International Journal of Pediatric Otorhinolaryngology 78 (2014) 1496–1502

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Main outcomes of a newborn hearing screening program in Belgium over six years

Bénédicte Vos [a,b,*], Raphaël Lagasse [a], Alain Levêque [a,b,c]

[a] Université libre de Bruxelles, School of Public Health, Research Center Health Policy and Systems – International Health, Route de Lennik 808, Brussels 1070, Belgium
[b] Centre d'Epidémiologie Périnatale (CEpiP), Route de Lennik 808, Brussels 1070, Belgium
[c] Université libre de Bruxelles, School of Public Health, Research Center Epidemiology, Biostatistic and Clinical Research, Route de Lennik 808, Brussels 1070, Belgium

ARTICLE INFO

Article history:
Received 14 March 2014
Received in revised form 10 June 2014
Accepted 14 June 2014
Available online 20 June 2014

Keywords:
Newborn hearing screening
Otoacoustic emissions
Hearing loss

ABSTRACT

Objective: To present the outcomes of the newborn hearing screening program in Belgium (French-speaking area) since its implementation and to analyze its evolution between 2007 and 2012 in the neonatal population without reported risk factors for hearing loss.
Methods: The study was descriptive and based on a retrospective analysis of six annual databases (2007–2012) from the newborn hearing screening program. The main outcomes were identified: prevalence of reported hearing impairment; coverage rates (first and second test, follow-up); proportions of conclusive screening tests; referral rate. Each outcome was presented for the six years and by year of birth. Chi-squares were used to study differences in the various outcomes according to time.
Results: Over the six years, 264,508 newborns were considered as eligible for the screening. Hearing impairment was confirmed in 1.41‰ ($n = 374$) of them, with significant disparities from year to year, between 0.67‰ and 1.94‰. Analysis of the screening process showed that only 92.71% ($n = 245,219$) of the eligible newborns underwent a first hearing test. This coverage rate varied greatly over time: at the beginning, less than 90% of the newborns had a first test and it rose to almost 95%. After the two screening steps, 2.40% ($n = 6340$) of the newborns were referred to an ENT doctor; the referral rate slightly decreased during the first years of the program and then stabilized around 2.4%. Over the period, only 62.21% of the referred newborns had a follow-up; the follow-up rate was particularly low for the first year (44.91%) and then strongly increased (+19.52% in 2008) but never exceeded 70%.
Conclusions: Outcome measures for the newborn hearing screening program in Belgium are lower than the benchmarks released by the Joint Committee on Infant Hearing. Nevertheless, the evolution of the outcome measures since the implementation of the program has been positive, particularly during the first years. At some point, most of the outcome measures decreased or at least did not change any further. The motivation and commitment of the professionals have to be supported in a variety of ways to improve outcome measures and thus, the quality of the program.

1. Introduction

European, American and international groups of experts have recommended the organization of universal newborn hearing screening (UNHS) for years [1–4]. The main purpose of a UNHS is to lower the age of hearing-impaired children at the time of diagnosis allowing earlier intervention. According to the Joint Committee on Infant Hearing (JCIH), comprehensive audiological assessment should be performed before 3 months of age, and appropriate intervention should begin before 6 months of age [2].

In the well-baby nursery population, prevalence of "significant bilateral hearing impairment" (HI), whose consequences are particularly severe for children's development, is 1–3 per thousand newborns [5]. However, UNHS programs aim to identify all kinds and degrees of HI among the bilateral or unilateral hearing-impaired newborns [2].

To assess UNHS programs, benchmarks and quality indicators have been released. The JCIH published the most frequently used benchmarks which are primarily related to the outcomes of the implemented UNHS programs, expressed as the minimum proportion of children who should be screened or who should be referred to

* Corresponding author. Tel.: +32 2 555 40 98.
E-mail addresses: benevos@ulb.ac.be (B. Vos), rlagass@ulb.ac.be (R. Lagasse), aleveque@ulb.ac.be (A. Levêque).

http://dx.doi.org/10.1016/j.ijporl.2014.06.019
0165-5876/

an ENT doctor, or the proportion of duly followed up newborns, required to be considered as a high-quality program [2,5].

UNHS programs have been implemented in different countries or areas and a lot of reports and studies have been published about the organization, protocols, and main outcomes of the programs [6–10]. In Belgium, the Fédération Wallonie-Bruxelles (FWB) (French-speaking area) has implemented a UNHS program since 2007, in collaboration with maternity hospitals.

The objectives of this study were to present the outcomes of the UNHS in the FWB since the beginning of the program and to analyze its evolution between 2007 and 2012 in the neonatal population free of risk factors for hearing loss. Specific outcome measures of UNHS programs, especially those defined by the JCIH, were used to assess the quality of the program and its potential development. In the discussion section, outcomes from the UNHS program in the FWB were compared to those from other UNHS programs.

2. Methods

2.1. Study design

Study design was descriptive and based on a retrospective analysis of six annual databases (2007–2012) from the UNHS program in the FWB. The same data management was applied annually to each database, and after the closure of the annual reports no new or updated hearing results or diagnosis were added to the database.

2.2. Population

Around 55,000 children were born annually in the FWB. In 2007, the FWB contained 50 maternity hospitals; three maternity wards were closed in 2008, and one more in 2010.

