J Exp Pathol. 2024;5(1):6-13.

Journal of Experimental Pathology Research Article

Retesting of Neonatal Hearing Screening Before and During the COVID-19 Pandemic: A Longitudinal Study

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Received date: February 01, 2024, **Accepted date:** March 08, 2024

Citation: Tiezerin CS, de Paiva KM, Woide LK, Cigana LB, Machado MJ, Haas P. Retesting of Neonatal Hearing Screening Before and During the COVID-19 Pandemic: A Longitudinal Study. J Exp Pathol. 2024;5(1):6-13.

Abstract

Introduction: Universal Newborn Hearing Screening (UNHS) plays an essential role in the early identification of hearing loss in neonates. Risk factors for hearing impairment may include family history, prematurity, and exposure to ototoxic substances. Coronavirus Disease 2019 (COVID-19) might be a significant contributing factor affecting the structures of the inner ear. **Objective**: To assess the auditory follow-up process of retesting for UNHS before and during the COVID-19 pandemic among neonates from an Outpatient Auditory Health Service (SASA) in the state of Santa Catarina with SUS (Unified Health System) assistance. **Methods**: A retrospective longitudinal study analyzed data from neonates attended at a SUS Auditory Health Service (SASA) from January 2018 to December 2022. Information related to UNHS and retest outcomes was assessed. Data were analyzed using Microsoft Excel® and MedCalc® Statistical Software version 22.006, utilizing statistical measures and regression analyses to identify factors associated with UNHS failures and retesting. **Results**: A failure to retest rate of 2.6% in the right ear and 2.2% in the left ear was observed among evaluated neonates. The average age of mothers of neonates who did not pass the test was 33 years, while the overall average was 27 years. Failure to pass the retest and a longer interval between UNHS and retesting were associated with UNHS Initial Retest Default (IRD). There was an increase in dropout rates for UNHS retesting, and the time interval between UNHS and retesting was extended during the pandemic. **Conclusion**: Several factors, including the interval between tests, mothers' age, and medical conditions, influenced the retest outcomes. The pandemic led to a significant increase in dropout rates and extended time for retesting.

Keywords: Neonatal screening, Newborn, Hearing, Auditory health, National Policy for Hearing Health Care

Background

COVID-19 occurred globally in late 2019, a period during which the World Health Organization and government agencies declared a global health emergency [1]. In Brazil, each region implemented measures to contain the spread of the virus, aligning with the established guidelines. According to information from the Ministry of Health, pregnant women undergo physiological changes that make them more susceptible to respiratory complications and other conditions. Clinical manifestations in pregnant women diagnosed with COVID-19 ranged from symptomatic to asymptomatic, similar to the general population, and in some cases, required hospitalization in intensive care units (ICU) [2].

Considering that congenital infections represent risk factors dependent on the relationship between the placenta and the fetus, any placental injury can result in the transmission of congenital infections such as syphilis, toxoplasmosis, human immunodeficiency virus (HIV), chickenpox, rubella, herpes, and cytomegalovirus [3]. These infections can cause direct damage to the structures of the fetal inner ear or trigger immune-mediated damage in the host. Therefore, COVID-19, being a viral disease, can also impact the structures of the inner ear, and considering that the examinations conducted to assess the functioning of the auditory structures are primarily

located in the inner ear, it is understood that COVID-19 infection can lead to failures both in the initial screening and in the retesting [4].

The first sense to be awakened in intrauterine life is hearing. Sounds produced by the pregnant woman and external sounds are auditory stimuli that lead the fetus to experience sensations that increase interest in sound immediately after birth. The lack of stimulation in the auditory system may lead to the development of speech and language disorders, learning disorders, and social issues in newborns. Early auditory intervention is suggested for these cases [5-7].

It is estimated that by 2050, approximately 2.5 billion people will have hearing impairments, with 1 in 4 individuals experiencing some degree of hearing loss, and at least 700 million will require access to healthcare and auditory rehabilitation services. Moreover, 60% of hearing losses in newborns can be prevented by expanding and improving screening for neonates with risk factors for auditory diseases during UNHS conducted in maternity hospitals after birth [8]. UNHS is the initial step for identifying childhood hearing impairments. From its screening program, if a newborn fails the test or presents any Indicators of Risk for Hearing Impairment (IRHI), they are referred for retesting and/or monitoring and appropriate referrals to other necessary levels of healthcare [9].

