Original Article :

Prevalence and Causes of Neonatal Hearing Loss and Follow-Up Default in a Cohort from Saudi Arabia: A Retrospective Analysis

Hanan Ibrahim Almuzaini, MD[1], Khaled Shahat, MD[2], Sara Ibrahim Almuzaini, MBBS[3], Badr Eyad Hafiz[4], Nora Tarq Alreefi[4], Reham Khaled Hawalh [5]

1. Assistant professor in Phoniatrics and Pediatric Audiology, Otorhinolaryngology, Head and Neck Surgery. General and Specialized Surgery Department ,College of Medicine.Taibah University ,Madinah, KSA

2. Assistant Professor of Pediatric and Adolescent Medicine, at the Pediatric Department, College of Medicine, Taibah University, Janadah bin Umayyah Road, Medina 42353, Saudi Arabia; and at Al-Rayan National College of Medicine, Madinah, Saudi Arabia.

3. General Practitioner, Madinah-Saudi Arabia; Email Sara.almuzaini95@gmail.com.

4. Medical interns, College of Medicine, Taibah University, Madinah, Saudi Arabia.

5. Pediatric nurse, Children and Maternity Hospital, Madina, Saudi Arabia.

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Corresponding author:

Hanan Ibrahim Almuzaini, MD, Assistant professor in Phoniatrics and Pediatric Audiology, Otorhinolaryngology, Head and Neck Surgery. General and Specialized Surgery Department ,College of Medicine.Taibah University, Janadah bin Umayyah Road, Madinah 42353, Saudi Arabia, Email dr.hanan.almuzaini@gmail.com

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Abstract

Background and Aims:

The Universal Newborn Hearing Screening (UNHS) program in Saudi Arabia has proven instrumental in curbing the burden of neonatal hearing loss in the country. Recent studies from Saudi Arabia have shown a high rate of neonatal hearing loss and follow up default, along with poor awareness among parents, in the Madina region of the country. The study aimed to investigate the prevalence and causes of neonatal hearing loss and follow-up default, among newborns screened at the Children and Maternity Hospital in Madina, Saudi Arabia.

Methods:

All 500 neonates who were born at the hospital from January 2021 to January 2022, and underwent UNHS were included. Prevalence and causes were expressed as descriptive statistics. A chi-square test was used to correlate clinical risk factors with the occurrence of neonatal hearing loss. A P value <0.05 was considered statistically significant.

Result:

The prevalence of neonatal hearing loss was 1.2%, with 17.8% follow-up default. Exposure to ototoxic drugs, low birth weight, craniofacial anomalies, neonatal intensive care unit admission, maternal age >30 years, and hereditary risk showed a statistically significant correlation with neonatal hearing loss. Transportation difficulties (33.7%), lack of education (28.7%) and poor economic condition (21.8%) were the top causes of follow-up default.

Conclusion:

The study reiterates the dire need to raise awareness among parents of newborns regarding the detrimental effects of neonatal hearing loss, the importance of the UNHS program and the necessity for complete follow-up. A wider and more efficient network of screening and referral centers is needed. Research studies conducted at individual institutions can help identify and address the inadequacies of the UNHS program at a grass root level.

Keywords:

Neonatal Hearing Loss, UNHS, Follow-Up Default, Saudi Arabia

Introduction:

The Center for Disease Control (CDC) describes hearing loss in children as a functional anomaly associated with any part of the ear like the inner, middle, or outer ear, the acoustic nerve or the auditory system[1]. Neonatal hearing loss is one of the most common congenital anomalies and presents a growing cause of concern. In 2019, the CDC reported the prevalence of neonatal hearing loss to be 1.7 per 1000 newborns [2]. Globally, the prevalence of moderate and severe bilateral hearing deficit (>40 dB) is estimated to be 1–3 per 1,000 live births in well-baby nursery population and 2–4 per 100 infants, in the neonatal intensive care unit (NICU) population [3]. Past evidence shows that the prevalence of sensorineural hearing loss in children in Saudi Arabia ranges between 7.7%-13% [1].