2.3. UNHS protocol

Participation of the maternity hospitals in the UNHS program is on a voluntary basis. The UNHS protocol proposes different tests and organizations depending on the presence or the absence of risk factors for hearing loss [11]. This study focused on the newborns without reported risk factors for hearing loss, thus only this specific part of the protocol was presented. A two-step screening is planned: automated otoacoustic emissions (AOAE) are performed during the stay in the maternity ward. The first step is performed on day two or day three, and a second step is performed the following day in the event of a failed test ("refer") on one or both ears. If the refer result persists on one or both ears on the second step, children are referred to an ENT doctor for an audiological assessment within two weeks (Fig. 1). When the screening process is not finalized during the stay in the maternity hospital, parents are invited to have the procedures performed in an outpatient clinic, during the four weeks thereafter. Professionals performing the screening tests work either in the maternity ward (midwives, nurses or childcare assistants) or in the outpatient clinic (nurses, speech therapists or audiologists). Each hospital is free to designate the professionals in accordance with its local resources.

2.4. Devices

Each maternity hospital chooses its own screening device, provided that the protocol can be applied: the Madsen Accuscreen® was the most frequently used device during the study, but the Natus Echo-Screen® and the Otodynamics Echocheck® were also used (some hospitals have used the new version of the Madsen Accuscreen® since 2011). All devices use the default "pass-refer" algorithm.

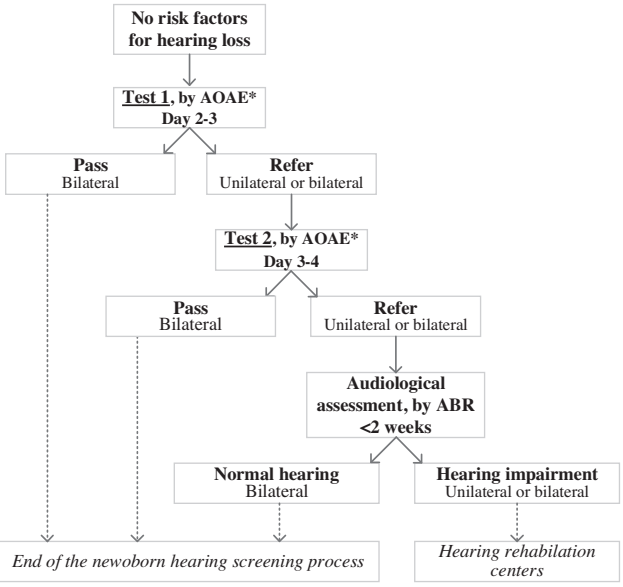


Fig. 1. Protocol of the newborn hearing screening program in the FWB, for the newborns without risk factors for hearing loss.

2.5. Data collection

At the beginning of the UNHS program, tests results were collected by the three neonatal blood screening centers in the FWB, and databases were sent annually to the coordinating agency for the UNHS program to monitor the program. Since 2011, computerized data collection based on the Internet has progressively replaced the initial system: screening results were transferred directly from devices to the central database, and results of the audiological assessments were directly typed in the database by the ENT doctors. Both systems still coexisted in 2012. The Internet database was managed by the coordinating agency and the neonatal blood screening centers did not participate in this system.

2.6. Inclusion and exclusion criteria

We included in this study children born between 1st of January 2007 and 31st of December 2012 in maternity hospitals participating in the UNHS program. However, due to the progressive recruitment of the hospitals in the UNHS program, we only included the children born in these specific maternity hospitals since the effective implementation of the program in these hospitals in this study.

We excluded newborns with risk factor for hearing loss: risk factors considered in the program[1] were based on the list from the JCIH (2000) [12] and adapted to the context and population in the FWB [11]. Risk factors were reported by professionals performing the screening tests, pediatricians, or ENT doctors.

[1] Family history of hereditary hearing loss; consanguinity (1st degree), in-utero infection (cytomegalovirus, toxoplasmosis, herpes, rubella, syphilis), poisoning (alcohol, drugs) by the mother during pregnancy, Apgar score of 0–6 at 5 min, gestational age <36 weeks and/or low birth weight (<1500 g), NICU admission for more than five days, exposure to ototoxic medications, hyperbilirubinemia at level requiring exchange transfusion, assisted ventilation lasting ≥24 h, head or neck anomalies and by extension each syndrome known to include a hearing loss, neurological or endocrine disease.

2.7. Outcome measures and statistical analysis

Different outcomes were used in this study. Firstly, parental refusal of the hearing test, parents who wanted their child to be tested by the Flemish UNHS program (because they speak Flemish and come under the Flemish program), and parental wished their child to be tested in another institution/by another ENT doctor were identified as three specific outcomes. We considered these newborns as not eligible for the program and we excluded them for the rest of the analysis, on the condition that no test was performed.

Secondly, as major outcomes of the program, we presented coverage rates and "pass-refer" rates for the first and second screening test, respectively, as "tested"/"not tested" and "pass"/"refer" as shown on the device screen. "Pass" means that the result was "pass" bilaterally. The denominator changed for each outcome: coverage rates were calculated according to the number of newborns that were supposed to undergo the first or second screening test or the audiological assessment; pass-refer proportions were calculated according to the number of newborns who underwent the mentioned test. The denominator for calculating the referral rates to ENT doctors was the newborns considered as eligible for the program.

Prevalence of HI included all kinds of hearing loss (sensorineural, temporary or permanent conductive hearing loss, and unspecified), whatever the degree of the impairment and whether it was unilateral or bilateral. The denominator for calculating reported HI was also the newborns considered as eligible for the program.

