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The economics of screening infants at risk of hearing impairment: An international analysis☆

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ABSTRACT

Objective: Hearing impairment in children across the world constitutes a particularly serious obstacle to their optimal development and education, including language acquisition. Around 0.5–6 in every 1000 neonates and infants have congenital or early childhood onset sensorineural deafness or severe-to-profound hearing impairment, with significant consequences. Therefore, early detection is a vitally important element in providing appropriate support for deaf and hearing-impaired babies that will help them enjoy equal opportunities in society alongside all other children. This analysis estimates the costs and effectiveness of various interventions to screen infants at risk of hearing impairment.

Methods: The economic analysis used a decision tree approach to determine the cost-effectiveness of newborn hearing screening strategies. Two unique models were built to capture different strategic screening decisions. Firstly, the cost-effectiveness of universal newborn hearing screening (UNHS) was compared to selective screening of newborns with risk factors. Secondly, the cost-effectiveness of providing a one-stage screening process vs. a two-stage screening process was investigated.

Results: Two countries, the United Kingdom and India, were used as case studies to illustrate the likely cost outcomes associated with the various strategies to diagnose hearing loss in infants. In the UK, the universal strategy incurs a further cost of approximately £2.3 million but detected an extra 63 cases. An incremental cost per case detected of £36,181 was estimated. The estimated economic burden was substantially higher in India when adopting a universal strategy due to the higher baseline prevalence of hearing loss. The one-stage screening strategy accumulated an additional 13,480 and 13,432 extra cases of false-positives, in the UK and India respectively when compared to a two-stage screening strategy. This represented increased costs by approximately £1.3 million and INR 34.6 million.

Conclusions: The cost-effectiveness of a screening intervention was largely dependent upon two key factors. As would be expected, the cost (per patient) of the intervention drives the model substantially, with higher costs leading to higher cost-effectiveness ratios. Likewise, the baseline prevalence (risk) of hearing impairment also affected the results. In scenarios where the baseline risk was low, the intervention was less likely to be cost-effective compared to when the baseline risk was high.

1. Background

Hearing impairment in children across the world constitutes a particularly serious obstacle to their optimal development and education, including language acquisition. According to a range of studies and surveys conducted in different countries, around 0.5–6 in every 1000 neonates and infants have congenital or early childhood onset sensorineural deafness or severe-to-profound hearing impairment [1–5]. Deaf and hearing-impaired children often experience delayed development of speech, language and cognitive skills, which may result in slow learning and difficulty progressing in school. Congenital and early childhood onset deafness or severe-to-profound hearing impairment may also affect the auditory neuropathway of children at a later developmental stage if appropriate and optimal interventions are not provided within the critical period of central auditory pathway development.

There are two main screening interventions generally available to a number of healthcare systems worldwide. These interventions are based on electrophysiological methods; transient evoked otoacoustic emissions (TEOAE) and automated auditory brainstem response (AABR). TEOAE measures sounds that are produced by the cochlea to response to acoustic stimulation and AABR measures electroencephalographic waveforms in response to clicks [6]. One common form of newborn hearing screening is a universal programme that occurs very soon after birth either at the hospital

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or in the community setting, in which all infants are screened [7]. More selective screening strategies have also been adopted in healthcare systems that only target the high risk population [3]. A family history of hearing impairment, admission to the neonatal intensive care unit or craniofacial abnormalities are general criterion which define an infant at high risk [3]. However, in reality this high risk criterion does not identify all at-risk infants [3].

It has been estimated that untreated deaf infants can cost society approximately $1,126,300 over the course of their lifetime [8]. Therefore, early detection is a vitally important element in providing appropriate support for deaf and hearing-impaired babies that will help them enjoy equal opportunities in society alongside all other children.

In essence, monetary and medical resources are scarce [9]. Choices have to be made about their deployment and an estimate of the additional resources that have to be used to obtain the additional benefit needed. This analysis aims to estimate the costs and effectiveness of various screening strategies to detect infants at risk of hearing impairment. Because situations are likely to differ substantially between settings and countries, case studies are included in the model, illustrating the likely impacts of cost and outcomes in a range of international settings.

2. Methods

The economic analysis used a decision tree approach to determine the cost-effectiveness of newborn hearing screening strategies. A decision tree allows the prediction of the number of patients who are likely to follow a particular pathway. Each pathway generates a unique outcome in terms of costs and health benefits. The outcomes can then be combined with the number of patients that experience each end state in order to calculate the expected cost for the patient cohort.

