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Influence of risk indicators on different Universal Newborn Hearing Screening steps

Influência dos indicadores de risco nas diferentes etapas da Triagem Auditiva Neonatal

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ABSTRACT

Purpose: To determine risk indicators for hearing loss affecting different evaluation steps of a universal newborn hearing screening program. **Methods:** Longitudinal, retrospective study of newborn hearing screening in a tertiary public hospital, including 832 newborns born between January and December 2012. Transient evoked otoacoustic emissions were measured at the first hearing evaluation in all newborns and retested in cases of "failure". When a "failure" result persisted, auditory brainstem responses (ABR) was performed. All newborns with risk indicator for hearing loss were evaluated with ABR screening regardless of the outcome of the otoacoustic emissions test. **Results:** The presence of at least one risk indicator for hearing loss, associated or not with craniofacial malformations, genetic syndromes and birth weight below 1500 g significantly increased the chances of "failure" in the otoacoustic emissions test. Meningitis and craniofacial malformations significantly increased the odds of an abnormal ABR. Two newborns with normal otoacoustic emissions were diagnosed with auditory neuropathy. **Conclusion:** Craniofacial malformation was an indicator strongly associated with a diagnosis of deafness, regardless of the hearing screening being performed by otoacoustic emissions or ABR at different steps of a universal newborn hearing screening program. This finding justifies continuous and systematic monitoring of the screening service seeking quality improvement of the newborn health hearing program.

Keywords: Neonatal screening; Risk index; Hearing; Electrophysiology; Infant, Newborn

RESUMO

Objetivo: Determinar os indicadores de risco para deficiência auditiva, que afetam as diferentes etapas de avaliação de um programa de triagem auditiva neonatal universal. **Métodos:** Estudo retrospectivo longitudinal de triagem auditiva neonatal, realizado em 832 neonatos nascidos em hospital público terciário, no período de janeiro a dezembro de 2012. O exame de Emissões Otoacústicas Evocadas por Estímulo Transiente foi realizado na primeira avaliação auditiva de todos os neonatos. Nos casos de "falha" na primeira avaliação, foi aplicado um reteste e, quando houve a permanência da "falha", foi realizado o Potencial Evocado Auditivo de Tronco Encefálico (PEATE). Os neonatos com indicador de risco para deficiência auditiva realizaram o PEATE independente do resultado das emissões otoacústicas. **Resultados:** A presença de, ao menos, um indicador de risco para deficiência auditiva, associado ou não a malformações craniofaciais, síndromes genéticas e peso menor que 1500 g ao nascimento, aumentaram significativamente as chances de "falha" na avaliação por emissões otoacústicas. Meningite e malformações craniofaciais aumentaram de maneira expressiva as chances de PEATE alterado. Dois neonatos com emissões otoacústicas normais apresentaram diagnóstico de neuropatia auditiva. **Conclusão:** A malformação craniofacial é um indicador fortemente associado ao diagnóstico da surdez, independente de a triagem auditiva ter sido realizada por emissões otoacústicas ou por PEATE em suas diferentes etapas, o que justifica o monitoramento contínuo e sistemático do serviço de triagem, na busca da melhoria da qualidade do programa de saúde auditiva do neonato.

Descritores: Triagem neonatal; Indicador de risco; Audição; Eletrofisiologia; Recém-nascido

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Conflict of interests: No

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INTRODUCTION

The universal newborn hearing screening is an effective process to identify hearing loss in newborns[1]. Identification of the hearing loss and early intervention (up to the age of 6 months) allow the best development of the child's linguistic abilities when compared with a late diagnosis of hearing loss[2,3].

The newborn hearing screening is commonly performed with otoacoustic emissions (OAEs) and auditory brainstem response (ABR) techniques. Depending on the protocol, each technique may be used alone or in sequence[4,5].

OAEs show direct evidence of the cochlear active mechanism, and their absence is related, in most cases, with sensorineural hearing loss[5]. ABR, in turn, indicate cochlear integrity, since the generation of neural responses requires cochlear functionality. ABR screening also includes electrophysiological measurement of the VIII nerve and low brainstem auditory function[5,6,7].

If on the one hand, OAEs testing is considered of easy evaluation, fast applicability, and low financial cost, on the other hand, ABR testing is less affected by noise and transient middle ear diseases, which decrease considerably the false-positive rates associated with this method while allowing detection of auditory neuropathy[6,7,8,9,10].