As a final summary outcome of the UNHS, the hearing status of the newborns was identified. It was composed of four groups: children who were not tested, bilateral normal-hearing children, children lost to follow-up (conclusion is unknown after one or more inconclusive tests) and hearing-impaired children.

Each outcome of the UNHS program in the FWB was presented for the six years as a whole and also by year of birth. We used chi-square tests to study the differences according to the year of birth. We also presented an overall figure to illustrate the steps of the protocol, including the number of newborns considered for the six years as a whole. We used STATA 12.0 for data management and statistical analysis.

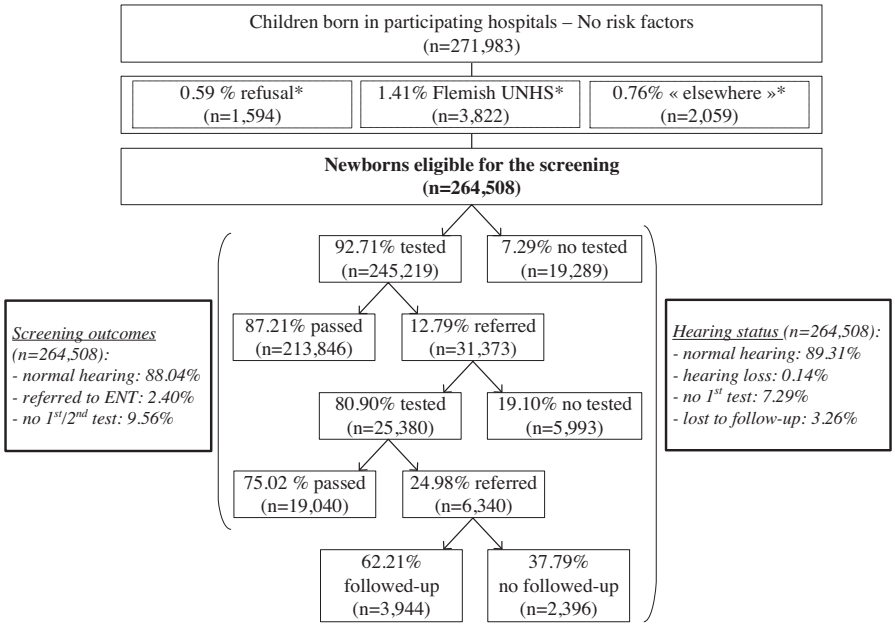
3. Results

3.1. Parental refusal, wish to have the screening performed by the Flemish UNHS program or by another institution/ENT doctor

Among the 271,983 children without reported risk factors for hearing loss born between 2007 and 2012 in the participating hospitals, the proportions of parental refusal, parental wish to have the test performed by the Flemish program or by another institution/ENT doctor were 0.59%, 1.41%, and 0.76%, respectively (Fig. 2). Over the six years, the annual proportion of parental refusal regularly decreased, from 1.07% in 2007 to 0.25% six years later. On the other hand, the proportion of parents who wanted the hearing test to be performed by the Flemish UNHS program gradually increased over the same period, tripling between the first (0.4%) and the second year (1.18%), and rising to 2.09% in 2012. Parental wish to have the hearing test performed in another hospital than the birth hospital or by another ENT doctor increased during the first three years, from 0.55% to 1.07% and then decreased during the following three years, to its lowest rate in 2012 (0.34%). Associations between each outcome and year of birth were statistically significant (Fig. 3). We did not consider these newborns as eligible for the UNHS program.

3.2. Evolution of the number of eligible newborns

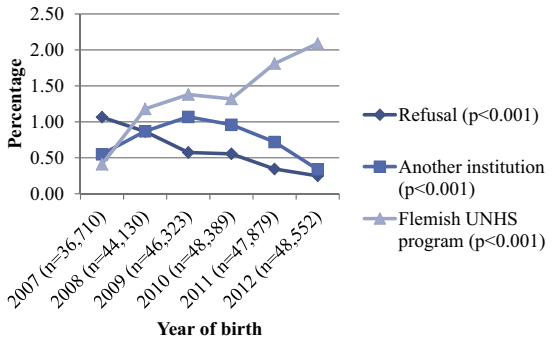
For the whole period, 264,508 newborns constituted the eligible population for the UNHS program. The largest increase of newborns occurred between 2007 and 2008 (+19.11%), followed



Flowchart of the UNHS protocol: each box presents the proportion and the number of newborns for the outcome analyzed; proportions are calculated on the box just above.

** Refusal: parental refusal of the screening; Flemish UNHS: parental wish to perform the screening in the UNHS Flemish program; Elsewhere: parental wish to perform the screening in another institution/by another ENT doctor*

Fig. 2. Process and results of the UNHS program for all children without risk factors born between 2007 and 2012 in the FWB.



(n): newborns without risk factors for hearing loss born in the participating hospitals

Fig. 3. Parental refusal, wish to have the hearing screening performed in another institution or by the Flemish UNHS program, by year of birth (2007–2012). (*n*): newborns without risk factors for hearing loss born in the participating hospitals.

by an increase of less than 5% in the two following years. Since 2011 the number of eligible newborns has stabilized (Table 1).