The model was developed using Microsoft Excel and aimed to capture the current pathway for a cohort of hypothetical newborn infants screened for deafness. The associated costs and outcomes for different screening strategies were included in the model. We have identified two countries to use as case studies within the model, namely the United Kingdom (UK) and India. This is due to the significant heterogeneity that exists between countries in terms of strategies, costs and the incidence of hearing loss in newborn infants. By focusing on two settings it increases the robustness and efficiency to contribute to policy decision-making. Although we are using the UK and Indian settings to illustrate the findings of the research, the model is built to demonstrate the likely impact of variations in basic model inputs, such as health care burden and intervention costs. This allows international decision makers to evaluate the relative impacts of each strategy in various international settings, such as those observed in developed and developing countries.

In the base case, only *healthcare* costs were considered. In an alternative analysis reported in Section 4, however, we took the perspective of society. That is, indirect costs (such as productivity and other patient-borne costs) were also included. This is because we are often assessing the strategies from a global perspective, rather than from the perspective of a single payer or healthcare provider.

This analysis includes the assessment of two unique models, each having been built to capture different strategic screening decisions. Firstly, the cost-effectiveness of *universal* newborn hearing screening (UNHS) was compared to *selective* screening of newborns with pre-specified risk factors. Secondly, the cost-effectiveness of providing a *one-stage* screening process vs. a *two-stage* screening process was investigated.

Previous studies have shown that, through the introduction of UNHS, the average age of identification of hearing loss in infants is reduced from approximately 12 to 18 months down to 6 months or less [8,10]. A limitation of UNHS is that, due to the low prevalence of hearing loss in infants, a significant number of false-positive are accumulated which results in a low positive predictive value (PPV). This is likely to result in unnecessary costs and monitoring of children who do not have hearing loss. An alternative strategy is the 'traditional' selective screening of newborns with risk factors. Risk factors for congenital bilateral deafness are infants with a family history of hearing impairment or infants with craniofacial anomalies [11]. Because the selective hearing strategy focuses on a smaller subset the strategy incurs fewer costs and has a higher PPV than UNHS strategies. Therefore, it is important to calculate the additional cost of providing a universal strategy in comparison to the selective hearing strategy, to estimate the potential economic impact from identifying a higher number of false-positives but fewer deaf infants.

Although examining newborn infants twice reduces the number of false-positive results, it also increases the number of false negative results, due to the fact that *both* readings must be positive in order to lead to a positive diagnosis. Consequently, this increases the specificity of testing and reduces its sensitivity. Therefore, it is equally important to assess the economic impact of providing a one-stage protocol in comparison to a two-stage process. For the purpose of this analysis, we assumed that these two strategies would be provided universally to newborn infants.

Two interventions for hearing screening programmes are considered in this analysis; transient evoked otoacoustic emission (TEOAE) and automated auditory brainstem response (AABR). For the purpose of this analysis, we have assumed newborn infants receive TEOAE followed by AABR if the first screen was positive. This is consistent and similar in structure with previous cost-effectiveness analyses [8,12]. In the case where only one-stage screening is analysed we have assumed patients will be assessed using TEOAE (see Figs. 1 and 2 for reference).

This cost-effectiveness analysis follows a hypothetical cohort of 100,000 newborn infants. Hearing loss in this analysis is defined as $\geq$40 dB which is the most consistent definition throughout the literature [1–3,8].

All relevant healthcare costs were incorporated. This included health care costs that were incurred as a direct consequence of the strategy employed. Further analysis that identifies the indirect costs such as travel time and lost productivity due to symptom-related work absence is presented in Section 4. All costs are evaluated in 2010 pounds (£) in the base case, although, since costs were drawn from international sources, the costs in this analysis can be converted into other currencies.

For each strategy (i.e. universal, selective, two-stage screening, one-stage screening), total costs (i.e. screening and all subsequent costs) were calculated by assigning costs to the corresponding numbers of patients who were at the end of each pathway in the model. The relevant costs comprised the costs of the interventions (i.e. screening interventions and medical supplies) and the associated resource use, such as the cost of different levels of staff involved; coordinator, screener, clerk or audiologist. The total costs include the costs associated with the different pathways followed by patients who attempt to achieve continence.

The model also included a cost for those patients who achieved a false positive screening result. This is the proportion of positive test results that are really negative events. False-positive results may cause parental anxiety and result in unnecessary follow-up tests and occasionally unnecessary interventions. In this analysis, it is assumed these infants incur an additional cost of an outpatient audiologist visit.

When required costs were adjusted to reflect the cost expressed in sterling and price year using the 'CCEMG – EPPI-Centre Cost Converter' (v.1.0) tool [13]. Table 1 lists the costs captured in the model.