry (BERA).

Data was analysed using SPSS 23. Mean and standard deviation were computed for quantitative variables, while frequencies and percentages were computed for qualitative variables. Chi-square test was used to evaluate association between risk factors and referral rates. $P \leq 0.05$ was considered statistically significant.

Annexure: Questionnaire

B/O: MRN #/OPD file #:
 DOB: Contact#:
 Mode of Delivery: Spontaneous/Caesarean Section/Induced Labour Gestational Age: (Preterm, Term, Post term) The duration of Labour:

Risk Factors

Prenatal	Natal	Postnatal
Family History of Hearing loss	Head Trauma/Birth Trauma	Mechanical ventilation more than 48 h
Radiation Exposure	Apgar Scores ** HNNE Scores ****	Craniofacial Anomalies Associated syndrome***
Addiction/ alcoholism	Very Low Birth Weight (<1.5kg)	NICU Stay > 2 Days
Maternal screening For In-Utero Infection (CMV*)	Singleton/Twins/or More	Hyperbilirubinaemia requiring Exchange
Ototoxic medication given to Mother		Neonatal Meningitis ECG for long QTc
Intrauterine Growth Retardation		Ototoxic Medication
No Identifiable Risk Factor		Parental Concern (Regarding Hearing, Speech, Language, or Developmental Delay)

*Cytomegalovirus (CMV), syphilis, herpes, rubella, toxoplasmosis; ** Appearance-Pulse-Grimace-Activity-Respiration (APGAR) scores of 0-4 in 1min, or 0-6 in 5min; *** Genetic syndromes; ****Hammersmith Neonatal Neurological Examination (HNNE).

Signature: Name of Screening Personnel:
 (Filled by duty doctor)
 OAE testing: PASS REFER
 Right Ear
 Left Ear
 Name and signature: Date:
 OAE: Otoacoustic emission

Results

Of the 319 children born during the study period, 267(83.7%) were screened. The remaining 52(16.3%) neonates could not be screened, including 39(12.2%) who expired.

Of the 267 neonates screened, 249(93.3%) passed the first screening. Of the remaining 18(6.7%) neonates, 8(44.4%) had problem unilaterally and 10(55.5%) bilaterally (Figure-1).

Risk factors were identified in 42(15.7%) newborns. The most frequent risks were ototoxic medications taken by the mother, radiation exposure to mother, NICU stay and preterm delivery <33 weeks of gestation, low APGAR score and twin pregnancies (Table).

In 36(13.48%) neonates with any risk factor, the result of OAE test was positive bilaterally. In infants with no risk, the result was positive in 213(79.78%) (Figure 2).

Among the 18(6.7%) subjects who were not positive on first

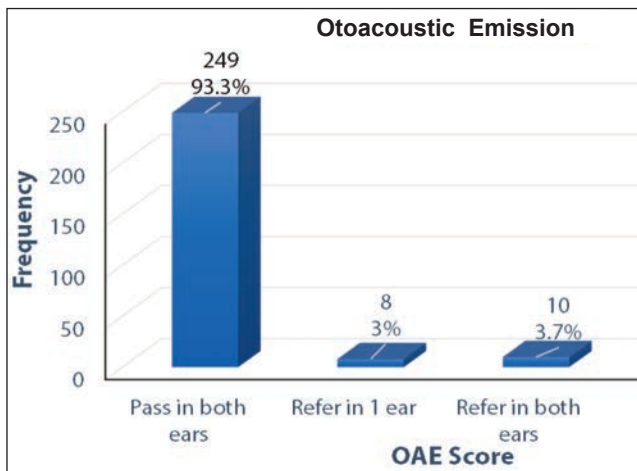


Figure-1: Otoacoustic emission (OAE) scores.