Past evidence shows that neonatal hearing loss can be attributed to several causative factors like NICU admission for ≥2 days, Usher's syndrome, Waardenburg's syndrome, other syndromes associated with hearing loss, family history of hereditary childhood sensorineural hearing loss, craniofacial abnormalities; and congenital infections such as cytomegalovirus, toxoplasmosis, bacterial meningitis, syphilis, herpes or rubella. In nearly 50% of affected infants, hearing loss is idiopathic [4]. Neonatal hearing loss can have a detrimental impact on a child's overall development by causing a significant delay or impairment of linguistic ability and communication skills. This can lead to social isolation, learning disabilities, poor academic performance; and also raises the risk of psychological complications and mental retardation [5].

Hence, timely and accurate screening of hearing function at birth and early diagnosis of neonatal hearing loss, play a crucial role in reducing the burden of this disorder. Tests like otoacoustic emission and automated auditory brainstem response (AABR) have been commonly used for screening neonatal hearing function. In order to establish a globally harmonized approach for newborn hearing screening, the Joint Committee for Infant Hearing (JCIH) has recommended the Universal Newborn Hearing Screening (UNHS) protocol, which encompasses screening all newborns for hearing disturbances, within the very first month of life. For those who fail screening, a thorough audiologic evaluation should take place within three months of age, and those with confirmed hearing loss should receive appropriate intervention not later than 6 months of age [4,6].

In Saudi Arabia, UNHS was adopted at a national level in the year 2016 and since then the coverage and referral rate of newborn hearing screening has significantly increased. It has been proven instrumental

in the early diagnosis of neonatal hearing loss and in mitigating the burden of this disorder to a significant extent. However, "loss-to-follow-up" has been identified as a key obstacle in the successful implementation of the UNHS program in Saudi Arabia. Large-scale studies in the country have stressed the need to investigate the causes of patients being lost to follow-up, during the screening stages of the UNHS protocol [2,7].

Recent evidence has also shown that the region of Madina has the highest referral rate under the country's UNHS program. Past evidence has also demonstrated alarmingly low levels of knowledge among parents in Madina, regarding hearing loss among neonates[2,7]. Hence, the current study retrospectively analyzed the prevalence and causes of neonatal hearing loss and follow-up default in the UNHS program, at a leading children's and maternity hospital, at Madina, Saudi Arabia. The study was planned with the objective of looking closely at the overall scenario of neonatal hearing loss, at an institutional level in the city of Madina.

Materials and Methods:

Study Objectives and Study Population: This study employed a retrospective, cross-sectional cohort analysis to assess the prevalence and causes of neonatal hearing loss and follow-up default (loss-to-follow-up), among neonates screened and treated at the Children and Maternity Hospital in Madina, Saudi Arabia, from January 2021 to January 2022. The study population comprised all babies born at the hospital during this period, each of whom also underwent the UNHS program at the study hospital.

Study Methods:

An assessment was conducted to determine the number of infants who underwent auditory screening, as well as the proportion of successful outcomes, referrals, and cases with default/loss of follow-up. Referral rate and loss to follow-up rate was assessed for all three stages of the newborn hearing screening program, and at the end of the screening. This evaluation was carried out utilizing the AABR test. The causative factors for neonatal hearing loss and reasons for follow-up default were documented. The Institutional Review Board (IRB116-2021) of the Faculty of Medicine approved the conduct of this research study. The ethics committee ensured that all data was anonymized and excluded the retrospective analysis of medical records from informed consent.

Key Risk Factors Included in the Study:

Neonates were considered to be at a high risk of hearing loss based on the presence of any of the following parameters: A family history of inherited sensorineural hearing loss in childhood, intrauterine Infections,

craniofacial anomalies, particularly those with pinna and ear canal morphologic abnormalities, birth weight less than 1500 g, hyperbilirubinemia necessitating exchange transfusions, ototoxic drugs, including, but not limited to, courses or combinations with loop diuretics, bacterial meningitis; appearance, pulse, grimace, activity, and respiration (APGAR) score <4 at one minute and <6 at five minutes, >5 days of mechanical ventilation needed and presence of a syndrome known to be associated with sensorineural and/or conductive hearing loss.

