Introduction and background information

Permanent neonatal hearing impairment occurs in approximately 1.4 per 1,000 newborns, affecting about 80 children every year in Flanders (Belgium). Children with a hearing impairment may have major difficulties in the fields of social and emotional development, motor skills, personality, reading comprehension, and speech and language development. Early hearing screening, with investigation before the age of 3 months and treatment before 6 months, considerably improves the prospects of children born deaf. The Joint Committee on Infant Hearing (JCIH) and the European Consensus Statement formulated the basic guidelines for hearing screening. This study describes the conception and elaboration of the programme for neonatal hearing screening in Flanders. It also reports on the results and the various analyses conducted to evaluate and adjust the programme. Finally, it identifies new socio-demographic risk factors for neonatal hearing impairment.

Hearing impairment can result from both the congenital and acquired malfunctioning of structures within the auditory system and it is the result of complex interactions between genetic, toxicological and social factors. The American Academy of Paediatrics (AAP) has described a standard list of risk factors for hearing impairment, mostly related to health and physical characteristics or to hereditary history.

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The Flemish universal hearing screening programme was conceived and implemented in the well-structured government organisation ‘Kind en
Gezin’ (K&G – Child and Family), which is responsible for health, child care, parenting support and well-being in young children until school age and covers the entire population of babies and young children in Flanders.

The programme was established in consultation with all the Flemish university ENT departments and with the certified centres of expertise which form the 24 referral centres. They are the partners in, and co-owners of, the screening programme.

The programme integrates screening, referral, specialised investigation, rehabilitation, family guidance and reporting in a single protocol.

All data and results are collected in a central K&G database. This makes it possible to monitor the programme and prevent drop-out, to evaluate the data and link them to socio-demographic characteristics.\textsuperscript{6} The screening uses the ‘Algo’ AABR screener (Natus Medical Inc San Carlos, Ca, US) and it leads to a ‘pass’ or a ‘refer’ result.

From 1998 onwards, the new hearing screening programme was implemented in all Flemish regions and incorporated in the normal preventive programme of the K&G nurses. All babies without a ‘pass’ result after a two-step screening procedure were referred to a certified referral centre. These centres also faced a new challenge in the field of medical and audiological diagnostics, early home intervention and rehabilitation for very young children.

**Analysis of the screening results and outcome data**

1. **The coverage of the hearing screening programme and related aspects** (Figure 1)

In the period to 2008, hearing screening was offered to 600,933 newborns, or 97.86% of newborns. K&G nurses tested 92.53% of the babies in a regional K&G centre, in a baby welfare clinic or during a home visit. Maternity wards tested 3.33% of the babies prior to discharge. These data remained stable over the years during the screening programme.

Some parents objected to the test or were not interested. The refusal rate decreased from about 3% in the first years to approximately 1% from 2004 onwards.

Only 1.60% of newborns could not be contacted.

In 1999, 3,072 babies were born into underprivileged families in the Flemish Region. The total target group approached in the underprivileged group was 91.54%. Kind & Gezin screened 85.18% with the Algo test. This is significantly lower than the percentage in the non-underprivileged population group. Data from 2011 confirm this difference in the screening levels linked to family origin. The screening level for newborns in underprivileged families continues to be a cause for concern.

The average age of the babies at screening was about four weeks in the period 2003-2004. Medical and auditory evaluation usually took place before the age of 3 months and intervention always began before the age of 6 months.

2. **The hearing screening referral rate and confirmation rate**

The referral rates and the confirmation rates were different with the Algo Portable (1999-2006) and with the Algo3i (2007-2012).

With the Algo Portable, 99.7% of the babies had a ‘pass’ result after the first or second test. 2.73‰ of the screened babies had two tests without a pass result and they were all referred to a certified specialised centre (Figure 2). The screening result was confirmed in almost 77% of the referred babies.

![Figure 1](https://example.com/figure1.png)

*Figure 1*  
Hearing screening offered to Flemish babies in the period to 2008.
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3. Studies looking at the optimal screening age

Before the universal hearing screening programme was implemented in Flanders in 1997, the optimal screening age was based on the age at which test duration was shortest: four weeks. Once extensive screening data were available, further investigation of the results was conducted to determine the optimal age for screening more accurately.

Studies of the screening data for the years 2003 and 2004, 2005 and 2007 resulted in the same conclusion: the later screening is performed, the higher the incidence of ‘failed’ and ‘refer’ test results. These differences are statistically significant. The number of ‘refer’ tests was significantly higher in the first week of life (Figure 4).

When the Algo3i device was introduced from 2007 onwards, the number of referrals for temporary impairment increased tremendously, pushing the referral rate after two tests up to 7.4‰. The confirmation rate for bilateral HI exceeding 40 dB was 1.24‰, an average of 86 a year (Figure 3).

The technical differences between the Algo Portable and Algo3i devices explain the increase in referrals. The Algo3i is more sensitive and it detects ‘temporary hearing loss’ by OME more frequently. However, this issue is outside the scope of our discussion of a hearing screening programme for infants.

The increased referral rate has a major impact on the daily workload for all involved, parental concern and costs.

Hearing loss exceeding 40 dB was confirmed in 58.87%, and 62.25% of these babies had a bilateral loss. The confirmation rate for bilateral severe hearing impairment after screening with the Algo Portable is therefore 1.01 per thousand, an average of 95 babies per year. Hearing loss was limited in only 13.15% of the referred babies. Another 4.71% of babies had a temporary hearing problem and 23% had no hearing impairment at all.

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4. Study of environmental and socio-demographic factors

Environmental and socio-demographic factors were investigated in the entire population of term babies in 2003 and 2004 (114,479 screened babies) to study any relationship with test results and congenital hearing impairment (CHI). The study revealed some interesting new findings about environmental factors.

