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Ethical issues in screening for hearing impairment in newborns in developing countries

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Screening of newborns for permanent congenital or early-onset hearing impairment has emerged as an essential component of neonatal care in developed countries, following favourable outcomes from early intervention in the critical period for optimal speech and language development. Progress towards a similar programme in developing countries, where most of the world's children with hearing impairment reside, may be impeded by reservations about the available level of support services and the possible effect of the prevailing healthcare challenges. Ethical justification for the systematic introduction of screening programmes for hearing in newborns based on the limitations in current primary prevention strategies, lack of credible alternative early-detection strategies and the incentives for capacity-building for the requisite support services is examined.

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The right of every child to survive and to thrive is well acknowledged,[1] notwithstanding the prevailing burden of adverse perinatal conditions on neonatal care in developing countries.[2] Many of these conditions cause substantial neonatal mortality and are also associated with lifelong disabilities such as congenital and early-onset (ie, occurring within the neonatal period) hearing impairment.[3]

Permanent congenital or early-onset hearing impairment (PCEHI) is one of the most common abnormalities in children, which is detectable shortly after birth.[4] PCEHI may markedly impair speech and language acquisition if detected late, and thereby compromise optimal childhood development and lifelong vocational prospects irretrievably.[5] Early detection of PCEHI makes early intervention possible, to achieve favourable outcomes in language skills and cognitive development.[5 6] After the development of objective, automated, simple, safe and reliable electrophysiological screening technologies consisting of otoacoustic emissions (OAE) and auditory brainstem response (ABR), screening for hearing in newborns before discharge from hospital or within the first 3 months of life has evolved as an essential component of childcare in most parts of the developed countries.[7–9] By contrast, routine screening for hearing in newborns or infants is rare in developing countries, where 90% of the estimated 665 000 babies born with PCEHI annually worldwide reside.[10]

In 1995, the World Health Assembly passed resolution WHA 48.9 urging member states "to prepare national plans for the prevention and control of major causes of avoidable hearing loss, and for early detection in babies, toddlers and children, as well as in the elderly, within the primary healthcare."[11] Some health professionals, however, believe that it is unethical to introduce a screening programme until adequate facilities and skilled personnel are available to deal with all the consequences of the screening programme,[12 13] and the rationale for introducing the screening for hearing in newborns in developing countries has been questioned because of current shortages in relevant services.[14] Ironically, it is doubtful if this interim constraint were in itself sufficient to foreclose any initiative towards early detection on moral grounds, given the evidence on the adverse consequences from the late detection of PCEHI on early childhood development and the need to offer every child the chance for the best start in life, as advocated by Unicef.[1 4] The feasibility of screening for hearing in newborns in developing countries has already been shown by a few pilot or ongoing programmes.[15–17] This review therefore examines the ethical considerations for screening for hearing in newborns when and where intervention services are limited but evolving.

ETHICAL STANDARDS FOR SCREENING FOR HEARING IN NEWBORNS

Every pregnant mother expects and longs to give birth to a healthy baby and to leave the hospital with the assurance that all is well, especially when there are no apparent complications at delivery. Routine neonatal examination by health professionals has therefore become an accepted practice for detecting potentially serious conditions in apparently healthy newborns before discharge from hospital. Mothers are reassured when no abnormalities are detected, and are alerted when any abnormality is detected from this subjective examination. Congenital hearing impairment, however, is an invisible disorder that cannot be detected through neonatal examination, even among babies that are considered to be at high risk. Screening for such abnormalities by using objective tests has therefore been acknowledged as vital care for the newborn.

Screening is "the systematic application of a test or enquiry, to identify individuals at

Abbreviations: ABR, auditory brainstem response; OAE, otoacoustic emission; PCEHI, permanent congenital or early-onset hearing impairment

sufficient risk to benefit from further investigation or direct preventive action, among people who have not sought medical attention on account of symptoms of that disorder.''[18] In cases when a condition is ''hidden'' and has no adverse and major consequences on a child's development, its detection may be difficult to justify on moral grounds,[19] especially where intervention services are limited. Congenital hearing impairment, however, does not fall into this category because its adverse effects are inevitable and become apparent at a time when intervention is at best suboptimal. The decision not to screen for a condition, which will eventually be apparent regardless of the availability of professional services, presents an ethical dilemma for healthcare providers.