It is crucial that the diagnosis of hearing impairment be conducted within the first month of life. Therefore, it is relevant to identify IRHI as early as possible. Some IRHI can be identified during pregnancy, highlighting the importance of providing guidance during prenatal consultations. This guidance is of great importance for early auditory diagnosis, as families need to be informed about the significance of UNHS and the auditory development of the newborn. However, some research indicates a high rate of families' lack of awareness regarding the importance of UNHS [11-14].

According to research, the most common IRHI include a family history of permanent deafness, stay in the Neonatal Intensive Care Unit (NICU), ototoxic medications, mechanical ventilation, low birth weight, prematurity, congenital infections, genetic syndromes associated with hearing impairment, Apgar score of 0 to 4 at 1 minute or 0 to 6 at 5 minutes, as well as alcoholism or drug use during pregnancy [11,15]. According to national and international guidelines, the flowchart of UNHS dictates that newborns without IRHI who failed the initial screening should undergo retesting, as well as newborns who passed or failed UNHS and have IRHI [16]. The attendance rate for retesting is one of the quality indicators of a UNHS program, with an expected attendance rate of 90% for newborns referred for retesting [17].

From the aforementioned information, it emphasizes the importance of preventive measures and early childhood auditory diagnostic assistance, as they are crucial in primary health care to remain updated and standardized in collecting data. This allows for the analysis of care, detected results, and leads to potential new public health policies for childhood auditory health. Following this guidance, this research aimed to verify the auditory follow-up process of retesting for UNHS before and during the COVID-19 pandemic among neonates from an Outpatient Auditory Health Service (OHS) in the state of Santa Catarina, Brazil, with SUS assistance.

Materials and Methods

Study design and location

This study constitutes retrospective longitudinal research involving the secondary analysis of data recorded in the database of neonates attended at a reference center in Auditory Health within the scope of the SUS from January 2018 to December 2022. The inclusion criteria encompassed neonates who underwent UNHS and were properly registered in the mentioned database, regardless of the presence or absence of risk indicators for hearing impairment, as long as they were referred to the OHS. Exclusion criteria were established to remove neonates who had satisfactory results in UNHS and did not present risk factors for hearing loss from the study. Neonates with incomplete information that was essential for the research were also excluded.

Data collection

The data collection was conducted by analyzing the available information in the database of the Auditory Health Service, a reference center in Santa Catarina. This service plays a crucial role in performing UNHS in a referenced Auditory Health Service that serves two large public maternity hospitals. It oversees retests and follows the protocol established by the Care Network for Persons with Disabilities in Santa Catarina [18].

Information was gathered encompassing prenatal, childbirth, and postpartum data, sociodemographic characteristics of the mother and newborn (including sex and age), results of Transient Evoked Otoacoustic Emissions (TEOAE) and/or Auditory Brainstem Response (ABR) tests classified as "PASS" or "FAIL," as well as the identification of IRHI. For newborns who did not pass the UNHS, information regarding attendance or non-attendance for retesting and the related care procedures involved in this process were also collected.

Data analysis

The data were organized into spreadsheets using Microsoft Excel® and subsequently exported for analysis using the MedCalc® Statistical Software version 22.006. For the analysis of quantitative (numeric) data, descriptive statistics were employed, providing summary measures (position and

variability), such as mean, median, minimum, maximum, and standard deviation. Relevant statistical graphs were utilized to ensure adequate visualization of the obtained results. Descriptive statistics with single and double entry tabulations were used to describe categorical variables, showing absolute and relative frequencies relevant to the study's objectives.

Additionally, regression analyses were conducted to identify factors associated with retesting failures and to analyze the non-attendance rate for retesting, as well as the time taken for retesting. These analyses were crucial to guide proposals for improving auditory health promotion in Santa Catarina.

Ethical aspects

This research was approved by the Research Ethics Committee (REC) of the Federal University of Santa Catarina, under protocol number CAAE: 39562720.8.0000.0121.