Statistical Analysis:

It was performed using GraphPad Prism 7 (GraphPad Software, CA, USA). Prevalence of neonatal hearing loss and follow-up default were presented using descriptive statistics (frequency and percentage). The correlation of risk factors with neonatal hearing loss was analyzed using the Chi-square test, correlation analysis and P-value computation. The threshold for statistical significance was set at $P<0.05$ for all analyses.

Results:

A total of 500 newborns with a mean age of one month were included in the study. The study sample was well balanced with regards to gender as it included 255 males (51%) and 245 females (49%). Table 1 summarizes the number of newborns who passed and failed at each stage of the hearing screening protocol as well as the percentage of newborns lost to follow-up at every stage. Out of the 500 newborns included in the study, a total of six newborns failed all three stages of screening and were referred for audiological assessment. Hence, the prevalence of neonatal hearing loss in the study was 6/500 (1.2%). With regards to follow-up default, 89 (17.8%) were lost to follow-up during the various stages of the screening protocol.

Table 1 Prevalence of Neonatal Hearing Loss and Follow-up Default Among the Study :

Stage of Screening	**Frequency of Screening Pass/ Fail N=500 (%)**
Age (Months)	1±0.55
Gender	
Female	255(51%)
Male	245(49%)
Number of newborns screened	500
First stage	
Pass	313(62.6%)
Failed and referred to stage 2	**187(37.4%)**
Patients lost to follow-up between stage 1 and stage 2	64 out of 187 (34.4%)
Second stage (N=123)	
Screened	123
Pass	81(65.86%)
Failed and referred to stage 3	**42(34.14%)**
Patients lost to follow-up between stage 2 and stage 3	21 out of 42 (50%)
Third stage (N=21)	
Pass	15(71.4%)

Failed and referred for audiological assessment	**6(28.6%)**
Patients lost to follow-up between stage 3 and Audiological assessment	2 out of 6 (34%)
Patients who underwent audiological assessment	4 out of 6 (66.6%)
Total number of newborns with neonatal hearing loss (referred for audiological assessment)	6/500 (1.2%)
The total number of newborns collectively lost to follow-up in all stages	89/500 (17.8%)

The top causes of follow-up default in the study were lack of proper transport facilities and lower educational level of parents. Poor economic status was also cited as a key reason contributing to loss of follow up. It is noteworthy that nearly 15 out of 89 newborns could not be followed up as their parents forgot to bring them for the next appointment. Table 2 summarizes the causes of follow up default in the study.

Table 2 Causes of Newborn Hearing Screening Default (Loss-to-Follow-Up) in the Study Population:

Total number of neonates with follow-up default	**N=89**
Reasons for Loss-to-Follow-Up (Default)	N (%)
Forgot to Attend	15 (16.8%)
Transport Problems	30 (33.7%)
Low Economic Status	19 (21.8%)
Education Level of parents (secondary or less)	25 (28.7%)

Table 3 lists the known risk factors for neonatal hearing loss and presents their distribution and frequency among newborns in the study. The risk factors of neonatal hearing loss were further correlated with the results of the AABR tests conducted in the study. Parental history of newborn hearing loss, craniofacial anomalies of the newborn, low birth weight, exposure to ototoxic drugs, maternal age >30 years, and admission to NICU had a statistically significant correlation with abnormal AABR test results.

Table 3 The known risk factors for neonatal hearing loss:

Factors Under Consideration for Neonatal Hearing Loss	**Frequency of Risk Factors in the Study Population**		**F value (One-way ANOVA)**	**p-value (Chi-Square)**
	Yes	**No**		
A history of hereditary sensorineural hearing loss as an infant	50	450	11.98	0.03*
Intrauterine Infections	20	480		
Craniofacial anomalies, particularly those with pinna and ear canal morphologic abnormalities.	15	485	2.19	0.07
Birth weight is less than 1500 g.	100	400	12.89	0.03*
Ototoxic drugs, including, but not limited to, courses or combinations with loop diuretics.	208	292	13.96	0.01*
Bacterial meningitis.	101	399	3.68	0.06
Sepsis	10	490	3.98	0.06
More than five days of mechanical ventilation are required.	10	490	3.76	0.06
Other findings linked to a syndrome known to be associated with sensorineural and/or conductive hearing loss.	5	495	2.19	0.07
Maternal diseases	300	200	3.49	0.06
Difficulties and complications of delivery	110	390		
Admission to a neonatal intensive care unit	55	445	4.96	0.07