Therefore, ABR testing is recommended when the newborn has risk indicators for hearing loss and in cases in which the OAEs screening fails[11,12,13]. The simple occurrence of risk indicators for hearing loss increases the number of abnormalities in both tests[14]. However, there are still doubts about which risk indicators for hearing loss affect each of these techniques, and whether they are the same in different protocols[15].

The objective of this study, therefore, was to determine the risk indicators for hearing loss affecting the different evaluation steps of a universal program of newborn hearing screening.

METHODS

This was a longitudinal, retrospective study performed in a high-risk pregnancy referral center at a tertiary public hospital. The study was approved by the Research Ethics Committee at *Universidade Estadual Paulista "Julio de Mesquita Filho"* (UNESP), process no. 3395/09. The data were collected from January to December 2012 at the same hospital where the study was conducted.

The inclusion criteria were: newborns with hearing screening performed at the site of the study, and a free and informed consent form signed by their parents or guardians.

The sample comprised 832 newborns and was stratified according to the three steps of the hearing screening protocol:

Step 1: testing of all newborns with transient evoked otoacoustic emissions (TEOAEs).

Step 2: retesting of the newborns who had failed step 1 within 30 days with TEOAEs.

Step 3: ABR performed in two situations:

- When a newborn with any risk indicator for hearing loss[4,16] presented the result "pass" at both steps 1 and 2. The record was only performed at the intensity of 80 dBNPS, to verify the integrity of the auditory pathway;
- When a newborn had failed in both previous steps. The minimum level of response was verified, and an audiological evaluation was performed.

Technical specifications of the TEOAEs test

The test was performed in newborns younger than 48 hours during natural sleep on their mothers' laps, using the equipment OtoRead® (Interacoustics). The criteria for a "pass/failure" result was adapted from the criteria described in the equipment's manufacturer protocol, in which a "pass" was only given when the signal/noise ratio was at least 6 dB in at least three consecutive frequency bands, with a mandatory occurrence at 4000 Hz.

Technical specifications of the ABR test

The ABR test was performed with the equipment EP15 – Eclipse® (Interacoustics, Denmark) in a quiet environment, with the newborn comfortably accommodated on his or her mother's lap during natural sleep. After cleaning the skin with an abrasive substance (Nuprep®), we placed Neuroline® surface electrodes at specific points. The active electrode was placed on the forehead (Fz), reference electrodes were placed on mastoid areas (M1 and M2), and the ground electrode was placed on the forehead. The stimulus was presented via ER-3A insert earphones using monaural stimulation with filtered clicks (high-pass/band-pass filters of 100 Hz and low-pass filter of 2000 Hz), duration of 100 μs, and rarefaction polarity. A total of 1024 clicks were presented and analyzed during 20 ms, and the procedure was repeated to confirm the reproducibility of the waves. The impedance of the electrodes was maintained constantly below 5 kohms. The rate of stimulus presentation was 20.1 clicks per second.

Abnormal ABR results were categorized as "mildly abnormal" (thresholds between 36–50 dBnHL), "moderately abnormal" (thresholds between 51–70 dBnHL), "severely abnormal" (thresholds between 71–90 dBnHL), and "profoundly abnormal" (thresholds > 90 dBnHL)[17].

Statistical analysis

To verify the chances of "failure" in the OAEs screening and abnormalities in the ABR testing based on risk indicators, we conducted a multivariate logistic regression with forward selection procedures. The associations were considered significant if $p<0.05$. The analyses were carried out with the software SPSS, version 15.0.

RESULTS

Characterization of the cohort

A total of 832 newborns participated in the study. There were 144 newborns (17%) with at least one risk indicator for hearing loss, of which the most frequent were: an ICU stay longer than 5 days, low Apgar score at 1 and 5 minutes after birth, and use of mechanical ventilation. Most newborns (54%) presented only one risk indicator and, at the most, six associated indicators (Figure 1).

The number of newborns evaluated decreased at each step due to failure to show up for the next step (Figure 2).

Step 1 - TEOAEs test

Out of the 832 newborns evaluated, 89 had a failure in at least one ear, of whom 31 had risk indicators of hearing loss (Figure 2 - Step 1).