Results

Table 1 presents a comprehensive summary of the distribution of retest examinations conducted. Two types of examinations, OAE (Otoacoustic Emissions) and ABR (Auditory Brainstem Response), were offered to the evaluated 1,206 neonates (100%). Among these, 118 (9.79%) underwent OAE, while 725 (60.14%) underwent ABR, and 363 (30.07%) did not attend the retest. The table also displays the results of the examinations by ear, combining the data from both types of examinations. Accordingly, 811 neonates (67.30%) obtained satisfactory results in the right ear, whereas 816 (67.7%) achieved satisfactory results in the left ear. Regarding cases of failure, 32 neonates (2.6%) presented failures in the right ear, while 27 (2.2%) showed failures in the left ear.

Based on the data presented in **Table 2**, several factors showed significant associations with the likelihood of failure

Table 1. Results of Retesting UNHS Examinations and Distribution of Results by Retested Ear (2018-2022).

Variables	**Distribution Total for Retesting UNHS**					
Exams	**n (Total)**	**(%)**			**P***	**P****
OAE	118	9.79%			**< 0.0001**	**< 0.0001**
ABR	725	60.14%				
No show	363	30.07%				
Total	**1206**	**100.0%**				
Results	**Right ear**		**Left ear**		**P***	**P****
	n (Total)	(%)	n (Total)	(%)		
Pass	811	67.30%	816	67.70%	< 0.0001	< 0.0001
Fail	32	2.60%	27	2.20%		
Not done	363	30.10%	363	30.10%		
Total	**1206**	**100.0%**	**1206**	**100.0%**		

P*: P-value (Chi-square test); P**: P-value (Fisher's exact test); OAE: Evoked Otoacoustic Emissions; ABR: Auditory Brainstem Response

Table 2. Neonatal Health Factors and Probability of Failure in UNHS Retesting (2018-2022).

Variable	**Gross OR***	**CI95%**	***P-value***	**Adjusted OR**	**CI95%**	***P-value***
Use of mechanical ventilation						
No	1			1		
Yes	3.66	1.3399 - 9.9975	0.0114	5.1903	1.6487-16.3397	0.0049
No prenatal follow-up						
No	1			1		
Yes	10.8514	0.9620 - 122.4015	0.0538	59.6179	3.2588-1090.6650	0.0058
Malformation						
No	1			1		

Yes	24.9688	7.6303 - 81.7052	<0.0001	30.4692	8.1816-113.4704	<0.0001
Neurological disturbances						
No	1			1		
Yes	14.8519	2.4061 - 91.6745	0.0037	12.2323	1.6792-89.1082	0.0135
Trisomy 21						
No	1			1		
Yes	5.4122	0.5902 - 49.6323	0.1353	9.937	0.9796-100.8028	0.0521
Admission to NICU						
No	1			1		
Yes	1.6533	0.7854 - 3.4803	0.1855	2.0713	0.8063-5.3208	0.1303
Jaundice						
No	1			1		
Yes	4.0101	0.8569 - 18.7662	0.0778	4.1374	0.6816-25.1132	0.1227
Family History of Hearing Loss						
No	1			1		
Yes	1.9509	0.2453 - 15.5148	0.5276	3.2307	0.3832-27.2351	0.2809
Prematurity						
No	1			1		
Yes	0.704	0.3550 - 1.3960	0.3149	0.5332	0.2263-1.2563	0.1504
Age of the mother						
x	1			1		
x + 1 year	nd	nd	nd	0.9919	0.9421-1.0444	0.7577

*= OR, unadjusted Odds Ratio; P value Z-test to unadjusted Odds Ratio;

aOR, odds ratio adjusted for the independent variables included in the models (Maternal age, Mechanical ventilation use, Did not undergo prenatal testing, malformation, neurological disorders, trisomy 21, ICU admission, family history of hearing loss, prematurity).

Model – Dependent Variable = falha na UNHS; *Method Enter, Overall Model Fit, Significance level P<0,00001 Constant -3,17538; Hosmer & Lemeshow test, Significance level P = 0,5015; Area under the ROC curve (AUC) = 0,751 (95%CI 0,719-0,782)

in the UNHS retesting. Notably, the use of mechanical ventilation emerged, revealing a substantially increased likelihood of failure for neonates undergoing this intervention. Additionally, the presence of congenital malformations and the manifestation of neurological disorders were also strongly associated with retesting failure. NICU admission and the occurrence of neonatal jaundice also demonstrated statistically significant associations with retesting failure.

