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# The risk factor profile of children covered by the Polish universal neonatal hearing screening program and its impact on hearing loss incidence



# Maciej J. Wróbel<sup>a,\*</sup>, Grażyna Greczka<sup>a</sup>, Witold Szyfter<sup>b</sup>

<sup>a</sup> Department of Otolaryngology and Oncological Laryngology, Poznań University of Medical Sciences, Poland <sup>b</sup> Medical coordinator of the Polish Universal Neonatal Hearing Screening Program in Poland, Poland

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#### ABSTRACT

*Objective:* The high frequency of risk factors detected within the newborn population increases the total number of children that should receive regular follow-ups. However, in some circumstances, this could be beyond the capacity of the health system. Therefore, careful interpretation and selection of risk factors, and in particular of those factors not strictly defined, should be carried out during screening. The aim of the study was to analyse the risk factor profile of children covered by the national universal neonatal hearing screening program and to correlate it with hearing loss incidence.

*Patients and methods:* The analysis of records in the program database collected from 472 neonatal and well-baby units over a period of 10 years (2002–2012), focused on children with at least one risk factor. The analysis was subdivided into distribution of risk factors as well as to risk factors and hearing loss correlation.

*Results:* In the studied cohort of n = 137,432 children (4% of the total number of screened children) single risk factors were most frequently detected, accounting for 71% of records. The association of two or more risk factors appeared in 659 configurations (29%), with a mean of 3.1 coexisting risk factors and a maximum of 9. Hearing loss was dependent on the number of risk factors in a child, but reached its maximum with the association of 6 factors.

*Conclusions:* The detection of postnatal hearing loss should be continued in order to increase our understanding of hearing incidence and the role of environmental factors. To optimize screening, discussions between specialists (mostly related to the issue of risk factors detected and registered in the earliest stage of screening programs) would be beneficial.

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## 1. Introduction

Hearing loss is the result of the overlapping factors of genetic predisposition and environmental impact. The calculated ratio is about 1:3, one third with prevalence of genetic factors, one third with a prevalence of environmental factors, and one third is a mix of those two. Currently, the estimated incidence of hearing impairment is 1–3 cases per 1000 newborns [1], however, the frequency of hearing loss detected in children treated in neonatal intensive care units (NICUs) is approximately 10 times higher, which is mostly due to concomitant risk factors (RF) for hearing loss [2,3].

Tel.: +48 61 8691 387; fax: +48 61 8691 690.

The list of risk factors in neonatal hearing screening has been prepared and modified over years to define the group of children at increased risk of future hearing problems [4–6]. This group should be put into a detailed and prolonged program to monitor their hearing and development. The incidence of hearing deterioration related to each risk factor is known, but still genetic predisposition and other environmental aspects may influence the incidence, resulting in some of those children developing hearing loss, and some not.

The high frequency of risk factors detected within the newborn population increases the total number of children that should be given regular follow-ups after the re-evaluation of hearing status. This in some circumstances could be beyond the capacity of the health system resulting in delayed diagnosis or audiological intervention. Therefore, risk factors (particularly not strictly defined, e.g. familial history of hearing loss or the use of ototoxic drugs) should be carefully interpreted and selected during screening.

In Poland, the universal neonatal hearing screening program was introduced in 2002 as a public initiative with the help of the

<sup>\*</sup> Corresponding author at: Poznan University of Medical Sciences, Department of Otolaryngology, ul. Przybyszewskiego 49, 60-355 Poznań, Poland.

E-mail address: wrobmac@ump.edu.pl (M.J. Wróbel).

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Great Orchestra of Christmas Charity Foundation [7]. The program is based on otoacoustic emissions testing (OAE) and a questionnaire aimed at identifying the 13 risk factors defined by the Joint Committee on Infant Hearing published in 2000 [8]. Children that do not meet the pass criteria of the screening, i.e. that receive the result "refer" for at least one ear after OAE testing and/or are identified as having at least one of the risk factors listed in the questionnaire, are directed to the laryngological and audiological centres responsible for hearing evaluation and appropriate intervention.

Despite the decreasing birth rate in Poland, the number of children referred to consultation departments after screening significantly overloads the capacity of centres. This is mostly due to the sum of risk factors detected and recorded at the screening level. We have therefore attempted to analyse the risk factor profile in the whole population of children covered by the program. This may contribute to the discussion on the importance of single risk factors and the interpretation of those factors not clearly defined, and potentially lead to a re-evaluation of the program's screening questionnaire.