3.3. Coverage rate (first test)

Only 245,219 (92.71%) of the eligible newborns underwent a first hearing test. This coverage rate varied greatly over the time ($p < 0.001$): in 2007, less than 90% had a first test and the coverage rate rose to 93.35% in 2008 (+4%). The rise continued for the following two years but since 2011, the coverage rate for the first test has fallen to around 92% (Table 1).

3.4. Pass rate (first test)

Among the 245,219 tested newborns, 213,846 (87.21%) had a bilateral pass result and 31,373 (12.79%) needed a second screening test. Over the six years, the annual percentage of pass at the first test ranged from 85.38% in 2007 to 87.95% in 2010 and statistically differed between years of birth ($p < 0.001$) (Table 1).

3.5. Coverage rate (second test)

Only 25,380 out of the 31,373 children (80.90%) with a refer result at the first screening test underwent the second screening test, as recommended by the protocol. The second-test coverage rate was considerably lower than the first, but it rose from 77.03% in 2007 to 84.61% in 2012. Improvements were particularly observed in 2010 and 2011 (+2.93% and +3.96% in comparison to the previous year) (Table 1).

3.6. Pass rate (second test)

Among the 25,380 re-tested newborns, 19,040 (75.02%) had a bilateral pass result, whereas 6340 children were referred to an ENT doctor after two inconclusive hearing screening tests (unilateral or bilateral). The proportion of pass results at the second step increased from 2007 to 2009 (from 72.87% to 76.37%) but it decreased in 2010 to the same level as the first year before increasing again (Table 1).

3.7. Referral rate, for further evaluation

After the two screening steps, 6340 newborns were referred to an ENT doctor, resulting in a global referral rate of 2.40% of the eligible population. The referral rate slightly decreased during the first years of the program and then stabilized around 2.4% (Table 1).

3.8. Coverage rate for the follow-up

Over the period, 3944 out of the 6340 newborns referred to an ENT doctor (62.21%) received a follow-up. The coverage rate for the follow-up was particularly low for the first year (44.91%) and then strongly increased in 2008 (+19.52%) but never exceeded 70%. Except during the first year, follow-up coverage ranged between 60.84% and 69.43% (Table 1).

3.9. Reported prevalence of hearing impairment

A diagnosis of HI was confirmed in 374 newborns (1.41‰ of the eligible population), with significant disparities between the years: for children born in 2007, reported prevalence was 0.67‰. It was never lower than 1.2‰ during the next five years; in 2008 and 2009, this prevalence was around 1.9‰ of newborns. Differences of reported prevalence of HI between the years were statistically significant ($p < 0.001$) (Fig. 4).

3.10. Hearing status of the newborns

Between 2007 and 2012, 19,289 eligible newborns (7.29%) did not undergo a hearing test. Throughout the entire process, whether the newborns had one or two screening tests or they were referred to an ENT doctor for further evaluation, 236,231 of the 264,508

Table 1
Main screening outcomes of the UNHS program in the FWB, by year of birth and globally (2007–2012).

		2007 *n* (%)	2008 *n* (%)	2009 *n* (%)	2010 *n* (%)	2011 *n* (%)	2012 *n* (%)	*p*	2007–2012[a] *n* (%)
1st test	Screening test	(*n* = 35,969)	(*n* = 42,844)	(*n* = 44,923)	(*n* = 47,020)	(*n* = 46,502)	(*n* = 47,250)	<0.001	(*n* = 264,508)
	Test performed	32,129 (89.32)	39,997 (93.35)	42,115 (93.75)	44,308 (94.23)	42,877 (92.20)	43,793 (92.68)		245,219 (92.71)
	Test result	(*n* = 32,129)	(*n* = 39,997)	(*n* = 42,115)	(*n* = 44,308)	(*n* = 42,877)	(*n* = 43,793)	<0.001	(*n* = 245,219)
	Pass	27,431 (85.38)	34,672 (86.69)	36,946 (87.83)	38,967 (87.95)	37,464 (87.38)	38,366 (87.61)		213,846 (87.21)
2nd test	Screening test	(*n* = 4,698)	(*n* = 5,325)	(*n* = 5,169)	(*n* = 5,341)	(*n* = 5,413)	(*n* = 5,427)	<0.001	(*n* = 31,373)
	Test performed	3619 (77.03)	4188 (78.65)	4042 (78.20)	4333 (81.13)	4606 (85.09)	4592 (84.61)		25,380 (80.90)
	Test result	(*n* = 3,619)	(*n* = 4,188)	(*n* = 4,042)	(*n* = 4,333)	(*n* = 4,606)	(*n* = 4,592)	<0.001	(*n* = 25,380)
	Pass	2637 (72.87)	3176 (75.84)	3087 (76.37)	3157 (72.86)	3471 (75.36)	3512 (76.48)		19,040 (75.02)
Follow-up	Newborns referred	(*n* = 982)	(*n* = 1,012)	(*n* = 955)	(*n* = 1,176)	(*n* = 1,135)	(*n* = 1,080)	<0.001	(*n* = 6,340)
	Follow-up performed	441 (44.91)	652 (64.43)	581 (60.84)	787 (66.92)	788 (69.43)	695 (64.35)		3944 (62.21)
Newborns eligible		(*n* = 35,969)	(*n* = 42,844)	(*n* = 44,923)	(*n* = 47,020)	(n = 46,502)	(*n* = 47,250)	<0.001	(*n* = 264,508)
Referred to ENT (>2 tests)		982 (2.73)	1012 (2.36)	955 (2.13)	1176 (2.50)	1135 (2.44)	1080 (2.29)		6340 (2.40)

n: number of newborns concerned by the outcome analyzed; *n* (%): number related to the proportion in brackets.
[a] Outcome measures combined for the years 2007–2012 were also presented in the flowchart (Fig. 3).