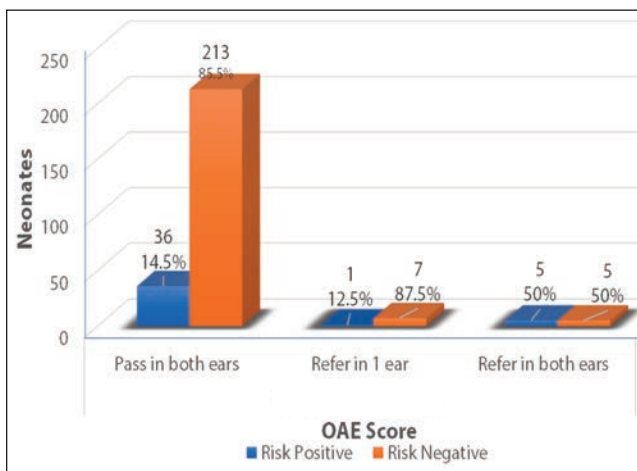


Figure-2: Otoacoustic emission (OAE) score and risks.

Table: Association of risk factors with otoacoustic emission (OAE).

Risk Factors	Otoacoustic Emission			p-value
	Pass in both ears	Refer in 1 ear	Refer in both ears	
Mode of delivery				
SVD	121 (48.6)	4(50)	4(40)	0.931**
C-section	128(51.4)	4(50)	6(60)	
Gestational Age				
Pre Term	10(4)	0(0)	3(30)	0.033*
Term	238(95.6)	8(100)	7(70)	
Post Term	1(0.4)	0(0)	0(0)	
Radiation Exposure				
Yes	2(0.8)	0(0)	0(0)	1.000**
No	247(99.2)	8(100)	10(100)	
Addiction				
Yes	1(0.4)	0(0)	0(0)	0.964**
No	248(99.6)	8(100)	10(100)	
In-utero Infection				
Yes	1(0.4)	1(12.5)	0(0)	0.060**
No	248(99.6)	7(87.5)	10(100)	
Ototoxic Medication to Mother				
Yes	12(4.8)	0(0)	1(10)	0.605**
No	237(95.2)	8(100)	9(90)	
Head/Birth Trauma				
Yes	0(0)	0(0)	1(10)	<0.001*
No	249(100)	8(100)	9(90)	
APGAR Score				
4-6	8(3.2)	1(12.5)	0(0)	0.274**
>7	241(96.8)	7(87.5)	10(100)	
HNNE Score				
7/5	3(1.2)	1(12.5)	0(0)	0.237**
8/5	2(0.8)	0(0)	0(0)	
9/5	244(98)	7(87.5)	10(100)	
Low birth weight				
Yes	1(0.4)	0(0)	0(0)	0.964**
No	248(99.6)	8(100)	10(100)	
Singleton/Twin or more				
Yes	6(2.4)	0(0)	1(10)	0.390**
No	243(97.6)	8(100)	9(90)	
MV>48 hours				
Yes	2(0.8)	0(0)	0(0)	1.000**
No	247(99.2)	8(100)	10(100)	
NICU stay >2 days				
Yes	1(0.4)	0(0)	0(0)	0.964**
No	248(99.6)	8(100)	10(100)	
Neonatal Meningitis				
Yes	2(0.8)	0(0)	0(0)	1.000**
No	247(99.2)	8(100)	10(100)	
Ototoxic Medication to Neonate				
Yes	2(0.8)	0(0)	0(0)	1.000**
No	247(99.2)	8(100)	10(100)	
Parental Concern				
Yes	3(1.2)	0(0)	0(0)	1.000**
No	246(98.8)	8(100)	10(100)	
Risk				
Positive	36(14.5)	1(12.5)	5(50)	0.020*
Negative	213(85.5)	7(87.5)	5(50)	

SVD: Spontaneous vaginal delivery, C-section: Caesarean section, APGAR: Appearance-Pulse-Grimace-Activity-Respiration, HNNE: Hammersmith neonatal neurological examination, MV: Mechanical ventilation, NICU: Neonatal intensive care unit.;Chi-square test was applied; *Significant. ; **Non-significant 0.05

screening, 8(3%) passed the second screening, while 10(3.7%) were asked to come for follow-up. Of them, 3(1.12%) returned for check-up, while 7(2.6%) did not show up.