Parents begin relatively	333	167	12.88	0.03*
Factors Under Consideration for Neonatal Hearing Loss	**Frequency of Risk Factors in the Study Population**		**F value (One-way ANOVA)**	**p-value (Chi-Square)**
APGAR (Appearance, Pulse, Grimace, Activity and Respiration) scores below four at one minute and below six at five minutes.				
Normal >6	433		1.17	0.07
Abnormal<6	67			
A serum level of hyperbilirubinemia necessitating exchange transfusions mg/dl				
>18=62	62		2.26	0.06
>15=338	438			
Maternal age				
<20	46		15.79	0.04*
20-30	350			
>30	104			

F value (One-way ANOVA) and P-value obtained from the Pearson Chi-Square. *P<0.05, AABR – Automated auditory brainstem response

Discussion:

Past evidence has demonstrated alarmingly low levels of knowledge regarding neonatal hearing loss among parents in Madina. A negative attitude towards screening for hearing loss has also been reported in this region [7]. Moreover, low levels of awareness regarding ear health have been reported from this region, among parents younger than 30 years of age and those with poor educational and economic backgrounds [8]. To our knowledge, we did not come across any previous study from a tertiary care children's hospital in Madina, focusing on the prevalence of neonatal hearing loss, its risk factors and follow-up default.

In the backdrop of such trends seen in Madina, we felt an unmet medical need to research the prevalence and causes of neonatal hearing loss and follow-up default, at the Children and Maternity Hospital in Madina, which is one of the leading tertiary care hospitals in the region for neonatal care. To investigate the same, we conducted a retrospective cohort study among newborns at the neonatal ENT unit of the hospital. All 500 neonates born from January 2021 to January 2022 in the hospital were screened for hearing loss and included in the study.

This retrospective cohort analysis showed the prevalence of neonatal hearing loss to be 1.2% while follow-up default or loss-to-follow-up was found to be 17.8%. The prevalence of neonatal hearing loss in our study was several folds lower than the Madina-specific prevalence of 7.72%, reported by N. Al Othman et al in their country-wide study [2]. This difference in prevalence can be most obviously attributed to the fact that N. Alothman et al had a much larger sample

size, wider demography and a much larger coverage of hospitals in the entire Madina province. We accept that a smaller sample size from a single hospital is a limitation of our study. However, as our hospital is one of the leading centers for UNHS in the city, we believe that data generated through our study will encourage and guide clinical teams from other hospitals in Madina, in their research endeavors on neonatal hearing loss. We believe this will spark a trend that can help gather better grass-root level data from individual hospitals in this region.

During 2019-20, Alanazi assessed the rate of referral in two UNHS programs from Riyadh, in a cohort of 20,171 newborns and reported the prevalence of neonatal hearing loss to be 1.33%[9]. Rehman et al assessed neonatal hearing loss in a cohort of 14860 newborns in the Eastern region of Saudi Arabia. The study included 13854 healthy babies and 826 NICU admissions. A total of 39 out of 14860 babies had confirmed hearing impairment amounting to an overall prevalence of 0.26%. The prevalence was 0.18% and 1.7% for the well-baby and NICU cohorts respectively [10].

Recently, N. Alothman et al published a country-wide analysis of neonatal hearing loss, among neonates screened under the country's UNHS program in the year 2021 alone; and reported a prevalence of 1.87% among 199,034 newborns[2]. In the year 2021, Alaql F reviewed the performance of the UNHS program of the Ministry of Health, Saudi Arabia ever since the inception of UNHS in the country and found that more than one million babies had been screened under the UNHS protocol with a referral rate of 0.7% [11]. Based on this recent evidence from large scale, country-wide studies, we can infer that under Saudi Arabia's current UNHS program, the overall prevalence of neonatal hearing loss ranges approximately between 0.7% to 1.8% [2,11]. In our study, we reported the prevalence of neonatal hearing loss to be 1.2%, which seems well within the national average shown by these larger studies.

