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A regional-based newborn hearing screening program: the Emilia-Romagna model after ten years of legislation

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Abstract

Background. *Hearing loss, occurring in 1-3/1,000 newborns in the well-babies population, is one of the most common congenital diseases, and hearing screening at birth still represents the only means for its early detection. Since 2011 the Emilia Romagna Regional Health Agency has recommended Newborn Hearing Screening for all babies at its birth points and for newborns moving to the region. The aims of this study are to analyze the results of this regional-based Newborn Hearing Screening program and to discuss the impact of the legislative endorsement on the organization.*

Material and methods. *This is an observational retrospective chart study. The recordings of well-babies and babies at Neonatal Intensive Care Units were collected during the period from January 1st 2015 to December 31st 2020. The following data were included: Newborn Hearing Screening coverage, percentage of refer at otoacoustic emissions, prevalence and entity of hearing loss, unilateral/bilateral rate, presence of audiological risk factors.*

Results. *More than 99% of a total of 198,396 newborns underwent the Newborn Hearing Screening test during the period January 1st 2015 to December 31st 2020, with a coverage ranging between 99.6% and 99.9%. Overall, the percentage of confirmed hearing loss cases was about 17-30 % of refer cases, 745 children received a diagnosis of hearing loss (prevalence 3.7/1,000). Considering profound hearing loss cases, these represent 13% of bilateral hearing loss.*

Conclusions. *A regional-based Newborn Hearing Screening program is valuable and cost-effective. In our experience, the centralization of the data system and of the data control is crucial in order to implement its efficiency and effectiveness. Healthcare policies, tracking systems and public awareness are decisive for a successful programme implementation.*

Introduction

Hearing loss, occurring in 1-3:1,000 newborns in the well-babies population, is one of the most common congenital diseases and hearing screening at birth still represents the only means for its early detection (1). Without hearing rehabilitation, permanent childhood hearing loss (PCHL) can have negative effects on language, developmental and educational outcomes in children (2). Moreover, approximately 20-40% of children born with hearing impairment also have significant additional disabilities (3). There is evidence that newborn hearing screening (NHS) significantly reduces the age of hearing loss diagnosis (4) and that hearing-impaired children who are identified and habilitated early, perform better in their language development (5, 6) obtaining a better quality of life (7).

Given the effectiveness of NHS programs in the diagnosis of congenital hearing loss (8), the adoption of a universal model has been recommended by several international organizations. The guidelines of the Joint Committee on Infant Hearing (JCIH) recommend that each country adopt appropriate protocols according to its administrative and budget possibilities (9). Furthermore, the diagnostic-therapeutic process and the resource allocation should be based on consolidated local epidemiological data concerning congenital hearing loss in children. In Italy, a universal NHS program was introduced in 2003 by the Italian Institute of Social Medicine (10), however its implementation throughout the national territory required time, especially for organizational reasons. The Italian National Health Service provides universal coverage, and it is organized at three levels: national, regional, and local. Administrative data are collected by local Healthcare Units and are processed at the regional level.

An accurate legislative plan plays an important role in improving healthcare policies and since 2011 the Health Agency of

Emilia Romagna Region has recommended the NHS for all babies at its birth points and also for newborns moving to the region, even if they are not officially registered as residents (11). The resolution, which was drafted by expert physicians, introduced a two-stage screening protocol: otoacoustic emissions (OAE) test on the second day after birth or before discharge for well babies and both OAE test and aABR (automated Auditory Brainstem Responses) for children with audiological risk factors, according to the recommendations of JCIH. A NHS program had already been adopted, independently, by all third level hospitals before 2012; however, second and first level facilities began the NHS after the regional endorsement.

Contextually, a regional Hearing Disabilities Group (HDG) was established, consisting of a multidisciplinary group that involves hospital and territory departments, in order to coordinate the procedures. HDG has the role of technical co-ordination, planning of support, maintenance and network facilitation. It involves several health care professionals such as Neonatologists, ENT specialists, Audiologists, Childhood and Adolescence Mental Health Services (CAMHS) professionals. CAMHS ensure the presence of a tracking service that integrates the therapeutic-diagnostic pathway, promoting a surveillance of the identified cases.