Discussion

A child born deaf or with delayed language and speech is felt excluded by society. Besides, 90% of the brain develops by the age of 5 years. A child born deaf must be recognised and rehabilitated as early as possible.¹¹ In the first year of life, undetected hearing impairment has profound adverse consequences that affect most developmental milestones, manifesting in significant lifelong deficits in gross and fine motor skills, cognitive performance, speech and language, and psychological development.¹² The overall socio-economic impact is substantial for the affected child, the family, and the community.¹³ This effect lasts for their whole life, and that child later in life is not a productive part of society.⁴

The Joint Committee on Infant Hearing has recommended the use of OAE in healthy neonates, while infants admitted to NICU should be screened with OAE and BERA due to the higher prevalence of auditory nerve dysfunction in this group, and because the sensitivity and specificity of BERA test is more than OAE test.¹⁴

The institution at which the current study was held is one of the largest private-sector hospitals in Karachi. It caters to people from both rural and urban areas of all provinces of Pakistan and neighbouring countries. The institution follows neonatal screening protocol in two steps.

Children are tested bilaterally, without any sedation with an OAE probe. If the "pass" result is not achieved at first, there is repeat OAE test after three weeks. The "pass" result of the second test is treated as final, and if the result is "refer" the family is advised to go for BERA test.

A study reported that in 51(6.6%) new-borns with risk factors, the result of OAE was either "refer" in one ear 1.8% or both ears 4.8%. In infants without risk factors, the result was "refer" unilaterally in 0.4% and bilaterally in 0.2% neonates. The relative risk of neonates with "refer" was found to be the highest in infants with anomalies, such as complex congenital (44.99%), craniofacial (17.46%), and mechanical ventilation (MV) for >5 days (10.69%). In another study, the highest relative risk of neonates with "refer" were in infants with family history, congenital malformations, and low APGAR score.¹ Lost to follow-up rates for neonatal screening have been reported to be 20% in single-centre studies and 21% in multicentre studies.¹⁵ Maternal factors, including age, marital status, addiction, number of children, insurance and poverty status are all

reasons for loss to follow-up.¹⁶ Child factors, such as birth weight, race, gender and NICU stay, also influenced dropout rates.¹⁷ Family acceptance and cooperation are an integral part of every infant's screening programme. In south-western Iran, 4.23% (457/10,804) of the infants did not come for the second screening stage after a failed newborn hearing screening.¹⁰ Rural areas have poor follow-up rates and more significant diagnostic delays due to limited expertise and audiological services.¹⁸ A study reported that >25% children were lost to follow-up by getting treatment from other institutions not participating in the national screening programme. The other reason was that parents ignored the risk of hearing impairment and showed laziness in scheduling appointments for their child's assessment.^{19,20}

A two-step screening protocol has been suggested for new-borns with an OAE on the second day of birth. A formal BERA test is reserved only for those who fail the OAE. The second BERA was performed on an average 28 days after the initial hearing screening.^{21,22} BERA can be used for hearing screening of high-risk neonates.¹⁰ Pakistan does not have an organised screening protocol at the national level.

In current study, parents of children whose results came "refer" after the first step of screening underwent repeat OAE test before discharge. Out of 18 neonates, 8 came out "pass" on the second OAE test. The rest were called for a follow-up after three weeks, but only 3 were brought for rescreening.

The reasons identified by talking with such parents was their failure to understand the importance of screening as well as early intervention and the need of collaboration between parents and service providers. Other reasons included lack of transport as they were living in small villages and cities, lack of interest as the family was not prepared for further testing because of financial issues, lack of awareness about early hearing loss identification, social and cultural stigma related to deafness, and negative parental attitude towards hearing screening protocol.