Meta-analysis of global data has estimated the prevalence of neonatal hearing loss to range between 0.1% to 0.6%, with a mean of 0.2% [12]. The prevalence of neonatal hearing loss is notably higher in Saudi Arabia as compared to the global average, often attributed to the rampant practice of consanguineous marriages in the country, which is known to increase the risk of several congenital cochleovestibular anomalies [9]. Low awareness levels among parents regarding the risk of neonatal hearing loss is another factor that explains the higher prevalence in Saudi Arabia [13].

In this context, it seems necessary to look at the prevalence of neonatal hearing loss,

from different parts of the world as well, to sketch a better comparison between Saudi Arabia and other countries. As per latest figures put forth by the Center for Disease Control (CDC), 0.17% newborn babies suffer from hearing loss in the United States [14]. Past studies from China have shown that the prevalence ranges between 0.23% to 0.63% [15]. In a recent pilot UNHS study, conducted among 506 neonates at a secondary care hospital in India, the incidence of hearing loss was found to be 1.83% for high-risk neonates and 0.53% for healthy ones [16]. An Iranian study consisting of a large cohort of 5500 newborns reported the prevalence of hearing impairment to be 0.43% [17]. Kaplama et al retrospectively analyzed the trend of neonatal hearing loss among 6034 Syrian and 37,219 Turkish newborns. The prevalence of hearing loss was found to be 0.2% and 0.4% among Turkish and Syrian newborns respectively [18]. We believe that varying prevalence estimates from different countries of the world, including Saudi Arabia, are most likely due to differences in socio-cultural practices, medical awareness, neonatal facilities, and different demographic patterns.

The most prominent risk factor in our study was the use of ototoxic drugs, especially loop diuretics, which showed the most robust statistical correlation with abnormal AABR test results. This was followed by low birth weight (<1500 g) which was found to be the second most important risk factor. Congenital craniofacial anomalies, mainly those involving the pinna and ear canal, and admission to the NICU also significantly impacted the risk of neonatal hearing loss. Maternal age >30 years and a positive hereditary history of neonatal hearing loss were also significant predisposing factors in our study. Other risk factors like intrauterine and perinatal infections, low APGAR score, hyperbilirubinemia, neonatal jaundice and the need for mechanical ventilation of the newborn, did correlate positively with the risk of neonatal hearing loss; however, the correlation was not statistically significant.

The profile of risk factors in our study seems in line with past evidence which has shown that positive familial history, low birth weight, craniofacial anomalies, exposure to ototoxic drugs, maternal age, maternal infections, perinatal infections, NICU admission, APGAR score <5, neonatal icterus and hyperbilirubinemia are frequently reported risk factors of neonatal hearing loss[19-23].

The overall prevalence of lost-to-follow-up or follow-up default in our study was 17.8%. Previous studies from Saudi Arabia have reported varying rates of follow-up default. In a study conducted at a tertiary care hospital in 2020-2022, N. Alothman et al

reported a follow-up default rate of 18%, in a cohort of 2312 Saudi newborns, which is almost equal to the rate of follow-up default reported in our study [24]. Almatrafi et al reported a higher lost-to follow-up rate of 24.14%, among a group of 1533 parents of infants / newborns in Saudi Arabia, in a questionnaire based cross-sectional survey done in 2019 [25]. Among recent studies conducted in Saudi Arabia, the highest rate of lost-to follow-up has been shown by Alanazi. In the study conducted by Alanazi in two major hospitals in Riyadh, the prevalence of follow-up default was at a staggering high of 34.9%, in a cohort of 20171 newborns, almost double the prevalence reported in our study [9]. The rate of follow up default in this study by Alanazi was significantly higher because it considered newborns lost to the system (failed the screening but never returned for referral) as well as the ones lost to documentation (newborns and infants whose screening status or final diagnosis has not been documented and updated into the system and remains unknown). In our opinion, if lost to documentation was not included by Alanazi, perhaps the prevalence of lost to follow up alone would have been much lower than 34.9% [9].