Step 2 - TEOAEs retest

Of 89 newborns referred to the retest, 22 (25%) were lost to follow-up. Among the 67 remaining patients, 31 (46%) remained with a "failure" result in at least one ear, of whom 18 had risk indicators (Figure 2 - Step 2).

Step 3 - ABR

A total of 117 newborns underwent ABR screening, excluding 39 who were lost to follow-up (Figure 2 - Step 3).

Fourteen newborns (22%) had a "failure" result in at least one ear, of whom 11 had risk indicators (Figure 2 - Step 3). Among these 14, only two had suggestive retrocochlear abnormalities (one newborn with unilateral abnormality and another with bilateral abnormalities).

Of the remaining 12 newborns, seven presented unilateral abnormality, resulting, therefore, in 17 ears with cochlear abnormalities (Table 1).

Indicators of risk X "failure" on the TEOAEs test

The presence of any risk indicator for hearing loss increased the chance of "failure" in the TEOAEs at steps 1 and 2, especially among newborns with genetic syndromes, craniofacial malformations, and birth weight below 1500 g (Table 2).

Risk indicators X abnormal ABR

The percentage of abnormalities in the ABR test was significantly greater in newborns who had meningitis and craniofacial malformation (Table 3).

DISCUSSION

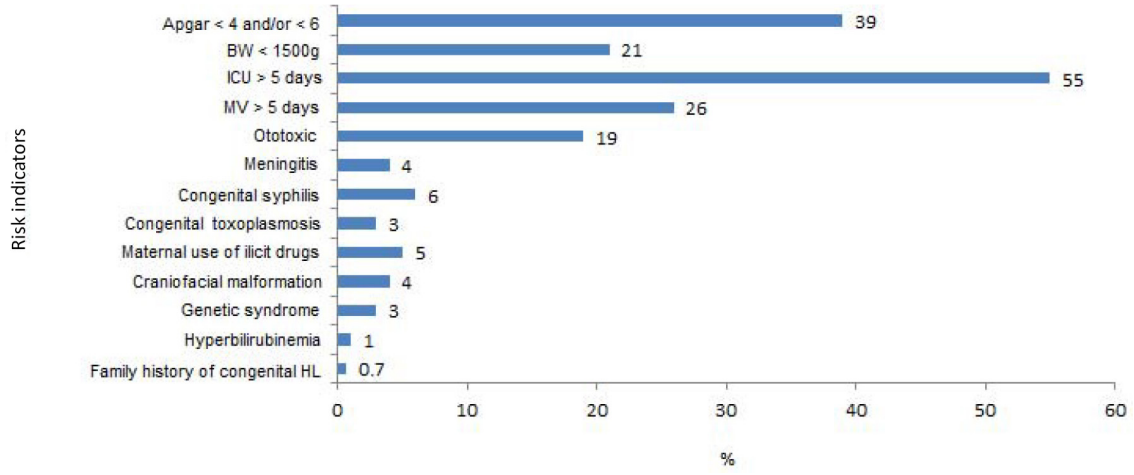
This study was conducted in a tertiary referral public hospital. The hearing screening service at this hospital serves the demand of the hospital itself in addition to that of other institutions, some not sufficiently structured to perform the TEOAES and ABR procedures.

TEOAEs testing was chosen as the initial evaluation due to its objectivity, speed, low cost, and easy implementation, as well as for being sensitive and reproducible, and allowing correct referral to subsequent evaluations[10].

On the other hand, once the hearing screening method chosen for this study could not identify retrocochlear abnormalities, all newborns with risk indicators for hearing loss were referred to ABR screening, regardless of the result of the OAEs evaluation.