The HDG has been fully operative since 2014 and it holds periodic meetings, dealing with clinical-technical problems and implementing data collection. Since it was not possible to adopt a unique regional database to collect the results of the screening procedures, a periodic survey was adopted to retrieve data and information, constantly, from all birth facilities and audiological services, in order to ensure a continuous information flow.

The aims of this study are (i) to analyze the results of the regional-based NHS program since its introduction and (ii) to discuss the impact of the legislative endorsement on the organizational system.

Methods

This is an observational retrospective chart study. All facilities and all audiology/ENT infantile services within the Emilia Romagna (ER) Region were screened. The recordings of well-babies and babies at the Neonatal Intensive Care Units (NICU) were collected over the period January 1st 2015 – December 31st 2020 by means of a questionnaire elaborated by the HDG.

NHS program

All children were initially screened by OAE test at birth facilities. The presence of OAE response in both ears was considered as a "pass". In case of a repeated unclear response ("refer") even only in one ear, a complete audiological evaluation was performed.

Children with audiological risk factors were tested using both OAE and aABR or clinical ABR (Auditory Brainstem Response).

Different instruments were used for OAE testing: third generation devices (ILO-292 by Otodynamics), fourth generation instruments (Accuscreen by Fischer-Zoth; Eclipse by Labat; Otoread by Interacoustics; Audioscreener by Viasys). Different devices were used to collect aABR, such as Natus (Algo portable), AccuScreen and Audioscreener.

Nurses, obstetricians, audiometry technicians, depending on each county organization, performed the first level OAE test. Retest by OAE was mostly performed by audiometry technicians.

In case of confirmed hearing impairment, the entity of hearing loss was defined by ANSI (American National Standards Institute) classification: normal hearing

(0-15 db), slight SNHL (sensorineural hearing loss) (16-25 db), mild SNHL (≥ 26 to < 40 dB); moderate SNHL (≥ 41 to < 65 dB), severe SNHL (≥ 66 to < 95 dB) and profound SNHL (> 96 dB) (12).

Questionnaires

The questionnaires employed were elaborated by the HDG to evaluate the implementation and to detect critical issues of the NHS program. They were subdivided in 4 sections: one to be filled in by each facility, one by each audiology service, two by each CAHMS.

In this study we have analysed reports from facilities and audiology/ENT infantile services, in particular. The questionnaire is filled in annually and is sent at least 5 months before the end of year. Therefore, all newborns refer at the screening usually receive an audiological evaluation, to confirm the presence of hearing loss as soon as possible, in the first months of life.

A few questions were about the organizational features of the program, the part concerning the instruments used and professionals involved in the pathway has been described in the NHS program methods section.

In this report, we analyzed:

- the way the test result was registered, for example, on a specific software or on paper register;
- if the test result was reported in the discharge letter;
- if the different professionals involved in administrating OAE tests, often characterized by high turnover, could benefit from periodical training. Concerning the demographic features of the tested children, the following data were asked:
- number of newborns and number of newborns who underwent NHS test;
- number of "refer" (unilateral/bilateral) children at OAE;
- number of children who underwent re-test and the results;
- number and entity of hearing loss cases, distinguishing unilateral and bilateral;
- number of children with audiological risk factors.

The questionnaire has been updated over the years: side and entity of hearing loss have only been requested since 2017, while the percentage of refer has been collected since 2016. Incidence of refer and hearing loss cases were calculated on the basis of data concerning the juvenile population.

Data Analysis

Data collected were analysed using STATA program version 14 (StataCorp LP 4905 Lakeway Drive College Station, Texas 77845 USA). The results are presented as percentages for categorical variables. First, for descriptive purposes, we conducted univariate analyses by generating absolute values and percentages for all variables, followed by weighted prevalence estimates and confidence intervals for the outcome variables.

Subsequently, prevalence and incidence values (two discrete measurements) were calculated using the following two denominators: (1) total number of cases of refer; (2) total number of cases of hearing loss. Point prevalence estimates were summarized descriptively and presented as the number of cases/10,000. Statistical significance was set at 5% (p= 0.05; two sided) throughout.