The general principles governing medical ethics (see box), which also must underpin screening for hearing in newborns, consist of non-maleficence (obligation to avoid causing harm); beneficence (obligations to provide benefits and to balance benefits against risks); autonomy (the right to choose); and justice (obligations of fairness in the distribution of benefits and risks).[20]

Ethical principles for screening for hearing in newborns*

- Parental autonomy: obligations to respect the decision-making capacity of parents to choose or decline screening
- Non-maleficence: obligations to avoid causing harm to the parent or child
- Beneficence: obligations to act for the benefit of the parent and child; and to balance benefits against risks
- Justice: obligations of fairness in the distribution of benefits and risks, and to ensure equitable access to screening

*Adapted from Beauchamp.[20]

BENEFITS AND RISKS OF SCREENING FOR HEARING IN NEWBORNS

Balancing benefits and risks in medical care is a moral duty and a frequent challenge for clinicians and other health professionals. It is a more difficult task in developing countries, where ordinarily it makes intuitive sense to devote considerable attention and resources to life-threatening conditions. Therefore, the identification of children with PCEHI from a normal population by health professionals through unsolicited screening (rather than waiting for affected children to present, by which time the optimal intervention has usually been missed) makes it ethically imperative that the benefits from screening outweigh any associated risks.

The primary benefit and goal of screening for hearing in newborns is the accurate identification of the few babies from a normal population who are likely to have PCEHI after a diagnostic or confirmatory test. Reliability is usually measured by the sensitivity (the proportion of those with the target condition that are rightly detected) and specificity (the proportion of those without the target condition that are rightly excluded) of the test. The current screening tests for hearing in infants (OAE and ABR) have proved to be quite reliable on the basis of these criteria, with a sensitivity and specificity of more than 90%, especially when combined in succession for a two-stage screening and conducted not earlier than 48 h after birth because of the high false-positive rates associated with vernix plugs.[21]

Screening for hearing in infants also offers the parents of a child with hearing impairment the knowledge of the special needs of their apparently normal baby as early as possible. In the absence of screening, hearing impairment is unlikely to be detected until the parents or caregivers observe a child's inability to respond to sound and the occurrence of inappropriate behaviour or speech and language defects when compared with their peers, from 12 to 18 months of age. During this process, parents who suspect the condition in their infants are often anxious, confused and make false assumptions about the nature, degree and full effects of the condition until they receive appropriate professional attention.

Screening for hearing in newborns is not without some risks for the child and the parents, because screening tests are not perfect. For instance, it is possible for a child to fail the screening test in the absence of PCEHI. Such an outcome may cause the parents unnecessary anxiety and stress until a diagnostic investigation is conducted. It is also possible for a child with or at risk of developing the disorder to pass the screening test and thus falsely assuring the parents that all is well. Parents can also be unnecessarily anxious about the outcome of the screening tests in their apparently normal babies. In addition, parents with unresolved grief after the diagnosis of hearing impairment in their child may be preoccupied with negative emotions, which could be detrimental to parent–child bonding. Notwithstanding, available evidence from parental accounts and other reports strongly suggests that these potential harms are unlikely to outweigh the benefits of screening tests for hearing in newborns for the child and the parents.[22–24]

PARENTAL AUTONOMY AND SCREENING FOR HEARING IN NEWBORNS

Parental informed consent is necessary before screening for hearing is undertaken and this should be sought within an established ethical framework.[8 25] Such consent is readily given if screening is presented within the context of the routine neonatal examinations, which parents expect shortly after delivery or before discharge from hospital. Parents are also likely to accept current screening tests for hearing because they are painless, non-invasive and quick to administer. Simple and culturally appropriate educational materials on the consequences of PCEHI and the value of early detection of hearing must therefore be provided to parents. Communication with audiovisual aids in local languages during antenatal and child health clinics, as currently practised in community-based public health interventions, would be valuable. It is equally important for service providers to refrain from creating unrealistic expectations, as it may undermine ongoing parental support for the screening and follow-up programmes.