## 2. Methods

Between 2002 and 2012, a total of 3,495,733 children were registered in the database of the Polish universal neonatal hearing screening program. According to protocol, all children were screened with otoacoustic emission (OAE) and outcomes were presented as either "positive" (i.e. hearing problem detected) or "negative" (i.e. hearing problem not detected). These results correspond to "refer" in at least one ear, and "pass" in both ears, respectively. Within the whole screened population (n = 3,435,800, on the date of analysis), n = 137,432 children (4%) had at least one risk factor registered. This group was the focus of the study. The full list of risk factors used in the questionnaire was adopted from the JCIH statement form 2000 and has not been changed since the beginning of the program (Table 1).

Analysis focused on the frequencies and configurations of risk factors during screening. The referral rate was also calculated for the same group of children, and this was compared with the positive and negative screening results. Subsequently, with respect to the final diagnosis of the hearing impairment, logistic regression with odds ratio was calculated for children with various groups of risk factors. Univariate analysis (Pearson's chi-square) was used to identify the role of each risk factor in the incidence of hearing loss. The applied method calculated the chance of detection of hearing impairment in patients with several associated risk factors, among which the analysed risk factor was present. Evaluation was conducted in a group of n = 74,355 children i.e. in 54.1% of those with RF referred from screening. Of those, 4.55% were diagnosed with hearing impairment and 95.45% as normal hearing individuals.

All calculations were performed with MS Excel 2010 and Statistica 10.0 software.

The study was design as the retrospective analysis of the coded data from the program database. According to the local ethics committee it did not require additional approvals.

#### 3. Results

### 3.1. Risk factor distribution

Analysis revealed that 71% (n = 97,503) of children in the studied population had only one risk factor. The most frequent was

Table 1

a. Risk factor list and corresponding results of hearing evaluation. b. Risk factor distribution list and corresponding results of hearing evaluation.

List of risk factors	Risk factor	Cohort occurrence (%)	Screening positive (%)	Screening negative (%)	Hearing impairment (%)
1	2	3	4	5	6
Total	One RF	70.9%	11.7%	88.28%	4.04%
Familial hearing loss	RF1	15.3%	11.7%	88.34	5.98%
Craniofacial anomalies	RF2	2.4%	48.9%	51.09%	31.90%
Complex congenital anomalies	RF3	0.0%	-	-	-
Premature birth (gestational age $<34$ weeks)	RF4	2.3%	16.8%	83.19%	4.09%
Ototoxic medications	RF5	32.1%	9.3%	90.66%	2.41%
TORCH infections	RF6	10.8%	7.6%	92.36%	2.04%
Low birth weight	RF7	1.0%	27.0%	73.04%	5.77%
Apgar <4 in 1st min	RF8	4.7%	10.6%	89.39%	2.20%
Apgar <6 in 5th min	RF9	0.4%	18.7%	81.26%	1.53%
Hyperbilirubinemia	RF10	0.5%	11.0%	88.95%	1.56%
Bacterial meningitis	RF11	0.1%	15.2%	84.83%	7.25%
Intensive care >7 days	RF12	1.1%	15.8%	84.25%	3.55%
Respirator support (mechanical ventilation)	RF13	0.2%	16.5%	83.50%	3.05%
Configurations of risk factors	Risk factors	Cohort	Screening	Screening	Hearing
	in single patient	occurrence (%)	positive (%)	negative (%)	impairment (%)
1	2	3	4	5	6
Total	$>1 RF^{a}$	29.1%	26.8%	73%	5.83%
	2	13.5%	23.5%	76%	5.23%
	3	6.9%	26.8%	73%	5.51%
	4	3.9%	29.6%	70%	5.77%
	5	3.1%	31.1%	69%	6.65%
	6	1.2%	37.4%	63%	10.67%
	7	0.5%	39.2%	61%	9.25%
	8	0.1%	48.8%	51%	8.70%
	9	0.0%	66.7%	33%	0.00%
	10	0.0%	-	-	-
	11	0.0%	-	-	-
	12	0.0%	-	-	-
	13	0.0%	-	-	-
Whole group of children with recorded RF	Total	100.0%	16.1%	84%	4.55%

<sup>a</sup> More than one risk factor recorded in single patient.

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