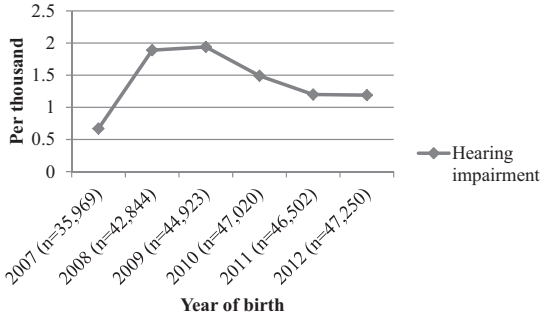
eligible newborns (89.31%) were considered as having normal bilateral hearing, 8614 (3.26%) began the hearing procedures without finishing and 374 (0.14%) had hearing impairment (Fig. 2). The proportion of children identified with normal bilateral hearing was lower than 85% in the first year and increased to 91% in 2010; it stabilized around 90% during the last two years of the study. The proportions of untested newborns followed a similar trend, improving only between 2007 and 2010 (Fig. 5).

4. Discussion

A major objective of a newborn hearing screening program is to identify children with HI, and to lower the age at the time of diagnosis. In our study, prevalence of HI reported by the UNHS program was 1.41‰ over the six years. This reported prevalence varied over the years: it was rather low in 2007 (0.67‰) and sharply increased to around 1.90‰ in 2008 and 2009. This improvement is due to better collaboration with the ENT doctors about notification of HI in databases: to improve quality data and data collection, each ENT doctor responsible for the UNHS program in the hospitals was asked annually, since 2009, to enter information about hearing-impaired children into the databases.

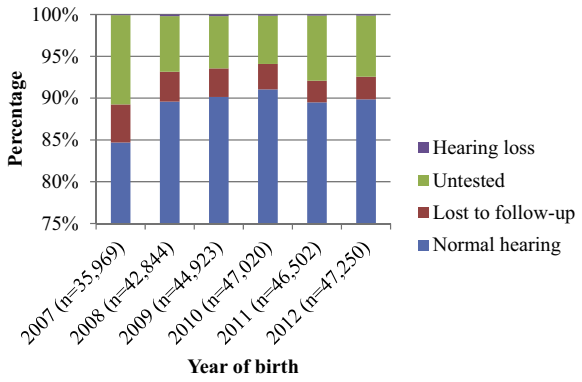
The prevalence of HI reported by the program in the FWB was in the same range as in other programs [10,13], but this comparison is difficult due to our methodology focusing on newborns without risk factors for hearing loss. Also, the quality of the data was rather poor: hearing threshold and type of HI could not be used. Moreover, HI was under-reported: in some cases, closure of the database a few months after the end of the previous year was too short a period to settle a diagnosis and this explains at least partially this under-reporting of HI. However, to standardize the method and compare the prevalence reported by year, we decided not to include these few hearing-impaired newborns in the study. As a consequence, one of the final outcomes of the program thus needed to be improved. Moreover, other outcomes such as age at diagnosis, assurance that children are taken care of in a specialized center, and type of hearing aid were not collected for global assessment of the program.

The coverage rates (first test, second test and follow-up) are important outcomes to monitor in a screening program. In the FWB, the analysis of the first-test coverage rate shows an increase from less than 90% of the eligible population in 2007 to more than 94% in 2010, however it then fell to a figure lower than the second year. A specific analysis conducted by maternity hospital showed a reduction in the coverage rate in some maternity hospitals in 2011 and 2012: a lack of data collection mostly explains these low rates, not an absence of hearing tests being performed. Therefore we strongly advise the development of daily data management and



(n): newborns without risk factors for hearing loss born eligible for the UNHS program

Fig. 4. Hearing impairment among population eligible for the UNHS program in the FWB, by year of birth (2007–2012) (<0.001).
(*n*): newborns without risk factors for hearing loss born eligible for the UNHS program.



(n): newborns without risk factors for hearing loss eligible for the UNHS program

Fig. 5. Hearing status by year of birth (2007–2012) for newborns without risk factors for hearing loss in FWB (<0.001).
(*n*): newborns without risk factors for hearing loss eligible for the UNHS program.

the increase of individual support to each hospital by the coordinating agency. This would support the commitment of the professionals involved in the UNHS and, as a consequence, improve the outcome measures and the quality of the program.

Annual and global coverage rates in the program in the FWB were lower than the minimum of 95% of newborns who completed screening, a benchmark recommended by the JCIH. The first-test coverage rate in the FWB was also lower than in other European (Hessen, Champagne-Ardenne) or Australian programs where coverage rates reached 95–98% [2,6,14,15]. However, the program in the FWB almost achieved a same coverage as in the Hamburg or Sienna programs (93–94%) [16,17].