The results below refer to **Table 3**, which demonstrates the quantity of IRHI present in 824 neonates who attended the UNHS Retest. Most neonates (45.63%) showed one IRHI, while 17.96% did not have any related IRHI. Additionally, 20.00% exhibited two IRHI, 11.77% had three IRHI, 3.16% recorded four IRHI, 1.09% presented five IRHI, and only 0.36% had six IRHI.

Table 4 shows the analysis of the maternal age variables in years and the retest results of the UNHS. Among the n=718 neonates who attended the retest, the median age of mothers whose neonates passed the retest was 28 years. In contrast, the 36 mothers who took their children to the retest and whose children failed the exam had a higher median age of 33 years.

Table 5 describes a total of 1,206 neonates who were scheduled for the UNHS retest. In the years before the pandemic, 2018 and 2019, 4.40% of the neonates were referred for the retest, with 0.70% not attending in 2018. In 2019, 35.10% of the babies were referred for the retest, with a non-attendance rate of 9.80%. However, during the pandemic years, in 2020, 24.90% were referred for the retest, with a non-attendance rate of 9.80%. In 2021, 17.00% of the babies were

Table 3. Quantity of IRHI present in the neonates who attended the UNHS Retest, Florianópolis, SC (2018-2022).

Variable	Number of IRHI present in neonates.			P*
	n	%	IQR	
None	148	17.96%	18.5-38.5	0.001437
One	376	45.63%	19.0-67.0	
Two	165	20.00%	21.8-103.5	
Three	97	11.77%	16.5-84.3	
Four	26	03.16%	81.0-206.0	
Five	9	01.09%	31.5-97.75	
Six	3	0.36%	25.25-111.50	
Total	**824**	**100%**		

IRHI: Indicators of Risk for Hearing Loss. P*: P value to Kruskal-Wallis test.

[1,2,3,4,5,6] P <0.05 compared to row number (Post-hoc analysis for pairwise comparison of subgroups If the Kruskal-Wallis test is positive -P<0.05).

Table 4. Analysis of the Influence of Maternal Age on the Results of the UNHS Retest, Florianópolis, SC (2018-2022).

Variable	Passes in the Retest			Fails the Retest			P**
	n	Median	IQR	n	Median	IQR	
Mother's Age (years)	718	28	23 -34	36	33	18-71	0.9556

P**: P values obtained to comparison of medians UNHS (Mann-Whithney test); UNHS: Universal Newborn Hearing Screening

Table 5. Distribution of Neonates by Year and Attendance at the UNHS Retest Before and During the Pandemic, Florianópolis, SC (2018-2022).

Variable	Forwarded to UNHS Retest						P*	P**
	Total		Did not return		Returned			
	n (Total)	(%)	n (No)	(%)	n (Yes)	(%)		
Year								
2018	53	4.40%	8	0.70%	45	3.70%	0.0013	ND
2019	423	35.10%	118	9.80%	305	25.30%		
2020	300	24.90%	104	8.60%	196	16.30%		
2021	205	17.00%	77	6.40%	128	10.60%		
2022	225	18.70%	56	4.60%	169	14.00%		
Total	**1 206**	**100.0%**	**363**	**30.10%**	**843**	**69.90%**		

P*: P-value (Chi-square test); P**: P-value (Fisher's exact test). MCD: Carmela Dutra Maternity, HRSJ: Regional Hospital of São José.

referred, and 6.40% did not attend. In the post-pandemic period, in 2022, 18.70% were referred, with a non-attendance rate of 4.60%. From the total sample of 1,206 neonates, there was an evasion rate of 30.10%, with the pandemic period registering the highest evasion rate.

Table 6 shows the time interval between the initial UNHS and the retest before the pandemic, with an average of 25 days in 2018 and 35 days in 2019. During the pandemic period, this average increased, being 40 days in 2020 and 44 days in 2021. After the pandemic, there was a decrease in this average, recording 37 days in 2022.

Table 6. Time for UNHS Retesting per Year Before and After the Pandemic, Florianópolis, SC (2018-2022).