The prevalence rates for CHI did not differ significantly between urban and rural areas. However, temporary hearing impairment was four times more prevalent in urban areas than in rural areas.

Babies born between October and January have more temporary hearing problems. This finding was confirmed in the years 2005 and 2006.

Boys are significantly more affected by congenital hearing impairment than girls. This gender effect is more pronounced for temporary hearing problems.

It was also observed that 1.6% of the children with a failed test ultimately proved to have a CHI. This was why, from 2005 onwards, these children were systematically classified as being at risk and were also referred for further investigation.

In the population of term babies, the sensitivity of the Algo screening test (true positive rate) was 94.02% and specificity (true negative rate) was 99.96%.

The positive predictive value was 79.36% and the negative predictive value was 99.99%.

The most important conclusion was that the socio-demographic factors of gender, birth length and birth order in the family, initial feeding type (breast or formula), level of education and origin of the mother were found to be independent predictors of CHI.

The probability of CHI:
- is 1.7 times higher in boys than in girls
- falls by 0.86 for each extra cm in birth length
- is 1.14 times higher for each successive baby
- is 1.75 times higher in bottle-fed than in breast-fed children
- is 2.46 times higher in mothers with low education levels than in mothers with high education levels
- is 2.72 times higher in babies of Eastern Europe origin than for other parts of the EU.

The logistic regression analysis results in ‘sensitivity’ and ‘specificity’ rates for the combination of these factors of 63.2% and 64.2% respectively.

The factors of head circumference, delivery (caesarean or vaginal), dwelling (urban or rural), age of the mother at birth and gestational age of the term babies were not associated with CHI prevalence.

Discussion

Hearing screening in Flanders was offered to almost the entire population of newborns. It was possible to contact 98.4% of babies and about 96% of them were screened.

All the characteristics of the programme comply with the JCIH 2007 principles and coverage complies with the international standard of 95% for neonatal hearing screening. The screening programme referral rate was 2.7‰ with the Algo Portable device and 7.4‰ with the Algo3i. As far as we know, these low referral rates are quite unique.

Without an extended database and thorough registration of the screening data linked to personal and family information, the monitoring, evaluation and scientific investigation would not be possible up to this level of population studies.

The findings relating to the links between socio-demographic factors and the risk of neonatal hearing impairment are new. The classic AAP list of risk factors can now be widened to include a socio-demographic cluster that includes gender, birth order, birth length, feeding type, and the educational level and origin of the mother. Including the new risk factors could reduce the number of
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hearing-impaired babies without any known classic risk factors. The assessment of these additional factors could alert clinicians to the increased risk of newborn hearing impairment and emphasise the need for accurate follow-up. Moreover, this extended assessment may improve decision-making in medical practice and in screening policy.

The complex relationship of these socio-demographic factors with CHI is still poorly understood. Several factors seem to be linked and contribute to this correlation, such as poverty, smoking, working conditions, poorer hygiene, foetal alcohol syndrome, cytomegalovirus (CMV) or other infections, inadequate prenatal care, single parent, consanguinity, unemployment and quality of housing.

The origin and educational level of the mother, number of children, birth length (suboptimal diet, smoking) and food type (more bottle feeding in disadvantaged families) may be linked to poverty. The type of initial nutrition is, as a postnatal factor, correlated to a factor that is present prenatally.

In the domain of epigenetics, it is becoming increasingly clear that a genotype can express in different ways depending on the environmental conditions (such as child abuse, malnutrition, alcohol, smoking).

The evaluation of the studied risk factors indicates that underprivileged people are a key risk group for CHI who can be screened with the highest cost-effectiveness.

The strength of this study is undoubtedly the huge sample - the total population of term babies in a geographic region (Flanders) - including the missed and late-discovered CHI cases. All data were obtained using the same strict protocol and they were recorded in a central database system. After careful quality control of all variables, a cluster of risk factors for congenital deafness was identified by multivariate statistical analysis.

One limitation is that the information about the AAP risk factors in children was not adequate. These data could not be processed. There were also no data about potentially important factors such as smoking and alcohol consumption during pregnancy.

Conclusions

The ultimate goal of an early hearing screening programme is to reduce the dramatic consequences of neonatal hearing impairment on the further development, prospects and quality of life of the babies concerned.

The Flemish hearing screening programme is the first one to have reached almost the entire population, covering 96% of all babies with almost no loss to follow-up.

Given the age for the first hearing aid, the increase in the number of cochlear implants, mainstream school attendance and the impact on auditory receptive skills, speech intelligibility and reading level, we must acknowledge that the futures of children born deaf have improved dramatically since the start of the new hearing screening programme in Flanders. The social impact of the screening programme has been considerable. In addition, the improvements in diagnostics, treatment, multidisciplinary rehabilitation, educational opportunities and socio-emotional follow-up have led to a true transformation of the professional landscape.

Obviously, the results have been achieved only thanks to the exceptional dedication of the people involved with the children, and above all the parents. The impact on the family is, and remains, particularly strong and, obviously, CIs are not a universal remedy but there are better prospects now.

Together with the partners, we conclude that this integrated programme has achieved its goals.

The relationship between a cluster of socio-demographic factors and the increased risk of neonatal hearing impairment is a new insight that extends the classic list of risk factors. It constitutes a step towards a better understanding of the pathogenesis and possible preventive measures for congenital deafness.

However, more research is needed into important questions relating to aetiology and prevention, as well as potential risk factors for CHI in lifestyle and environment. More research is also needed into optimal treatment, rehabilitation and support, and into the long-term outcomes of CIs and their potential problems in adulthood.

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