Compared to the first-test coverage, the second-test coverage rate in the FWB was significantly lower and changed mainly between 2009 and 2011: this improvement could have been caused by a change in the protocol, which encouraged performing hearing tests one day earlier with the aim of reducing incomplete screening procedures. The updated recommendation was to perform the first hearing test on the second day of life (and possibly sooner in the event of a short stay at the hospital) and the second test the following day. In the FWB, the hospital stay in maternity ward generally lasted at least three days, but the screening was not necessarily organized seven days a week, so this change could explain the improvement in the second-test coverage, by the reduction of newborns lost to follow-up between the first and second screening tests. Logically, this change in the protocol did not show similar consequences on the first-test coverage because the organization of the screening in the maternity wards allows at least one test to be performed before discharge, regardless of the length of the hospital stay. So, coverage rates still need to be improved to reach at least the benchmarks from the JCIH, while the length of the hospital stay in the FWB is favorable to the organization of an inpatient two-step screening. Indeed, other programs reached higher second-test coverage than in the FWB, for example, in the Milanese program (with outpatient procedure) or the Israeli program (with inpatient screening) [7,10].

Beyond screening test-coverage, the monitoring of the follow-up coverage is required. In the FWB, as the first and second-test coverage, the follow-up coverage rate also needs to be improved. In 2007, fewer than one out of two referred newborns had a documented follow-up and during the next five years, only between 60 and 69% had a documented follow-up. Advanced analysis showed that follow-up coverage was underestimated due to a lack of systematic data transmission of audiological assessment results, such as a diagnosis of HI. Analysis of other programs with an effective tracking system shows that the proportions of

newborns lost to follow-up was reduced to 16% (Milan) or less than 10% (Hessen) and reached the benchmark set by the JCIH (90% of the referred newborns should complete an audiological assessment by 3 months of age) [2,10,14]. In the program in the FWB, a centralized tracking system has been progressively implemented but was not yet working for all of the hospitals in 2012. As a consequence, ENT doctors should be more thorough to improve the completeness of collected data and parents should be systematically invited to perform their child's audiological assessment, after two inconclusive tests.

The monitoring of a UNHS program is not entire without the analysis of the pass-refer rate: indeed, a high refer rate implies the performance of more complementary tests and also raises the risk of newborns lost to follow-up, compromising the quality of the UNHS program. In our study, less than 88% of the tested newborns had a bilateral pass result at the first screening test. The pass rate showed a slight improvement from 85% in 2007 to 88% in 2010 and thereafter it has stabilized. Improvements in the pass rate are in line with other programs [7,18] and could be explained by improved skills, even when devices are automated, because most of the professionals had never performed a neonatal hearing test before the implementation of the program. When the test is performed also has consequences on the pass-refer results, particularly when the screening is performed early [19–21]. In our study, association between the day of the test and the screening outcomes was not investigated.

Compared to other UNHS protocols using OAE in newborns without risk factors, the pass rate in the FWB was slightly lower than local Italian programs or the Albanian program (between 87.8% and 91%) [8,22,23]. In our study, comparisons of the pass-refer outcomes did not aim to assess devices performance but tried to monitor the hearing screening process and to compare it with other programs. We notice that comparisons are weakened due to methodological difficulties: protocols are different regarding when the tests are carried out (especially the second test that could be performed as an inpatient or outpatient procedure), the number of tests allowed (sometimes repeated tests are considered as one procedure or a step in the program [24]), outcome of the screening (unilateral or bilateral "pass" outcomes are looked for [18]), devices (automated or not), population ("well-babies"; "NICU"; "healthy newborns"; "all newborns") and denominator used (eligible population, newborns tested). Furthermore, comparability of the pass-refer rate for the second test is challenging because few protocols organize a two-step screening with exclusively OAE: most of the time, in order to reduce referral rate, OAE are combined with automated auditory brainstem response (AABR) and some protocols use exclusively AABR [25]. We also noticed that the protocol is not always strictly implemented, but is sometimes locally adapted; thus the reality of the outcome measures could not exactly reflect the official protocol.

Despite these methodological arguments, referral rate is a commonly used outcome for UNHS assessment, particularly the referral rate between screening and audiological assessment and it is well-documented that a two-step protocol contributes to lowering the proportion of newborns referred for further evaluation, compared with a one-step protocol [19]. In our study, at the end of the screening process, 2.4% of the eligible population were referred to an ENT doctor. Again, due to differences between protocols, comparison is difficult. But if this outcome is used as a general quality benchmark, summarizing the UNHS organization, it highlights a rather high referral rate in the program in the FWB, even if it was lower than the 4% recommended by the JCIH [2]. The two-step protocol in the FWB based exclusively on AOAE is one of the major reasons for this high referral rate: programs using AABR in one or two steps of the screening generally present lower referral rates [25,26]. In the FWB, it was decided to favor the AOAE technique because it is easier to perform and less expensive, at the price of a higher referral rate. At this time, we should investigate whether keeping this technique still affects the quality of the program (by increasing referral rates and risk of newborns lost to follow-up). We should consider whether changing the AOAE in favor of AABR would improve program outcomes or, on the contrary, would jeopardize its success (by a decrease in skilled professionals performing the screening or by a higher testing time for example).

