

Evolution of Italian Universal Newborn Hearing Screening Programs

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Key words: Neonatal hearing loss, Newborn hearing screening legislation, congenital deafness
Parole chiave: Sordità neonatale, Screening uditivo neonatale, Legislazione, sordità congenita

Abstract

Background. The aim of this study was to evaluate the state of implementation of the Universal Newborn Hearing Screening Programs in Italy and to determine the effect that a health specific legislation may have on the percentage of infants screened for detection of hearing impairment in nurseries.

Material and methods. Italian Newborn Hearing Screening data were obtained during four national surveys (years 2003, 2006, 2008, and 2011). The screening rates obtained by the Regions which adopted or did not adopt a legislation to increase the newborns' coverage were compared.

Results. In 2011, the average coverage rate was 78.3%, but in 12 out of 20 Regions it exceeded 95%. Coverage rate was greater in Regions that implemented a health specific legislation compared to Regions that did not. As a matter of fact, Regions which passed the legislation screened more than 95% of infants, whereas Regions without legislation reported a mean screening rate of nearly 67% of newborns.

Conclusion. Current results seem to confirm that a specific legislation might have a decisive effect on the increase of rate of coverage of newborn hearing screenings.

Introduction

Hearing impairment is one of the most frequent defects at birth, occurring in about one to three infants per 1,000 live births (1-2). Congenital, profound deafness is the most socially debilitating condition, as it causes serious delays in speech, language, and cognitive development. In the absence of Early Hearing Detection and Intervention (EHDI) programs, the diagnosis of profound deafness is performed too late.

Recent studies on cortical plasticity during the postnatal life demonstrate that the development of the auditory cortex

is affected by the absence of hearing experience(2).

This delay prevents the children from getting good results in receptive language skill performances, educational attainment, and psycho-social development (3-4). Children with mild or moderate hearing loss are often identified even later, typically at school age.

Early hearing loss detection is a key issue to decrease social impact, disability and costs associated with the disease. In Italy, a recent study estimated that the lifetime mean cost assessed for a subject affected by profound pre-lingual deafness turned out to

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be equal to Euro 737,994.76 for a single male and Euro 755,404.02 for a female (5).

During the past years, the Universal Newborn Hearing Screening (UNHS)- and more generally the EHDI programs - have been endorsed worldwide by Governments and many international professional and advocacy organizations (6-11). Guidelines were developed by these national and international bodies recommending to perform hearing screening in infants by one month of age, to evaluate referral cases within three months and to initiate rehabilitation programs within the first six months of life.

In Italy, UNHS for early detection of congenital hearing loss is close to the final approval by the Ministry of Health and it is officially included in the so-called Essential Levels of Assistance (LEA), the list of medical performances granted by the National Health Service (NHS) in every Region, which got its final approval in Fall 2016.

In line with the current legislation, the NHS delegates the design and implementation of health programs for activation of UNHS to every Regional Administration.

By the end of 2011, Health Regional Laws, either prescribing or encouraging UNHS, were enacted only in seven Regions (Campania, Emilia, Friuli, Liguria, Lombardy, Marche, and Tuscany) out of 20.

Since 2001, the Italian Institute of Social Medicine (IIMS) promoted research and activities focused on prevention of hearing diseases and on neonatal hearing screening. Since 2003, IIMS performed systematical nationwide UNHS surveys. The current study analyses data of UNHS surveys performed by IIMS in the years 2003, 2006, 2008, and 2011.

Material and methods

An *ad hoc* questionnaire was developed by IIMS for the nationwide surveys on the

state of implementation of EHDI programs in Italy. Total annual births and number of screened newborns in each Birthing Hospital/Birthing Centre participating in the surveys were collected and saved in a national repository database. The surveys were conducted in the years 2003, 2006, 2008 and 2011. The questionnaire was sent to all maternity wards, including both well-baby nurseries and neonatal intensive care units of public hospitals, and of private hospitals affiliated with the NHS; only private clinics were not enrolled, due to the frequent lack of official registers.

The number of Hospitals involved in the four reporting years was 618 in 2003, 607 in 2006, 711 in 2008 and 505 in 2011. They accounted for 532,221 births in 2003 (corresponding to 97.8% of total live births), 541,970 births in 2006 (corresponding to 96.7% of total live births), 535,577 births in 2008 (corresponding to 92.8% of total live births), and 527,308 births in 2011 (corresponding to 96.7% of total live births).

The questionnaire swere filled in by either the Chief of the Hospital or the UNHS program Coordinator. All the Hospitals that were invited to participate in the surveys returned the filled-in questionnaires.

