The organisation of universal newborn hearing screening in the Wallonia-Brussels Federation

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Abstract. The organisation of universal newborn hearing screening in the Wallonia-Brussels Federation. Objective: A universal newborn hearing screening programme based on the voluntary participation of maternity hospitals has been implemented in the Wallonia-Brussels Federation since November 2006. This paper presents the results of this programme and its evolution since its implementation (2007-2011). Method: Two-step screening by automated otoacoustic emissions is performed on newborns without risk factors for hearing loss and, if abnormal responses or risk factor(s) are found, auditory brainstem response audiometry is performed. Descriptive analyses of the eligible population, coverage rates and prevalence of hearing loss were presented by year of birth (2007-2011) and globally. Results: Over five years, the first screening test coverage rate increased to almost 95%, 2.4% of the newborns without risk factors were referred for auditory brainstem response audiometry and 42.6% of the newborns referred were lost to follow-up. Of the newborns with risk factor(s) for hearing loss, 6.3% were not tested, 87.4% had normal bilateral hearing and 3.7% were lost to follow-up. Between 2007 and 2011, hearing impairment was reported in 3.18‰ newborns eligible for the programme, whatever the kind of hearing loss. Conclusions: Like many other countries and areas, the Wallonia-Brussels Federation has implemented a universal newborn hearing screening programme. The results of this programme evolved favourably during the period 2007-2011, even if some efforts had to be made to improve the follow-up of the referred newborns and data quality.

Introduction

Permanent childhood hearing loss is a public health concern, particularly because of its severe consequences and high prevalence. Although estimating prevalence depends on many factors, in particular the definition adopted by the authors, permanent childhood hearing loss is one of the most common congenital anomalies: one to three newborns per thousand in the well-baby nursery population are affected by severe or profound bilateral hearing loss and this prevalence is ten times more common in the neonatal IC-unit population, where significant bilateral hearing loss is seen in 2-4% in this specific group of newborns.¹²

The children in question are severely affected by permanent bilateral hearing loss and, without appropriate intervention, deaf children’s cognition and socio-emotional development lag behind those of their hearing peers.¹³ It is known that the first six months of life are particularly important for the early development of language and speech: children in whom hearing loss is identified by six months of age have better language outcomes than children identified later, whatever the degree of hearing loss.¹³ As a result, the Joint Committee on Infant Hearing recommended that hearing loss should be confirmed before 3 months of age and that hearing-impaired children should receive appropriate interventions before the age of 6 months.³

When hearing is not screened, the average age of diagnosis for severe to profound hearing loss is higher than recommended (30 months in the USA and 39 months in Germany, whatever the degree of hearing loss).⁴ Universal newborn hearing screening is therefore advisable in preference to programmes

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targeting newborns with risk factor(s). Given this consideration, different national and international groups of experts have been recommending the organisation of universal newborn hearing screening programmes (UNHSP) for almost 20 years now. Despite these recommendations, there was no coordinated programme in the Wallonia-Brussels Federation (FWB, the French-speaking part of Belgium) until November 2006. At that time, the FWB proposed a UNHSP for the maternity hospitals located in Brussels and Wallonia and data were collected starting on 1 January 2007.

The objective of this study is to present the results of the UNHSP in the FWB and its evolution since implementation (2007-2011).

Materials and methods

Protocol and organisation of the UNHSP

The UNHSP was implemented in the maternity hospitals of the FWB that agreed to participate on a voluntary basis. A protocol was released in November 2006 by a group of experts in ENT, public health and paediatrics. It specified that newborns without risk factors for hearing loss should undergo a two-stage screening programme: the first step is an automated otoacoustic emissions test (AOAE) at the maternity unit on the second or third day of life. When there is an abnormal response (unilaterally or bilaterally), the second step is a control performed the day after using the same test. Children who fail the second test for one or both ears are referred for audiological assessment using auditory brainstem response audiometry (ABR) within two weeks, but preferably before hospital discharge. Newborns with at least one risk factor (Table 1) undergo immediate audiological assessment by ABR, if possible before being discharged from hospital. In the case of newborns leaving the hospital before a hearing test has taken place (screening or audiological assessment), an appointment is made with the parents for an outpatient test during the four weeks thereafter. This protocol is implemented locally in line with the hospitals’ human and financial resources. Each hospital is free to decide on its own approach to organisation (professionals who conducted the screening, number of days a week allocated to the screening programme…) in accordance with the protocol.