Screening all newborns is one of the challenges of a UNHS program. However, some newborns are not tested, sometimes due to explicit reasons. In the FWB, we observed that the parental refusal rate of the hearing screening sharply decreased between implementation of the program in 2007 (1.07%) and 2012 (0.25%), and it is now considered very marginal, which is a sign of acceptance of hearing screening by the parents. However our results also suggest that the outcome "another institution/ENT doctor" could be an informal refusal by the parents who do not feel comfortable about explicitly refusing the screening. In this case, the refusal rate would be underestimated. There is no possibility to confirm this hypothesis, in the absence of data collection of hearing screening performed outside of the FWB program. It is also impossible to confirm that the screening was actually performed when "Flemish UNHS" was noted. This outcome measure tripled between the first and second year: joining of some Brussels hospitals, which is a bilingual area, could explain this increase. The data collection form allowed the three outcomes to be filled in separately (refusal; another institution/ENT doctor; Flemish UNHS) but they are likely related, especially if the parents were not asked if they are willing to perform the test outside the hospital and the FWB program.

If we consider refusals alone, rates in the FWB were similar to those observed in other programs, for example in Western Australia (0.4%), the Ligurian program (0.6%) or in the USA (between 0.4% and 3%) [6,9,22,27]. But if we consider refusals, testing in another institution/by another ENT doctor and by the Flemish UNHS as a combined outcome, the rate in the FWB was similar to the Flemish program (2.6%) or the program in Israel (3.9 or 2.1% depending on the year) [7,28].

The objectives of this study were to present the main results and outcomes of the UNHS program in the FWB since its beginning and to analyze changes, year by year, between 2007 and 2012. Analysis of the changes was based on the year of birth; this method is arbitrary and did not take account of previous experience of UNHS before the program in the FWB. So, results of 2007 are composed of both experienced and non-experienced maternity wards. Despite this, all outcome measures were quite low in the first year, in particular when they are compared to the benchmarks from the JCIH, but they improved in the second year. This improvement continued for the majority, but at some point (in 2009 or 2010 principally) these levels plateaued or even declined. Obviously, it is a challenge to continue improving outcome measures or, at least, to stay at the same level as in the preceding year. Organizational difficulties or workload related to the program could reduce the motivation and commitment of the staffs, particularly after a few years. The screening tests possibly being performed more routinely and a decline in the support from management or from the coordinating agency could explain these outcome measures.

Our study highlighted a recurrent weakness about data quality: lack of accurate information and lack of transmission of the information have been mentioned. This weakens the assessment of the entire program. In order to correct this weakness, the data management, based on regular contacts between the professionals and the coordinating agency and a closer monitoring of the data, should be re-enforced, as the awareness of the professionals about

data quality. Also, outcome measures about the process (from the local organization) should be integrated to develop a comprehensive assessment.

5. Conclusion

Outcome measures for the UNHS program in the FWB were slightly lower than the benchmarks released by the JCIH. Nevertheless, despite the difficulty in comparing with other programs, some outcomes from the program in the FWB were similar or higher, while some outcomes (such as screening coverage or follow-up coverage) clearly needed to be improved. Completeness and quality of data particularly deserve the attention of the professionals involved in the program. The evolution of outcome measures since its implementation and over six years was positive during the first years but most of the outcome measures decreased or at least did not increase further after a point. The motivation and commitment of the professionals has to be supported in a variety of ways to improve the outcomes and thus quality of the program.

Conflict of interest

The authors declare that they have no competing interests.

Funding source

The Fédération Wallonie-Bruxelles funded the newborn hearing screening program and had no other involvement in this article.

Authorship

Bénédicte Vos participated in the design of the study, performed the data management and the statistical analysis, and drafted the manuscript. Raphaël Lagasse and Alain Levêque participated in the design of the study, in the editing of the manuscript and critically reviewed the text. All authors read and approved the final manuscript.

Acknowledgements

The authors would like to thank the maternity hospitals and their staff for participating in the program and the neonatal blood screening centers. They also would like to thank ISO Translation for the editing of the manuscript.