Data analysis and statistical evaluation

The percentages of newborns screened on a nationwide basis in the four reporting years were analyzed and compared. Additional analysis was done by clustering the regional data in five geographical areas: North West (Liguria, Lombardy, Piedmont, and Aosta Valley); North East (Emilia-Romagna, Friuli Venezia Giulia, Trentino Alto Adige and Veneto); Center (Abruzzo, Latium, Tuscany, Umbria and Marche); South (Basilicata, Calabria, Campania, Molise, and Apulia); and Islands (Sicily and Sardinia). Statistical evaluation with the t-test was done to assess

whether the percentage of newborns screened was modified according to reporting year and geographical area. The SPSS statistical software package (SPSS Statistical Data Analysis, SPSS Inc. Chicago, IL, 2007) was used. Statistical significance was accepted at $p < 0.05$.

Results

Data were collected through a questionnaire. The questionnaire was sent to all the public Birthing Hospitals/Centers and active in Italy and was filled in either by the Chief of the Hospital or by the UNHS program coordinator.

For each Hospital/Birthing Center participating to the survey, the following items were collected:

1. hospital profile: geographic location of the hospital (i.e., rural or urban position); total annual births; category of newborn units (i.e., level I or basic neonatal care, level II or specialty care, and level III or subspecialty Neonatal Intensive Care Unit - NICU).

2. UNHS program data: number of newborns screened for hearing loss; hearing screening and rescreening protocols used; percentage of infants referred for audiologic evaluation.

3. UNHS program structure and functions: program co-ordinator and staff performing the screening.

4. communication: staff in charge to convey hearing screening results to the parents and type of information given to parents.

In this study we have analyzed only the number of newborns screened at first level during admission to the nursery

All questionnaires were filled.

Figure 1 shows the distribution of total births in each geographical area in the four reporting years. It is noted that the number of births remained nearly stable across the reporting periods; the geographical area with the highest number of births was the North

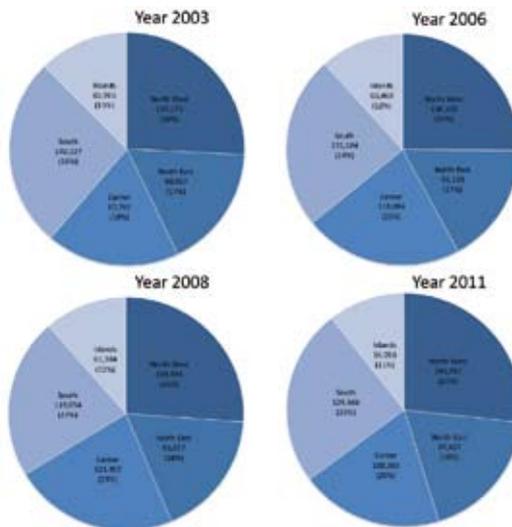


Figure 1 - Annual total births (and percentages on the total births in Italy) in the four reporting years (2003, 2006, 2008 and 2011) in each geographical area.

West, which accounted for 25-26% of total births in Italy, followed by the Center and the South (accounting for 18-26% of births), the North East (accounting for 17-18% of total births), and finally the Islands (11-13% of total births).

As shown in Fig. 2, screening coverage increased progressively from 29.3% (156,048 out of 532,221 births) in 2003 to

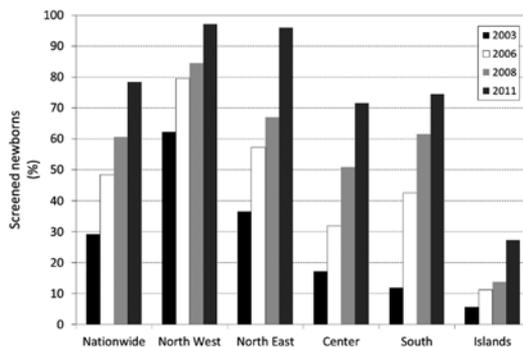


Figure 2 - Percentage of newborns screened in the four reporting years in Italy ('nationwide') and in each geographical area.

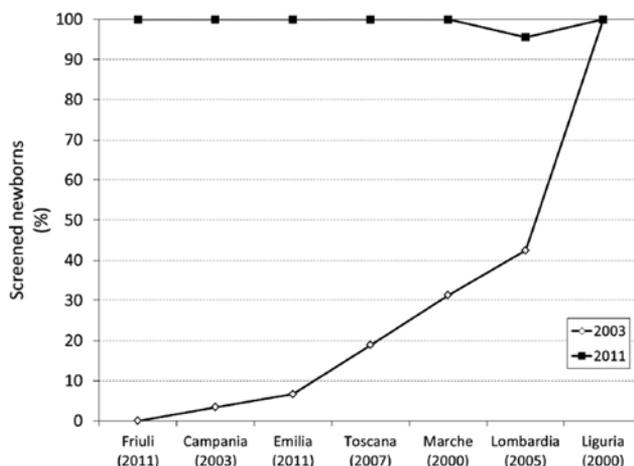


Figure 3 - Percentage of newborns screened in the years 2003 and 2011 in the seven Regions with UNHS legislation. Numbers under brackets are the years when the UNHS legislation passed in each Region. Lombardy and Liguria belong to the North West area, Emilia Romagna and Friuli Venezia Giulia belong to the North East area, Tuscany and Marche belong to the Center area, and Campania belongs to the South area.