Data collection

Data were collected on paper records at the beginning of the UNHSP with the collaboration of the three neonatal metabolic screening centres of the FWB: data for the screening (otoacoustic emissions tests) were written on the reverse of the Phenylketonuria test form and the results of the audiological assessment were collected using a specific form. These results were stored in specific databases by the metabolic screening centre associated with the hospital and analysed anonymously by the Coordinating Agency on an annual basis. Since 2011, data collection using a computer system with an Internet connection has gradually replaced the paper records. In this system, the Coordinating Agency processes on a daily basis the data collected electronically. Data collected (by paper or by computer) are linked to the screening tests and the audiological assessment; the political organisation of the preventive health care system in Belgium means that no information is collected at the moment from the hearing rehabilitation services.

Table 1

<table>
<thead>
<tr>
<th>Risk factors related to</th>
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<tbody>
<tr>
<td>the prenatal period</td>
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<tr>
<td>- Family history of hereditary hearing loss</td>
</tr>
<tr>
<td>- Consanguinity (1st degree)</td>
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<tr>
<td>- In-utero infection:</td>
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<tr>
<td>o cytomegalovirus</td>
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<tr>
<td>o toxoplasmosis</td>
</tr>
<tr>
<td>o herpes</td>
</tr>
<tr>
<td>o rubella</td>
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<tr>
<td>o syphilis</td>
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<tr>
<td>- Poisoning (alcohol, drugs) by the mother during pregnancy</td>
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Recall system
Untested newborns or newborns with anomalous results are recalled for outpatient testing. The recall system depends on the kind of data collection: when data are collected on paper, hospitals are expected to contact the parents three times; when data are collected by computer, the Coordinating Agency is required to send two recall letters (on the 15th and 30th days of life) and the hospital is required to contact the parents a third time by phone (on about the 45th day of life).

Funding
The UNHSP is funded in different ways: in 2007, the FWB paid 5 euros to participating hospitals per tested child without a risk factor whether one or two tests were performed, and maternity hospitals were allowed to charge the parents up to 10 euros. Most social security organisations reimburse parental participation in the screening procedures. In the case of children with a risk factor or children referred after two AOAEs to an ENT doctor (audiological assessment), the financial arrangements depend on the Federal Sickness Fund. Data collection is also financed by the FWB but screening and diagnosis devices are not funded by the FWB: hospitals must therefore buy their own devices and they are responsible for training users. Since 2009, all financing from the FWB has been indexed annually.

Inclusion criterion
Due to the approach with voluntary participation, some hospitals joined the UNHSP at the beginning of the programme in November 2006, while others joined in later years. In this paper, the data include newborns screened from the outset of protocol implementation in the corresponding maternity hospital, not necessarily since 1 January 2007.

Variables used and data analysis
The results were presented globally for the first five years of the programme as proportions. They were also set out by year of birth from 2007 to 2011. The eligible population and the proportions of newborns with or without risk factors for hearing loss were presented first. The coverage rates were then presented separately for the first screening test and for follow-up after referral to the ENT specialist. These rates were calculated only for the children who required the test in question.

A “hearing status” variable was established. It covered four groups of children: newborns with bilateral normal hearing, untested newborns, lost to follow-up (after at least one inconclusive test) and hearing-impaired newborns. The kind of hearing loss was presented separately for unilateral and bilateral impairments on the basis of the following items: conductive, auditory neuropathy, perceptive (temporary, permanent, unspecified) or unspecified hearing loss.

Analysis was performed with Stata 12 I.C. software.

Results
At the beginning of 2007, 23 hospitals were participating in the UNHSP; 19 hospitals joined the programme during the year 2007, two more in 2008 and one more in 2010. At the end of 2011, all the maternity hospitals of the FWB had agreed to implement the protocol, with the exception of three institutions. Due to this progressive involvement, the annual number of children newborns covered by the programme rose from 38,265 (in 2007) to more than 52,000 (since 2010) (+35%) (Table 2).

In 2007, the UNHSP covered 66.5% of the children born in the FWB and it evolved progressively to cover almost 90% of newborns in the FWB.

The parental refusal rate fell steadily: from 1% at the beginning of the programme to 0.3% five years later. The proportion of parents who wanted the hearing test in another institution or from another ENT doctor rose sharply from 1% (2007) to 2.4% (2011). More than half of these parents (58.3%) wanted the test to be performed by the Flemish Maternal and Child Health Agency (Kind en Gezin). In the case of parental refusal or a stated intention to arrange for testing outside the UNHSP of the FWB, the newborns were not considered to be part of the programme population (2.6% of the children born in participating hospitals) (Table 2).

Between 2007 and 2011, 233,397 newborns in the FWB entered the UNHSP. Due to the progressive involvement of the hospitals in the programme, the number of children in the programme population increased, especially between 2007 and 2008 (+22%), and gradually stabilised between 2009 and 2011. In the programme population, 217,258 newborns (93.1%) had no risk factor for hearing loss.
and 16,139 (6.9%) had one or more risk factors. The proportion of newborns with risk factor(s) for hearing loss increased gradually, doubling over the period 2007-2011 from 4.1% to 8.6% (Table 2).