The present study was conducted in compliance with the Helsinki Declaration (2008). It was performed retrospectively through a systematic hospital case file review and therefore did not affect patient care in any way, since it only incorporates the recordings of a database and its evaluation.

Results

The number of births decreased by about 5,000 babies in six years. Indeed,

when the NHS program started, in ER there were 27 facilities in total, while, currently, these are reduced to 23. Audiology / ENT infantile services are 15 in total. All regional services involved answered the questionnaire annually. Over the years, birth points using specific software to register test results increased from 33% (2015) to 56% (2020), while additional use of paper registers decreased from 37% to 17%; there was not a great variation in the use of other systems such as excel system or hospital databases. In 2015, only 4 birth facilities did not report the test directly in the discharge letter, while in 2020 all adopted it. Training for professionals involved in OAE tests were periodically organized in all regional services.

The collected data were analyzed globally and on an annual basis. About 99% of a total of 198,396 newborns underwent the NHS test during the period January 1st 2015 - December 31st 2020. Coverage, across the studied period, ranged between 99.6% and 99.9%; newborns also presenting audiological risk factors were 4.11% to 6.88% of tested babies. Overall, 745 children were diagnosed with hearing loss (prevalence 3.7/1,000) (Table 1). The percentage of children referred for an audiological evaluation was 4.4% in the first year of observation and thereafter ranged between 1.3 and 2.4% in the subsequent years.

In Table 2 we have reported the incidence of hearing loss and refer cases in relation to the juvenile population. For all years, except for 2020, the incidence was between 1.8-2/10,000. Apart from the first year of data collection (2016), the percentage of confirmed hearing loss cases was about 17-30% of refer cases (Table 2). Our analysis yields an evidence-based estimate for the population incidence of hearing loss of 1.4–2/10,000, with no statistical significant difference in the reference years.

Data concerning entity and side of hearing loss have only been collected since 2017 (Figure 1). Between 2017 and 2020, 323/503 (64.2%) cases were affected by bilateral hearing loss (BHL). Mean prevalence of BHL was 2.5/1,000, while the mean prevalence of unilateral hearing loss (UHL) was 1.3/1,000 (180 cases).

As regards hearing loss severity, moderate hearing loss is the most frequent, affecting 39% of UHL and 47% of BHL; unfortunately, some of the 2017 data concerning hearing loss entity are missing. Profound hearing loss (40 cases in total) represents 13% of BHL and 7.9 % of the total hearing loss cases (prevalence 0.3/1,000). Considering severe and profound cases of hearing loss, these account for 33% of all BHL (see also figure 2).

The distribution in absolute numbers of BHL and UHL according to severity is indicated in figures 3 A and 3 B.

Table 1 - Results per year of observation (2015-2020).

Year	Newborns	Screened newborns	Coverage	Refer	Audiological risk factors	Cases of hearing loss	Prevalence/1000
2015	35,876	35,773	99.71%		4.11%	115	3.20/1000
2016	34,776	34,640	99.60%	4.4%	5.05%	127	3.65/1000
2017	33,410	33,289	99.63%	2.3%	6.88%	130	3.89/1000
2018	32,818	32,745	99.77%	1.35%	6.79%	134	4.08/1000
2019	31,195	31,081	99.63%	2.45%	5.77%	143	4.58/1000
2020	30,321	30,308	99.95%	1.76%	5.71%	96	3.16/1000
	198,396	197,836	99.71%	2.45%	5.71%	745	3.7/1000

Table 2 - Incidence of hearing loss and refer cases across the studied years. Juvenile population data were supplied by the Emilia Romagna Healthcare Service.