48.4% (262,103 out of 541,970 births) in 2006, to 60.6% (324,537 out of 535,577 births) in 2008 and to 78.3% (413,212 out of 527,308 births) in 2011. Similarly, screening coverage increased during the 2003-2011 reporting period also in all the five geographical areas: from 62.2% (85,291 out of 137,175 births) to 97.1% (137,013 out of 141,067 births) in the North West; from 36.6% (33,315 out of 90,957 births) to 95.9% (93,433 out of 97,427 births) in the North East; from 17.3% (16,927 out of 97,797 births) to 71.5% (73,940 out of 103,395 births) in the Center; from 12.0% (16,795 out of 140,327 births) to 74.4% (96,260 out of 129,366 births) in the South; and from 5.6% (3,720 out of 65,965 births) to 27.4% (12,566 out of 56,053 births) in the Islands. The larger increase was observed in the Southern Regions, where the coverage in 2011 was 62.4 percentage points greater than in 2003, followed by the North-East (59.3 percentage points of increase), the Regions of the Center (54.2 percentage points of increase), the North-West (34.9 percentage points of increase), and the Islands (21.8 percentage points of increase).

The area with the highest coverage in all the four reporting years was the North-West followed by the North East; the lowest coverage was found in the Islands. In 2003, the coverage in the Center area was greater than that in the South, whereas in years 2006, 2008 and 2011, the South showed a greater coverage than the Center.

Figure 3 shows the comparison of the percentages of screening coverages in year 2003 and 2011 for the seven Regions with UNHS legislation. As reported at the bottom of the figure, in all these Regions, apart from Liguria and Marche, UNHS legislation was approved later than (or, at least, during) year 2003; Liguria and Marche were the only two Regions where legislation was passed before year 2003 (i.e., in 2000). For all seven Regions but Liguria, which already reached 100% coverage in 2003, screening coverage had a steep increase from year 2003 (when legislation was not yet approved or its application was at the very beginning) to year 2011 (when legislation was well established). Coverage was in all Regions but Liguria below 50% in year 2003 and increased well above 95% in the year 2011

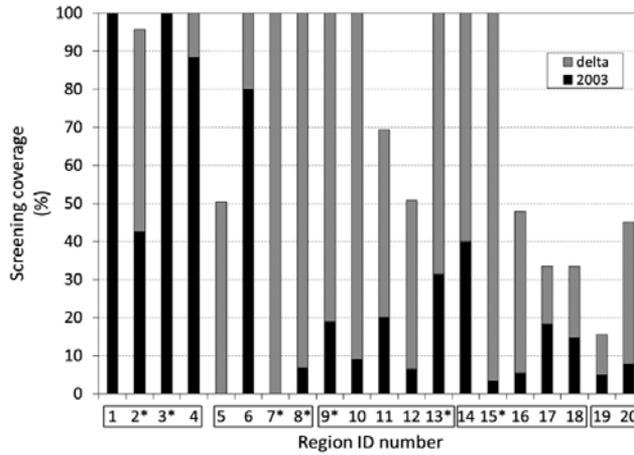


Figure 4 - Screening coverage (percent value) in 2003 (black bars) and difference ('delta') with the screening coverage in 2011 (gray bars) in each Region. The symbols * mark the seven Regions with UNHS legislation, as reported in Figure 3. Regions are grouped in five geographical areas: North West (1: Aosta Valley, 2: Lombardy, 3: Liguria, 4: Piedmont); North East (5: Trentino-South Tyrol, 6: Veneto, 7: Friuli Venezia Giulia, 8: Emilia Romagna); Center (9: Tuscany, 10: Umbria, 11: Abruzzo, 12: Latium, 13: Marche); South (14: Molise, 15: Campania, 16: Apulia, 17: Basilicata, 18: Calabria); Islands (19: Sardinia, 20: Sicily).

(only for Liguria, screening coverage was 100% both in 2003 and 2011).

Details on the difference between screening coverage in 2003 and 2011 in each Region are given in Figure 4. It can be seen that the Regions with UNHS legislation had the largest increase in coverage rate among the Regions belonging to the same geographical area (Liguria was an exception,

as the coverage had already reached 100% in 2003 and in 2011).