Beliefs that are based on superstition and cultural taboos have a strong impact and influence on refuting the screening test. Parental denial of hearing loss along with illiteracy and poor economic status are key factors linked with noncompliance, and delays both diagnosis and timely rehabilitation.⁹ Das et al. reported that mothers' education plays an important role in the follow-up of babies.²³ Social pressures and financial issues are primary barriers encountered during interviews with parents. The reason of poor rate of follow-up was due to multiple visits for screening tests and the waiting time which makes parents

postpone BERA, the confirmatory test. Middle ear diseases, such as effusion, also prevent infants from returning.²⁴

Rosenfeld et al. suggested that using hearing aids for speech and complete resolution of effusion should never affect the usage of amplification device.²⁵

More than 60% of the participants felt that difficulty obtaining appointments was critical for delayed screening. One solution would be to structure hearing screening, diagnostic follow-up, and intervention as integral parts of the childcare system.²⁶ Early identification of hearing loss can prevent speech and language delays and benefit the child socially and academically.¹⁶

The current study has limitations as it comprised short-term data from a single centre. The current study may act as a pilot study giving insight into the benefits of an early screening programme and possible ways to make it better and more organised for the future.

Conclusion

Neonatal risk factors associated with hearing loss need to be identified, and a comprehensive hearing screening programme is required for neonates. Follow-up of infants who needed further evaluation was a difficult task and posed a challenge to the current screening programme which required regular monitoring and setting up goals to implement at the regional and national levels.