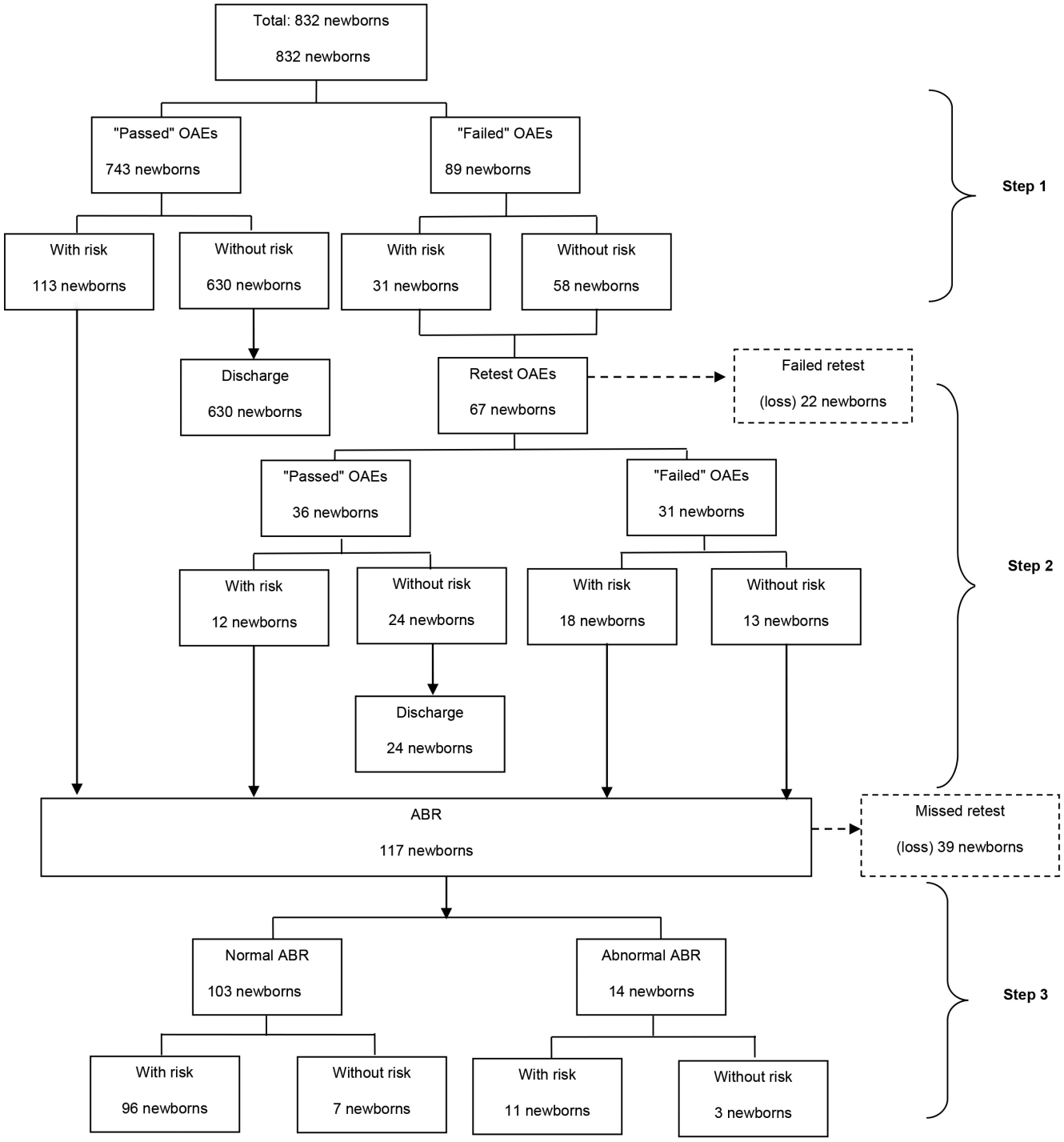
The literature recommends several protocols incorporating the techniques of TEOAEs and ABR for programs of universal newborn hearing. A combination of both techniques, as adopted in this study, has been recommended for over 20 years[18,19].

In this study, we were interested in identifying possible risk indicators that would increase the chances of "failure" in the OAEs testing and abnormalities in the ABR screening, due to



SubtitIte: BW = birth weight; ICU = Intensive Care; MV = mechanical ventilation; HL = hearing loss

Figure 1. Frequency of risk indicators in all newborns evaluated in the study



Subtitle: OAEs = Otoacoustic emissions; ABR = Auditory brainstem responses

Figure 2. Steps in the program of universal newborn hearing screening

the high demand of newborns at risk in the institution where the study was conducted.

When we verified the association between risk indicators and an increase in the possibility of a "failure" in the OAEs, we observed that the chances of the occurrence of at least one risk indicator were significant for newborns with birth weight <1500 g, genetic syndrome, or craniofacial malformation. There is evidence that a risk indicator for hearing loss increases the number of "failures" in the OAEs evaluation and that the identification of the real cause of the failure may help detect those cases that could indeed have an auditory abnormality[1,20,21,22].