Variable	The time between initial UNHS and retest for neonates who attended (days).			P*
	n	Median	IQR	
Year				
2018	148	25[2,3 e 4]	18.5-38.5	0,001578
2019	376	35 [1]	19.0-67.0	
2020	165	40 [1]	21.8-103.5	
2021	97	44 [1]	16.5-84.3	
2022	38	37	8.0-85.0	
Total	**824**			

IRHI: Indicators of Risk for Hearing Loss; [P*]: P value to Kruskal-Wallis test

Discussion

The rate of detection of hearing impairment in this research is relatively low, indicating that the majority of neonates have hearing within normal limits (2.6% failed in the right ear, and 2.2% failed in the left ear). These data are satisfactory and suggest that the initial screening conducted in neonates might be effective in early identification of hearing problems, noting that there were more failures in the right ear, but without statistical significance. According to Marinho et al. [19], there is no consensus on the predominance of ears in failures in neonatal hearing screening.

Furthermore, the statistically significant association ($p < 0.0001$) between the OAE and ABR examination types, as described in **Table 1**, and the obtained results indicates that the screening procedures are effective. This statistical validity strengthens confidence in the study's results and suggests that the screening method is robust and reliable. Presenting a detailed analysis of the influence of maternal age on the retest result, the study reveals significant observations about the median age of mothers whose neonates passed the retest (28 years), while mothers whose neonates failed the retest had a higher median age (33 years). This difference in maternal age suggests that neonates from older maternal age groups may be more likely to fail the UNHS retest, as corroborated by Cheung et al.'s study [20]. The research does not contain data regarding whether mothers contracted COVID-19 postpartum, nor whether neonates were infected subsequent to the initial screening.

Different factors have shown significant associations with the likelihood of failure in the UNHS retest. Notably, the use of mechanical ventilation emerged as a critical factor associated with an increased probability of failure in neonates undergoing this intervention. Additionally, the presence of birth malformations and the manifestation of neurological disorders also showed strong associations with retest failure, corroborating the findings in Keihanidost et al.'s study [21]. Hospitalization in the NICU and the manifestation of neonatal jaundice also exhibited statistically relevant correlations with retest failure, as indicated by Werkineh et al. [22].

Highlights a significant concern related to neonatal evasion from the UNHS retest. It is observed that, in the pre-pandemic period (2018 and 2019), the referral rate for retesting considerably increased from 4.40% to 35.10%. However, simultaneously, the non-attendance rate also increased from 0.70% to 9.80%. This may indicate greater awareness about the importance of retesting but also emphasizes the need to address factors contributing to non-attendance.

During the pandemic period (2020 and 2021), although the referral rate for retesting was lower compared to the pre-pandemic period, the non-attendance rate remained notably high. The results suggest that even in a scenario of reduced referrals, evasion continues to be a relevant concern. In the post-pandemic period (2022), the referral rate increased while the non-attendance rate decreased. This might indicate a positive trend in awareness and neonatal attendance for retesting following the end of the pandemic period.

Before the pandemic, the average return time was 25 days in 2018 and 35 days in 2019. During the pandemic, there was a significant increase in this interval, with averages of 40 days in 2020 and 44 days in 2021. In the post-pandemic period (2022), the average return time decreased to 37 days. These data suggest that the pandemic directly impacted the delay in neonates returning for retesting, possibly due to health-related restrictions and safety measures, corroborating with Besen et al. [23]. However, it is encouraging to note that after the end of the pandemic, there was a decrease in the average time interval, which may indicate a gradual recovery towards the normalcy of screening processes, and this fact might be considered a limitation of the study.

Conclusion

The results of this research indicate that the statistically

significant association between the types of exams and the obtained results validates the reliability of the screening procedures, strengthening confidence in the study's outcomes. However, the influence of maternal age and other factors such as the use of mechanical ventilation, birth malformations, neurological disorders, NICU hospitalization, and neonatal jaundice demonstrates the complexity of factors that can affect the auditory screening results. These findings highlight the need for a more comprehensive approach in neonatal auditory screening, considering various risk factors like maternal age, for more targeted intervention.

Regarding the time interval between the initial UNHS and retesting during the pandemic variations, there were significant delays in the return of neonates. However, after the end of the pandemic, there was a gradual recovery towards the normalization of screening processes. These findings underscore the importance of awareness, targeted strategies, and continuity of screening services to ensure healthy hearing development in newborns, especially in challenging situations like a pandemic.

Data Availability Statement

The authors declare the availability of Data Availability Statement.

Acknowledgments

The authors would like to thank Instituto Otovida for supporting this work.

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