The 217,258 newborns without risk factors for hearing loss constituted the screening group and entered the two-step screening protocol based on AOAE performed at the maternity unit. The first screening test coverage rate was 89.3% in the first year, increasing to 94.2% in 2010; since 2007, the coverage rates have constantly improved, except for 2011 (92.2%) (all newborns with the exception of the “untested” children – Figure 1). During the five years studied, the overall coverage rate for the first screening test was 92.7% (n = 201,426). After screening tests, 5,260 newborns were referred for audiological assessment, resulting in a referral rate of 2.4%. Data on the follow-up of these referred children showed that some of them were lost to follow-up: hearing status was unknown for 42.6% of the referred newborns (either the children were untested after the two AOAEs or no diagnosis was made after audiological assessment). This figure varied from year to year: from 57.3% in 2007 to 36.2% in 2011.

In the newborn population with no risk factors who entered the UNHSP, hearing loss was reported for 318 infants (1.46‰), with major variations from year to year (0.67‰ in 2007; 1.94‰ in 2009). The proportions of children with normal bilateral hearing evolved positively over the years and this improvement was correlated with the fall in numbers of newborns who were untested and lost to follow-up (Figure 1).

During the first five years of the UNHSP, 16,139 newborns were identified with at least one risk factor for hearing loss and were directly referred to an ENT doctor for audiological assessment. Of these children, 1,008 were not tested (6.3%), 602 were lost to follow-up before a diagnosis was made (3.7%), 14,104 had normal bilateral hearing (87.4%) and 425 had bilateral or unilateral hearing loss. In this specific group of newborns with risk factor(s), the proportion of hearing-impaired children varied considerably from year to year: from 0.5% in 2007 to 3.7% in 2009. By contrast,
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The proportions of newborns with bilateral normal hearing remained quite stable during the years studied (Figure 1).

Between 2007 and 2011, hearing impairment was reported for 3.18‰ of newborns in the programme, irrespective of the kind of hearing loss and whether hearing loss was unilateral or bilateral. Conductive hearing impairment (temporary, permanent or unspecified) was reported in 394 newborns (1.69‰) and perceptive hearing impairment was reported in 124 newborns (0.53‰); for 222 newborns (0.95‰), the kind of impairment was not specified in the data (Table 3).

Discussion

The Task Force on Newborn and Infant Hearing (1999) has identified five elements that are essential for an effective UNHSP: initial screening, tracking and follow-up, identification, intervention and evaluation.1

Performing initial screening in maternity hospitals is a good way of recruiting newborns and of ensuring that a high proportion of them undergo neonatal screening.11 As in many other UNHSPs,12-14 this approach has been implemented in the FWB, where it is based on the voluntary participation of the hospitals. With the improvement in the hospital participation rate and the increase in the UNHSP population, the acceptance of hearing tests by parents improved. Indeed, the proportion of parents refusing the hearing test fell from 1% to 0.3% over five years. These parental refusal rates were comparable to those in other programmes,12-14 even when a financial contribution was required from parents in the FWB. In this study, we did not look at whether the parents were aware that there was a charge for screening, and so no link could be made between financial contribution and the refusal rate. From a public health point of view, every newborn should be screened in a UNHSP to ensure that all hearing-impaired children benefit from early intervention. However, ethically speaking, parents should be given the opportunity to refuse the test after full information has been supplied.

On the other hand, the proportion of parents who preferred the test to be performed outside the programme more than doubled during the five years studied (from 1 to 2.4%). This is explained by the increasing number of participating hospitals (especially during 2007 and 2008) in Brussels (a bilingual area) where Flemish-speaking parents preferred their child to be monitored by the Flemish Maternal and Child Health Agency. However, the item “performing the test outside the maternity hospital” also included newborns referred to another institution for audiological assessment (due to the absence of an ABR device in the maternity hospital), not exclusively at the parents’ request.