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References

1. Wroblewska-Seniuk K, Greczka G, Dabrowski P, Szyfter-Harris J, Mazela J. Hearing impairment in premature newborns—Analysis based on the national hearing screening database in Poland. *PLoS one* 2017; 12: e0184359.
2. Poonual W, Navacharoen N, Kangsanarak J, Namwongprom S, Saokaew S. Hearing loss screening tool (COBRA score) for newborns in primary care setting. *Korean J Pediatr* 2017; 60: 353-8.
3. Yong M, Willink A, McMahon C, McPherson B, Nieman CL, Reed NS, et al. Access to adults' hearing aids: policies and technologies used in eight countries. *Bull World Health Organ* 2019; 97: 699-710.
4. Global Research on Developmental Disabilities Collaborators. Developmental disabilities among children younger than 5 years in 195 countries and territories, 1990-2016: a systematic analysis for the Global Burden of Disease Study 2016. *Lancet Glob Health* 2018; 6: e1100-21.
5. Ahmed S, Sheraz S, Malik SA, Ahmed NR, Farooq S, Raheem A, et al. Frequency of Congenital Hearing Loss in Neonates. *J Ayub Med Coll Abbottabad* 2018; 30: 234-6.
6. Yoshinaga-Itano C, Manchaiah V, Hunnicutt C. Outcomes of universal newborn screening programs: Systematic review. *J Clin Med* 2021; 10: 2784.
7. World Health Organization. New-born and infant hearing screening: current issues and guiding principles for action: outcome of a WHO informal consultation, 09-10 November 2009. Geneva: WHO; 2010
8. Bussé AM, Mackey AR, Hoeve HL, Goedegebure A, Carr G, Uhlén IM, et al. Assessment of hearing screening programmes across 47 countries or regions I: provision of newborn hearing screening. *Int J Audiol* 2021; 60: 821-30.
9. Lwanga SK, Lemeshow S. Sample size determination in health studies: a practical manual. Geneva, Switzerland: World Health Organization; 1991. [Online] 1991 [Cited 2023 May 10]. Available from: URL: <https://apps.who.int/iris/handle/10665/40062>
10. Saki N, Bayat A, Hoseinabadi R, Nikakhlagh S, Karimi M, Dashti R. Universal newborn hearing screening in south-western Iran. *Int J Paediatr Otorhinolaryngol* 2017; 97: 89-92.
11. Davis AC, Hoffman HJ. Hearing loss: rising prevalence and impact. *Bull World Health Organ* 2019; 97: 646-646A.
12. Zehnhoff-Dinnesen A, Albuquerque W, Bolz HJ, Brockmeier SJ, Langer T, Narayan R, et al. Basics of Disorders of Hearing Development. In: Phoniatrics I. Berlin, Heidelberg: Springer: 2020, pp 751-836.
13. Khoza-Shangase K. Early hearing detection and intervention in South Africa: Exploring factors compromising service delivery as expressed by caregivers. *Int J Pediatr Otorhinolaryngol* 2019; 118: 73-8.
14. Joint Committee on Infant Hearing. Year 2007 position statement: Principles and guidelines for early hearing detection and intervention programs. *Pediatrics* 2007; 120: 898-921.
15. Kolethekkat AA, Al Abri R, Hlaiwah O, Al Harasi Z, Al Omrani A, Sulaiman AA, et al. Limitations and drawbacks of the hospital-based universal neonatal hearing screening program: first report from the Arabian Peninsula and insights. *Int J Paediatr Otorhinolaryngol* 2020; 132: 109926.
16. Crouch EL, Probst JC, Bennett KJ, Carroll MC. Evaluating loss to follow-up in newborn hearing screening in a southern state. *J Early Hearing Detection and Intervention* 2017; 2: 40-7.
17. Dudda R, Muniyappa HP, Puttaraju S, Lakshmi MS. A qualitative study on knowledge and attitude towards risk factors, early identification and intervention of infant hearing loss among puerperal mothers-A short survey. *J Clin Diagn Res* 2017; 11: MC01-5.
18. Ghavami N, Haghani S, Borimnejad L. Loss to Follow-up After Newborn Hearing Screening and Its Related Factors. *J Client-Centered Nursing Care* 2022; 8: 159-66.
19. Hunter LL, Blankenship CM, Keefe DH, Feeney MP, Brown DK, McCune A, et al. Longitudinal development of distortion product otoacoustic emissions in infants with normal hearing. *Ear Hear* 2018; 39: 863-73.
20. Sachdeva K, Sao T. Outcomes of newborn hearing screening program: a hospitalbased study. *Indian J Otolaryngol Head Neck Surg* 2017; 69: 194-8.
21. Blanař V, Škvřňáková J, Pellant A, Vodička J, Praisler J, Boháčová E, et al. Effectiveness of neonatal hearing screening system: A 12-year single centre study in the Czech Republic. *J Pediatr Nurs* 2021; 59: e32-7.
22. Schwarz Y, Kaufman GN, Daniel SJ. Newborn hearing screening failure and maternal factors during pregnancy. *Int J Pediatr*

- Otorhinolaryngol 2017; 103: 65-70.
23. Das S, Seepana R, Bakshi SS. Perspectives of newborn hearing screening in resource constrained settings. *J Otol* 2020; 15: 174-7.
 24. Farinetti A, Raji A, Wu H, Wanna B, Vincent C. International consensus (ICON) on audiological assessment of hearing loss in children. *Eur Ann Otorhinolaryngol Head Neck Dis* 2018; 135(1S): S41-S48.
 25. Rosenfeld RM, Tunkel DE, Schwartz SR, Anne S, Bishop CE, Chelius DC, et al. Clinical practice guideline: tympanostomy tubes in children (update). *Otolaryngol Head Neck Surg* 2022; 166(suppl 1): S1-S55.
 26. Wroblewska-Seniuk K, Chojnacka K, Pucher B, Szczapa J, Gadzinowski J, Grzegorowski M. The results of newborn hearing screening by means of transient evoked otoacoustic emissions. *Int J Pediatr Otorhinolaryngol* 2005; 69: 1351-7.
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