Year	Juvenile population (0-17y)	N. refer	Hearing loss cases	% hearing loss cases/ refer	Hearing loss Incidence/10,000	Incidence N. refer /10,000
2016	713,391	1542	127	8.2	1.8	21.6
2017	711,765	758	130	17.2	1.8	10.6
2018	708,622	444	134	30.2	1.9	6.3
2019	704,439	678	143	21.1	2.0	9.6
2020	698,003	501	96	19.2	1.4	7.2

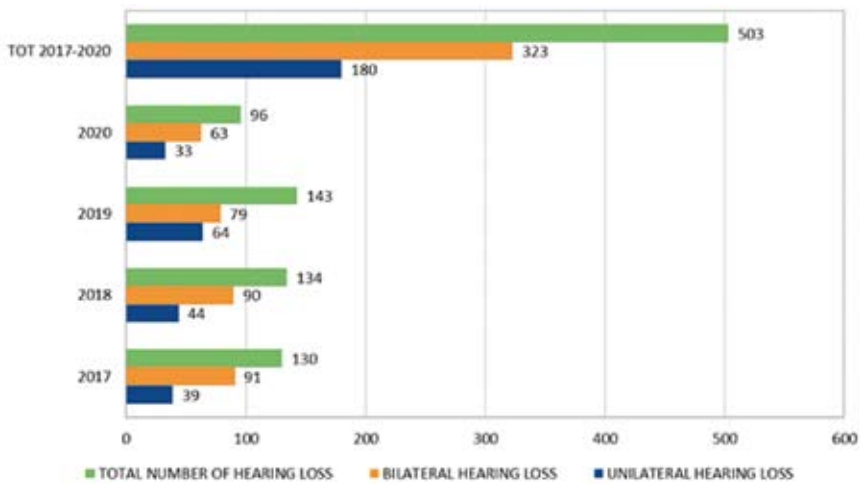

Figure 1 - Bilateral vs Unilateral cases of hearing loss, per year, and in total

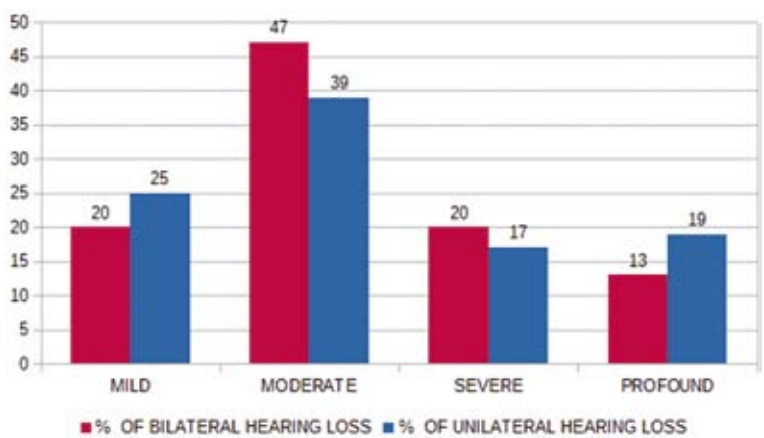

Figure 2 – Percentage values for total cases by hearing loss severity

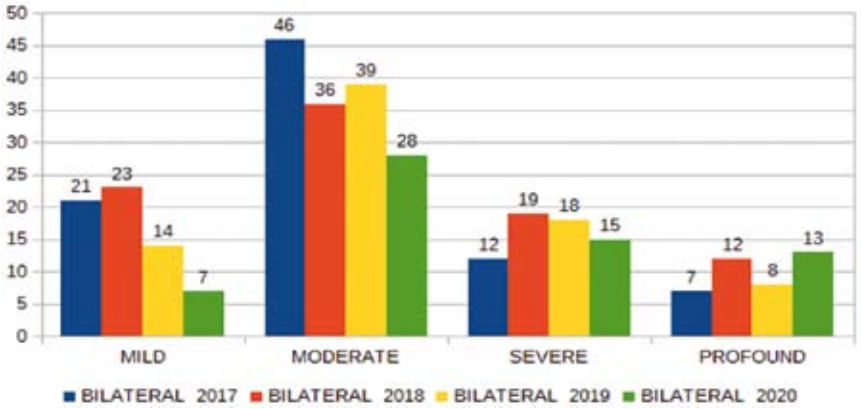

Figure 3A - Distribution in absolute number of bilateral hearing loss according to severity during years