However, when this relationship was established with the ABR (a test considered the gold standard in the diagnosis of hearing loss in newborns), only meningitis and craniofacial malformation showed statistical significance. Similarly, a study[23] has found an increased risk of deafness in the association

Table 1. Distribution of abnormal ABR per ear

ABR	RE	LE	Total
Mild abnormality (36-50 dBnHL)	3	6	9
Moderate abnormality (51-70 dBnHL)	1	2	3
Severe abnormality (71-90 dBnHL)	0	0	0
Profound abnormality (>90 dBnHL)	2	3	5
Retrocochlear abnormality	1	3	4
Total	7	14	21

Subtitle: ABR = Auditory brainstem responses; RE = right ear; LE = left ear; dBnHL = decibel normal hearing level

between meningitis and craniofacial malformation. Other studies, in turn, have found increased rates of deafness associated with the use of mechanical ventilation, ototoxic drugs, and family history of congenital hearing loss. These differences are probably related to the profile of the population at each center[24,25,26].

We must emphasize that craniofacial malformation was the only risk indicator that consistently affected different steps of the newborn hearing screening program.

Another aspect to be considered is that the interpretation of the abnormalities in both tests is different. For example, a failure in the OAEs test does not mean that the child has hearing loss, and the investigation of this result must be conducted carefully to prevent erroneous interpretations. The abnormalities found in the ABR tests, in turn, are of greater relevance and justify the importance of the cross check in the audiological diagnosis in children[27].

The present study showed that 22% of the newborns presented some degree of hearing abnormality, which was more common among those with risk factors for hearing loss. Most abnormalities were characterized by cochlear damage, a finding similar to that of other studies in the literature[22,23].

Although this study had a small number of retrocochlear abnormalities (characterized by an abnormal response in the OAEs and ABR evaluations), the identification of these cases allowed early intervention, reducing the impact that the impairment would otherwise have in the newborns' auditory development.

Loss of follow-up is a frequent problem in protocols that adopt more than one step, especially due to challenges in taking the newborn back to the hospital after discharge. This problem is commonly observed in cases that "passed" the first assessment and were expected to return for ABR screening due to a risk indicator. Thus, a direct referral for ABR testing of those newborns at risk for hearing loss would be an attempt to reduce the lack of adherence to the program. Other reasons related to loss of follow-up for many patients is residence in another city, rehospitalization, and/or a choice by the mother to retest the newborn at their hometown.

Lastly, the knowledge of the particularities of each service and the relevance of actual risk indicators currently affect the various stages of a newborn hearing screening program. Therefore, it is necessary to guide the multidisciplinary team involved in the newborn's health towards correct referral, diagnosis, and therapy.

CONCLUSION

Craniofacial malformation was an indicator strongly associated with a diagnosis of deafness, independent of the hearing screening being performed by OAEs or ABR testing at its various stages. This justifies continuous and systematic monitoring of the center in which the screening is performed in search for improvement in the quality of the auditory health program of the newborn.

Table 2. Chance of abnormality in the TEOAEs (test or retest) due to risk indicators (n=810)

Variable	β	SE	Wald	p-value	OR	(95% CI)
Low-birth weight	1.30	0.49	7.07	0.008	3.68	(1.41 – 9.63)
Malformation	2.64	0.92	8.31	0.004	14.00	(2.33 – 84.20)
Genetic syndrome	3.04	1.19	6.52	0.011	21.00	(2.03 – 217.26)
Presence of any risk factor	0.89	0.34	6.78	0.009	2.43	(1.25 – 4.75)
Constant	-2.83	0.17	280.84	0.000	0.06	

Model adjusted with forward selection procedures (p<0.05)
Subtitle: TEOAEs = Transient evoked otoacoustic emissions; SE = Standard error; Wald = Wald's test; OR = odds ratio; CI = confidence interval

Table 3. Chance of abnormality in the ABR due to risk indicators (n=117)

Variable	β	SE	Wald	p-value	OR	95% CI
Meningitis	1.887	0.971	3.775	0.052	6.60	(0.98 – 44.29)
Malformation	2.986	1.269	5.536	0.019	19.80	(1.65 – 238.10)
Constant	-2.293	0.332	47.735	0.000	0.10	

Model adjusted with forward selection procedures (p<0.05)
Subtitle: ABR = Auditory brainstem responses; SE = Standard error; Wald = Wald's test; OR = odds ratio; CI = confidence interval

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