Overall, in light of previous studies from Saudi Arabia and based on the outcomes of the current study reported by us, we believe that the average rate of follow-up default in neonatal hearing screening in the country ranges approximately between 18% to 25% [24, 25]. However, we also concur with the recommendation of N. Alothman et al that a large-scale, country-wide assessment must be conducted to evaluate the rate of follow-up default in Saudi Arabia's UNHS program, to accurately determine the magnitude and etiology of such default [24].

It is noteworthy that the most frequent cause of follow-up default in our study was difficulty in transportation and commuting to the referral or screening center, reported by 33.7% of parents. Though transportation problems have been cited previously as one of the causative factors for follow-up default, but were never reported as the leading cause, to the best of our knowledge[24]. Hence, relevant authorities need to urgently look into providing better transportation facilities to parents of newborns being screened under the UNHS program.

As expected, nearly 29% of parents of newborns in our study had low awareness and poor knowledge about the significance of following up till the end of the UNHS protocol. This is in congruence with the findings of previous studies from Saudi Arabia that have shown poor awareness among parents as a key cause for follow-up default [9, 24]. We believe such a lack of awareness is attributable to lower educational levels

among parents and a lack of clear communication or reminders from healthcare staff regarding the importance of follow-up. Also, economically weaker parents often have a lower educational status and limited resources, which in turn, leads to poor awareness levels and low commitment towards follow-up. Poor economic status was reported by 21.8% of parents as the reason for loss-to-follow-up.

A sub-group of 16.8% of parents cited forgetting the appointment, as their reason for follow-up default. Previous studies have also reported "forgot to attend" as one of the reasons for follow-up default [24]. We believe that parents tend to forget these follow-up appointments because they do not realize the importance of follow-up or haven't been well informed on the potential adverse fallouts of missed follow-up under the UNHS protocol. This circles back to the underlying problem of poor awareness and knowledge among parents, which has been cited previously as the key cause of follow-up default in UNHS programs in Saudi Arabia [9].

We opine that large-scale public education initiatives are needed in Saudi Arabia to raise awareness regarding the importance of neonatal hearing screening and its follow-up. A concerted effort from the government, administration, hospital authorities and clinicians can go a long way in making parents well aware of the disastrous effects of neonatal hearing loss and the benefits of the UNHS program. Awareness initiatives should be mandatorily conducted at all possible hospitals and social platforms, to ensure maximum public access and participation. In today's era, the power of social media can be harnessed mostly in raising awareness on neonatal hearing loss and the UNHS program. Widespread digital campaigns can help take the message to a much larger audience of parents, guardians and caregivers. The need of the hour is to build a better infrastructure which ensures screening and referral centers are easily available, accessible, affordable and well-equipped. We recommend clinical colleagues and researchers from other UNHS centers in the Madina region, to deep dive into the prevalence and causes of neonatal hearing loss and follow-up default in their respective institutions. This will provide a grass-root level view of the situation at an institutional level. This, in turn, will help address the inadequacies in the UNHS program at each individual center and exercise evidence-backed remedial measures.

Conclusion:

The prevalence and causes of neonatal hearing loss and follow-up default in the study were in congruence with previous data reported by larger studies from Saudi

Arabia. The study reiterates the dire need to raise awareness among parents of newborns regarding the detrimental effects of neonatal hearing loss, the importance of the UNHS program and the necessity to follow-up till the end. Mass awareness campaigns with large scale public participation are needed to educate parents on this sensitive issue. Infrastructural improvement in terms of a wider and more efficient network of screening and referral centers is a huge unmet need. Research studies conducted at individual institutions can help identify and address the inadequacies of the UNHS program at a grassroots level.

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Hanan Almuzaini – Writing editing and final review.

Khaled Shahat, Sara Ibrahim Almuzaini, Badr Eyad Hafiz, Nora Tarq Alreefi, Reham Khaled Hawalh

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All authors declare that they have no competing interests.

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The data that support the findings of this study are available from the corresponding author upon reasonable request.

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