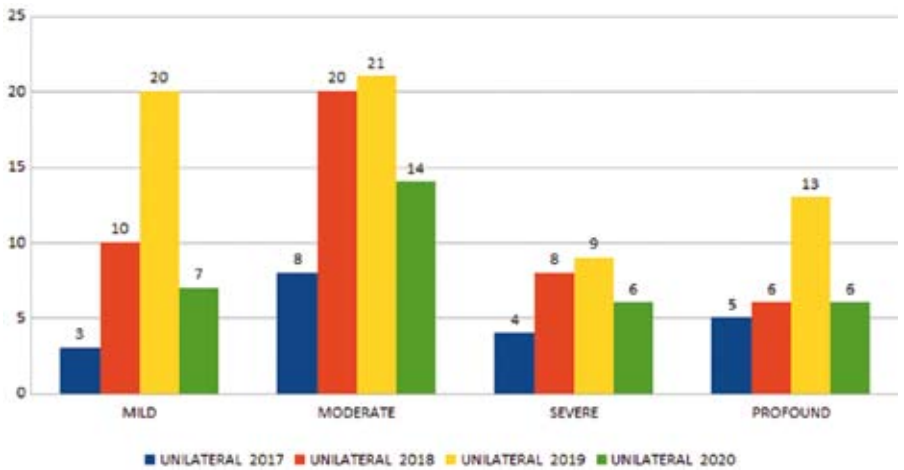

Figure 3B - Distribution of unilateral hearing loss according to severity, during years (by number of cases). In 2019 entity of one case is missing

No statistically significant differences were found across the studied years.

Discussion

Programs to identify hearing impairment have significantly improved over the last few years, and their implementation continues to grow throughout the world (13). The present study reports the ten-year experience of a NHS program, which was implemented on a regional basis and 6 years of data collecting, representing one of the most extensive in Italy in terms of study length. To the best of our knowledge, the originality of this experience lies also in its organizational system, in which the program, since its introduction, was monitored by a permanent multidisciplinary regional group, the HDG.

The NHS efficiency was analysed according to quality indicators such as coverage and referral rate (9). Coverage resulted above 95% and ranged over 99%,

immediately from the start of the NHS program. A high coverage rate is important when adopting universal screening (14), especially when it is compared to selective screenings based on high-risk criteria, since the latter do not detect all infants with congenital hearing loss, according to literature data (15). Indeed, it has been estimated that targeted screening would miss up to 40% of newborns with hearing loss, a considerable percentage (16). Significant contribution to the achievement of this high coverage is also due to the implementation of tracking results through software and to the reporting of OAE result in the discharge letter, which considerably reduces the possibility for children to skip the test.

These optimal results highlight the role of institutional promotion and reflect the strong support of healthcare administrators (17). Moreover, the performance of the screening test in birth facilities, carried out in 93% of countries in the world (2), appears the most efficient in reaching most newborns in the first days of life.

The referrals OAE rate, with the exception of the first year, was below 4%, ranging between 2.3 and 1.4%. This is a positive data, as a recent overview about NHS all over the world reports that, on average, 4.5% of the babies who underwent NHS failed the screen (2); we have to consider that in several facilities OAE are not performed by audiometry technicians and there is a high personnel turnover. A factor that mainly contributed to obtaining these percentages was the benefit of periodical training of the personnel, in addition to the institution of a central monitoring system.

In the present experience only 20-30 % of refer received a confirmed diagnosis of hearing loss, therefore it is likely that children were over-tested. According to many studies, the main reason for false-positive outcomes with OAE testing are transient conditions in the external auditory canal and middle ear, as well as high ambient noise level (13, 18). According to the literature, referral rates were greater for programs that screened newborns within the first 48 h of life (18). Moreover it is necessary to contemplate that hearing levels in premature infants can improve over time (19).

The problem of false positive at screening is well known. A low false positive rate avoids worrying unnecessarily about hearing impairments and minimizes unnecessary diagnostic tests (20) but parental uncertainty about screening results for newborns still can represent a partial obstacle, even though the benefits of early identification and intervention outweigh disadvantages (21, 22).