As to the comparison of the average screening coverage rates between Regions without and the seven Regions with UNHS legislation, the former had an improvement in coverage from year 2003 to year 2011 much greater than that observed in Regions without legislation: as a matter of fact, the average coverage rate increased from $28.8 \pm 34.9\%$ to $99.3 \pm 1.6\%$ in Regions with legislation and from $30.3 \pm 35.3\%$ to $66.8 \pm 28.6\%$ in the remaining Regions (See also Fig. 5). The increase of screening coverage in Regions with and without UNHS legislation was statistical significant ($p < 0.01$). Interestingly, it was noted that the average screening coverage in year 2003 was almost similar both in Regions with and without legislation and was well below 30%; on the contrary, in the year 2011, screening coverage was highly different, being well above 95% in Regions with UNHS legislation and still below 70% in those without.

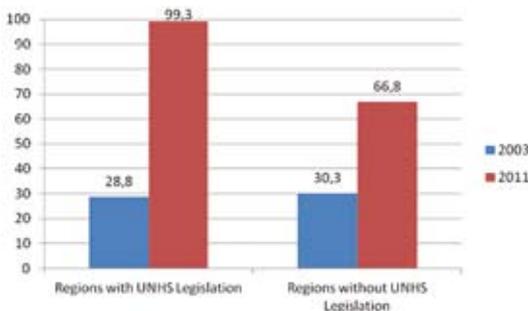


Figure 5 - Comparison of average screening coverage rates between the Regions without and the seven Regions with UNHS legislation.

Discussion

The majority of UNHS programs were implemented in the two most economically developed areas – the North West and North East: in these two areas, the coverage rate was greater than 95% in year 2011. Coverage rate still remained quite limited in a very few areas, such as in the Islands, where it remained below 30%. In 2011, screening coverage was greater than 95% in twelve out of 20 Regions. Of these twelve Regions, four were in the North West area, three were in the North East, three in the Center, and the last two were in the South.

A general trend of increasing coverage was observed in the period 2003–2011, both nationwide and in all the five geographical areas. An effect of this increase was that the difference in coverage rates among the geographic areas was less pronounced in year 2011 than in year 2003. As a matter of fact, in the year 2003, the coverage rate was greater than 60% only in one out of five areas (the North West) and was well below 40% elsewhere (in some areas, see e.g., the South and the Islands, it was even below 15%), whereas in 2011 the coverage rate was still very low (nearly 30% only in one area, the Islands) and reached values higher than 70% in the remaining areas. In three out of five areas (North East, Center, and South), the coverage increased from year 2003 to year 2011 more than 50 percentage points. Unexpectedly, the greatest increase (more than 60 percentage points) was observed in the South. The outperformance of this area was mainly due to the dramatic increase of coverage rate in Region Campania, which is one of the seven Regions with UNHS legislation and also is the region with the highest number of births/year within the southern area.

As a general trend, coverage rates in Regions with UNHS legislation were significantly higher than those in Regions without legislation. These findings seem to be in line with other recent analyses in the USA

(12) which confirmed that UNHS legislation seems to have a very positive effect on the performance of UNHS programs.

Conclusions

Our results revealed a progressive and steep diffusion of UNHS programs in Italy from 2003 to 2011; in year 2011 the average screening coverage nation wide was 78.3%. At the time of writing this paper, screening coverage remained low only in very limited parts of the Country. Many are the factors that contribute to a successful implementation strategy for the UNHS program. The analysis of the data collected in the present survey seems to confirm that Health Regional Legislation plays an important role in ensuring that newborns are screened for hearing loss, a good achievement for public health.

Riassunto

Sviluppo dei Programmi di Screening Uditivo Neonatale Universale in Italia

Obiettivo. Lo scopo del presente studio è quello di valutare lo stato di copertura dei programmi di Screening Uditivo Neonatale Universale in Italia, e determinare l'effetto che una specifica legislazione sanitaria può avere sull'incremento della percentuale di neonati sottoposti a screening per la rilevazione della sordità nei punti nascita.

Materiali e metodi. I dati relativi allo Screening Uditivo Neonatale in Italia sono stati ottenuti nel corso di quattro indagini nazionali (nel 2003, 2006, 2008, e 2011).

Sono stati confrontati i tassi di copertura dello screening delle Regioni che avevano adottato una legislazione sanitaria e di quelle senza legislazione.

Risultati. Nel 2011, il tasso di copertura medio nazionale è stato del 78,3%, mentre in dodici regioni su 20 superava il 95%. Il tasso di copertura è stato maggiore nelle regioni che hanno emanato una legislazione, rispetto alle regioni che non lo hanno fatto.

I dati dimostrano che le regioni che hanno approvato una legislazione sullo screening, hanno incrementato oltre il 95% il numero di bambini selezionati, mentre le

regioni, senza una legislazione approvata, presentavano un tasso medio di screening del 67%.

Conclusioni. I nostri risultati sembrano confermare che l'approvazione di una legislazione specifica potrebbe avere un effetto determinante sull'incremento del tasso di copertura dello screening uditivo neonatale.

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