Coverage rates are important elements for the monitoring of UNHSPs. In the FWB, the annual first-test coverage rates have improved gradually from 89.3% to 94.2%. However, a decrease was observed in the data for 2011. This was caused by a lack of systematic data communication from two hospitals (and not a fall in the number of hearing tests). Regular data management, such as computer data transmission, is therefore required to resolve this data problem in real time. Early discharges from hospital may explain low coverage rates and so outpatient appointments should be arranged when the test is not performed before discharge. These coverage rates need to improve to achieve the recommended coverage of at least 95% of newborns (before one month of age).3

The referral rate in the FWB programme is lower than the recommended 4% and is therefore acceptable.3 However, this referral rate should be

Table 3

Hearing losses identified by the UNHSP in the FWB among children born between 2007 and 2011 (n = 233,397)

<table>
<thead>
<tr>
<th></th>
<th>Unilateral &amp; Bilateral</th>
<th>Unilateral</th>
<th>Bilateral</th>
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<tbody>
<tr>
<td>n</td>
<td>%</td>
<td>n</td>
<td>%</td>
</tr>
<tr>
<td>Perceptive</td>
<td>743 3.18</td>
<td>357</td>
<td>1.53</td>
</tr>
<tr>
<td>Auditory neuropathy</td>
<td>124 0.53</td>
<td>41</td>
<td>0.18</td>
</tr>
<tr>
<td>Conductive (temporal)</td>
<td>197 0.84</td>
<td>105</td>
<td>0.45</td>
</tr>
<tr>
<td>Conductive (permanent)</td>
<td>26 0.11</td>
<td>14</td>
<td>0.06</td>
</tr>
<tr>
<td>Conductive (unspecified)</td>
<td>171 0.73</td>
<td>101</td>
<td>0.43</td>
</tr>
<tr>
<td>Hearing loss - unspecified</td>
<td>222 0.95</td>
<td>96</td>
<td>0.41</td>
</tr>
</tbody>
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analysed with caution: some newborns thought to be lost to follow-up between the first and the second screening test would have been referred if they had been tested and so the referral rate has been underestimated. Screening results in a UNHSP should therefore tend to achieve low proportions of referral rates for audiological assessment: high referral rates may reflect the re-organisation of ENT outpatient clinic due to a higher workload and require specific skills to be represented in paediatric audiology departments. High referral rates also exacerbate the risk of losing newborns to follow-up.

The proportions of newborns lost to follow-up between screening and audiological assessment should also be monitored. In the FWB programme, this specific element should be improved: even if the percentages of loss to follow-up have fallen since 2007, fewer than 70% of the referred newborns were known to have undergone audiological assessment, whereas the recommended minimum is 90% (by three months of age). Some of the children classified as being lost to follow-up by the UNHSP may have undergone the appropriate audiological assessment and the result may simply not have been included in the database: it would be better to describe these children as being “lost to documentation”. It is impossible to distinguish between newborns lost to documentation and those lost to follow-up and the proportion of newborns that are actually lost to follow-up is certainly an overestimate. The centralised data collection and tracking system that has been progressively implemented since 2011 will reduce the number of children lost to follow-up and lost to documentation by a more systematic approach to data management. Moreover, this new system and active tracking will enhance collaboration between professionals in the hospitals and the Coordinating Agency and therefore enhance the overall quality of the programme.

In addition to initial screening and tracking and follow-up, the identification of hearing loss is one of the major objectives of a UNHSP. Hearing loss (bilateral or unilateral, whatever the kind or the degree of hearing loss) was reported in 3.18‰ of newborns. The prevalence of hearing loss identified by the UNHSP concurs with other programmes and studies. However, data quality is an area requiring particular attention: some information about hearing loss is incomplete (type or degree of deafness). Similarly, information about risk factors is incomplete: the increase in reported risk factors from 4.1% to 8.6% was probably due to improvements in the communication of this information and it could be a positive effect of the training given by the Coordinating Agency. Moreover, this data collection may be biased: risk factors have to be reported only for children with risk factors and no data was collected for the item ‘no risk factor’.

The Task Force on Newborn and Infant Hearing has stated that intervention is the fourth prerequisite for an effective UNHSP. At the moment, the UNHSP in the FWB does not collect any data about intervention for hearing-impaired children. This absence of data collection could be explained by the political organisation of the Belgian healthcare system: preventive services are funded by the Communities and rehabilitation services by the Regions or the Federal Sickness Fund. From a public health point of view, this absence of data collection from rehabilitation services weakens the UNHSP by making global evaluation impossible. The other components (initial screening, follow-up, identification of hearing loss) of the UNHSP are monitored on an ongoing and annual basis by the Coordinating Agency.

**Conclusion**

By implementing this UNHSP, the FWB complied with the international guidelines recommending early hearing tests. The ultimate objectives of a UNHSP are to identify hearing loss during the first months of life and to initiate early and adequate intervention. Our study showed that the results of the UNHSP in the FWB have evolved favourably over the five years, even if some efforts have still to be made to improve the follow-up of the referred newborns and data quality, in particular with relation to diagnosis. Unfortunately, the UNHSP in the FWB did not collect data from hearing rehabilitation services and so a global evaluation of the UNHSP is not possible. However, the voluntary participation of maternity hospitals did not seem to be a constraint and choices made in the protocol (kind of test, early screening) have resulted in an effective programme in the FWB.

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References


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