Early enrollment of babies presenting PCHL in intervention programs, requires strong tracking and follow-up procedures: data regarding the prevalence and distribution of hearing loss severity are crucial in order to plan and allocate economic resources. Prevalence of congenital hearing loss in our region ranged around 3.71,000 and, since the high coverage of OAE test, it appears to reflect the local reality, specifically because we did not find any statistically significant differences from 2016. Congenital hearing loss is linked to genetic and environmental factors, which are strictly related to socio-geographic aspects; therefore, it is extremely important to know the territorial variance in order to implement the healthcare system and to optimize resources (23).

These data are not far from the results of a recent systematic review and meta-analysis, which reported an overall prevalence of 2.21/1,000 (range 1-6) (24).

Another indirect measure of workload deriving from the NHS programme can be calculated through the incidence of hearing loss cases that was around 2/10,000, related to the regional juvenile population.

Major strengths of the study were the sample size, which expresses the results of a whole region and the high coverage rate of the NHS program. Moreover, it was possible

to extrapolate the incidence for each year.

The major drawback is the mild discrepancy of questionnaires during the observation period that did not always allow us to extrapolate the same data over the years. Another limitation is the lack of information about the composition of audiological risk factors.

Conclusions

The adoption of the regional NHS program is valuable and cost-effective; in our experience, the centralization of the data system and of the data control is crucial in order to implement the efficacy and effectiveness of the NHS. The constant information flow (birth points to HDG and viceversa) is very important to control the coverage and quality of the screening. Furthermore, the multidisciplinary approach is functional to monitoring the NHS results, and healthcare policies, tracking systems and public awareness are decisive for a successful programme implementation.

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Riassunto

Screening audiologico neonatale in Emilia-Romagna: valutazione dei dati a 10 anni dalla sua adozione

Premessa. L'ipoacusia è una delle patologie congenite di più comune riscontro, con una prevalenza stimata di 1-3/1.000 neonati. Lo screening audiologico neonatale, ad oggi rappresenta lo strumento più efficace per ottenere diagnosi e trattamento precoci dell'impairment uditivo. Nel 2011 l'Agenzia per la Salute della Regione Emilia Romagna ha emesso una delibera che ha raccomandato l'adozione dello screening uditivo neonatale per tutti i nuovi nati in regione ed anche per quelli che vi si trasferiscono.

Gli obiettivi di questo studio sono analizzare i risultati del programma di screening uditivo e discutere l'impatto della delibera regionale su di esso.

Materiali e metodi. Si tratta di uno studio osservazionale retrospettivo. Sono stati raccolti i dati relativi allo screening uditivo presso tutti i punti nascita e tutte le Unità di Terapia Intensiva Neonatale, nel periodo compreso tra il 1° gennaio 2015 ed il 31 dicembre 2020. In particolare, sono stati valutati i seguenti dati: la copertura dello screening, la percentuale di casi "refer" alle otoemissioni acustiche, la prevalenza e la incidenza dei casi di ipoacusia, la entità dell'ipoacusia, la presenza di fattori di rischio audiologico alla nascita.

Risultati. Oltre il 99% di 198.396 neonati è stato sottoposto a screening uditivo nel periodo di studio, con una copertura compresa tra il 99,6% e il 99,9%. Complessivamente, la percentuale di casi confermati di ipoacusia è stata di circa il 17-30 % di tutti i risultati "refer". Circa 745 bambini hanno ricevuto una diagnosi di ipoacusia, pari ad una prevalenza del 3,7/1.000. Il 13% dei casi di ipoacusia neurosensoriale è profonda.

Conclusioni. La organizzazione del programma di screening uditivo neonatale su base regionale è vantaggiosa. Nella nostra esperienza, la centralizzazione del sistema di controllo dei dati è stata determinante per implementarne l'efficacia. L'approccio multidisciplinare, le politiche sanitarie, i sistemi di monitoraggio e la sensibilizzazione del pubblico sono fattori importanti per ottenere una elevata copertura di un programma di screening audiologico.

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