References

[1] European Consensus Statement on Neonatal Hearing Screening, Finalized at the European Consensus Development Conference on neonatal hearing screening Milan, 15–16 May 1998, Acta Paediatr. 88 (1999) 107–108.
[2] Joint Committee on Infant Hearing, Year 2007 position statement: principles and guidelines for early hearing detection and intervention programs, Pediatrics 120 (2007) 898–921.
[3] US Preventive Services Task Force, Universal screening for hearing loss in newborns: US Preventive Services Task Force recommendation statement, Pediatrics 122 (2008) 143–148.
[4] World Health Organization, Newborn and infant hearing screening. Current issues and guiding principles for action, Geneva, Switzerland. 9–10 November 2009, Outcome of a WHO informal consultation held at WHO headquarters, 40 (2010).
[5] A. Erenberg, J. Lemons, C. Sia, D. Trunkel, P. Ziring, Newborn and infant hearing loss: detection and intervention. American Academy of Pediatrics. Task force on newborn and infant hearing, 1998–99, Pediatrics 103 (1999) 527–530.
[6] H.D. Bailey, C. Bower, J. Krishnaswamy, H.L. Coates, Newborn hearing screening in Western Australia, Med. J. Aust. 177 (2002) 180–185.
[7] P. Gilbey, C. Kraus, R. Ghanayim, A. Sharabi-Nov, S. Bretler, Universal newborn hearing screening in Zefat, Israel: the first two years, Int. J. Pediatr. Otorhinolaryngol. 77 (2013) 97–100.
[8] S. Hatzopoulos, B. Qirjazi, A. Martini, Neonatal hearing screening in Albania: results from an ongoing universal screening program, Int. J. Audiol. 46 (2007) 176–182.
[9] A.L. Mehl, V. Thomson, The Colorado newborn hearing screening project, 1992–99: on the threshold of effective population-based universal newborn hearing screening, Pediatrics 109 (2002) E7.
[10] G. Pastorino, P. Sergi, M. Mastrangelo, P. Ravazzani, G. Tognola, M. Parazzini, et al., The Milan Project: a newborn hearing screening programme, Acta Paediatr. 94 (2005) 458–463.
[11] B. Vos, R. Lagasse, A. Leveque, The organisation of universal newborn hearing screening in the Wallonia-Brussels Federation, B-ENT (Suppl. 21) (2013) 9–15.
[12] Joint Committee on Infant Hearing, American Academy of Audiology, American Academy of Pediatrics, American Speech-Language-Hearing Association, Directors of Speech, Hearing Programs in State Health and Welfare Agencies, Year 2000 position statement: principles and guidelines for early hearing detection and intervention programs, Pediatrics 106 (2000) 798–817.
[13] J. Jakubikova, Z. Kabatova, M. Zavodna, Identification of hearing loss in newborns by transient otoacoustic emissions, Int. J. Pediatr. Otorhinolaryngol. 67 (2003) 15–18.
[14] K. Neumann, M. Gross, P. Bottcher, H.A. Euler, M. Spormann-Lagodzinski, M. Polzer, Effectiveness and efficiency of a universal newborn hearing screening in Germany, Folia Phoniatr. Logop. 58 (2006) 440–455.
[15] P. Schmidt, M. Leveque, J.B. Danvin, B. Leroux, A. Chays, Systematic hearing screening for newborns in the Champagne-Ardennes region: 32,500 births in 2 years of experience, Ann. Otolaryngol. Chir. Cervicofac. 124 (2007) 157–165.
[16] B. De Capua, D. Costantini, C. Martufi, G. Latini, M. Gentile, C. De Felice, Universal neonatal hearing screening: the Siena (Italy) experience on 19,700 newborns, Early Hum. Dev. 83 (2007) 601–606.
[17] A.K. Rohlfs, T. Wiesner, H. Drews, F. Muller, A. Breitfuss, R. Schiller, et al., Interdisciplinary approach to design, performance, and quality management in a multicenter newborn hearing screening project: introduction, methods, and results of the newborn hearing screening in Hamburg (Part I), Eur. J. Pediatr. 169 (2010) 1353–1360.
[18] M. Leveque, P. Schmidt, B. Leroux, J.B. Danvin, T. Langagne, M. Labrousse, et al., Universal newborn hearing screening: a 27-month experience in the French region of Champagne-Ardenne, Acta Paediatr. 96 (2007) 1150–1154.
[19] O.V. Akinpelu, E. Peleva, W.R. Funnell, S.J. Daniel, Otoacoustic emissions in newborn hearing screening: a systematic review of the effects of different protocols on test outcomes, Int. J. Pediatr. Otorhinolaryngol. 78 (2014) 711–717.
[20] E. Berninger, B. Westling, Outcome of a universal newborn hearing-screening programme based on multiple transient-evoked otoacoustic emissions and clinical brainstem response audiometry, Acta Otolaryngol. 131 (2011) 728–739.
[21] M. Lupoli Lda, L. Garcia, A.R. Anastasio, A.C. Fontana, Time after birth in relation to failure rate in newborn hearing screening, Int. J. Pediatr. Otorhinolaryngol. 77 (2013) 932–935.
[22] M.G. Calevo, P. Mezzano, E. Zullino, P. Padovani, G. Serra, STERN Group, Ligurian experience on neonatal hearing screening: clinical and epidemiological aspects, Acta Paediatr. 96 (2007) 1592–1599.
[23] A. Ciorba, S. Hatzopoulos, M. Busi, P. Guerrini, J. Petruccelli, A. Martini, The universal newborn hearing screening program at the University Hospital of Ferrara: focus on costs and software solutions, Int. J. Pediatr. Otorhinolaryngol. 72 (2008) 807–816.
[24] H.C. Lin, M.T. Shu, K.C. Chang, S.M. Bruna, A universal newborn hearing screening program in Taiwan, Int. J. Pediatr. Otorhinolaryngol. 63 (2002) 209–218.
[25] G.X. Papacharalampous, T.P. Nikolopoulos, D.I. Davilis, I.E. Xenellis, S.G. Korres, Universal newborn hearing screening, a revolutionary diagnosis of deafness: real benefits and limitations, Eur. Arch. Otorhinolaryngol. 268 (2011) 1399–1406.
[26] J.I. Benito-Orejas, B. Ramirez, D. Morais, A. Almaraz, J.L. Fernandez-Calvo, Comparison of two-step transient evoked otoacoustic emissions (TEOAE) and automated auditory brainstem response (AABR) for universal newborn hearing screening programs, Int. J. Pediatr. Otorhinolaryngol. 72 (2008) 1193–1201.
[27] A. Young, E. Andrews, Parents' experience of universal neonatal hearing screening: a critical review of the literature and its implications for the implementation of new UNHS programs, J. Deaf Stud. Deaf Educ. 6 (2001) 149–160.
[28] E. Van Kerschaver, A.N. Boudewyns, L. Stappaerts, F.L. Wuyts, P.H. Van de Heyning, Organisation of a universal newborn hearing screening programme in Flanders, B-ENT 3 (2007) 185–190.