The arrival of the newborn is a joyous and emotional event for the family and the disclosure of a permanent abnormality in an apparently normal baby must be handled with sensitivity. Parental reaction to this information would normally be characterised by shock, denial, grief and depression.[26] In some communities, unfavourable and superstitious beliefs are held towards congenital abnormalities or childhood disabilities, which may confound parental reactions to medical diagnosis. For instance, in one study in rural Papua New Guinea, sorcery and other supernatural factors were deemed to be the most common causes of disabilities.[27] Well-trained medical personnel must therefore handle the communication of the screening results, the possibility of false-positive and the diagnostic results to parents. The effective management of this process is crucial for speedy parental adjustment, good parent–child bonding and the

child's subsequent enrolment into a family-oriented early intervention programme.[28 29]

DISTRIBUTIVE JUSTICE IN SCREENING FOR HEARING IN NEWBORNS

Poor public funding or the lack of adequate funding to implement screening programmes in developing countries has led to the suggestion that it is probably more efficient or cost-effective to embark on primary prevention of hearing impairment by the same efforts aimed at reducing infant mortality.[2 30] Unfortunately, healthcare services in many of these countries are unlikely to develop rapidly to levels that will markedly curtail the incidence of congenital and early-onset hearing loss in the near future. For instance, although rubella and meningitis are well-documented causes of PCEHI, the relevant vaccines are not currently included in national immunisation programmes in many developing countries.[1] The equity in screening for hearing in newborns is therefore largely derived from the lack of an effective primary prevention strategy to tackle the full spectrum of PCEHI attributable to hereditary or genetic factors presently.

With the significant involvement of the private sector in healthcare delivery, especially in urban centres, the requirement for distributive justice may be redefined in terms of equity of access[31] and the identification or recognition of best practices in medical care to guide health professionals regularly.[8] Efforts should be made to create awareness and facilitate access to screening for all those who stand to benefit. These approaches are likely to diminish the resources available to other healthcare needs inequitably and they may result in raising the overall standard of healthcare across the communities. Screening for hearing in newborns may in fact stimulate interest and capacity-building for other screening programmes for hearing in newborns not currently offered in developing countries.

If screening for hearing in newborns is currently mandated in most parts of the developed countries, where only about 10% of children with PCEHI worldwide are found, then distributive justice on a global level suggests that this vital public health programme should be offered also in the developing countries where most of the potential beneficiaries reside.

The traditional approach of first seeking to ascertain quantitatively the cost-effectiveness of introducing a screening programme may be inappropriate ethically, where the alternative option is not to screen. Such an analysis is perhaps more appropriate when considering several screening options such as universal versus selected screening or hospital-based versus community-based screening. Failure to screen, as is currently the case in many developing countries, cannot be regarded as an option for this condition. Leveraging on existing well-established child health platforms, such as Unicef's expanded programme on immunisation, is perhaps a practical and cost-effective option for implementing an early-detection programme for PCEHI.[15 23]

SHOULD LIMITED INTERVENTION SERVICES FORESTALL EARLY DETECTION OF HEARING?

An essential requirement for the introduction of any screening programme for hearing in newborns is evidence that an efficacious treatment exists, which is demonstrable by current communications options for children with PCEHI.[32] Early detection makes early and appropriate intervention possible. The term ''early intervention'' refers to a broad array of activities aimed at optimising early childhood development. Early intervention services are therefore designed to meet the developmental needs of children from birth to 3 years of age, who have a developmental delay in physical, cognitive, communication, social, emotional or adaptive development or have a diagnosed condition that has a high probability of resulting in developmental delay.[33]

The availability of facilities to deliver these services, however, varies and grows with time even in developed countries. For example, in the UK, a national screening programme for hearing in newborns was introduced in 2002, even though there was a dearth of adequate professional staff to support the programme. This shortage was subsequently addressed through accelerated on-going training programmes for audiologists, along with a package of incentives. Similar arrangements were made in the US to tackle the shortage of skilled professionals to support the rapidly expanding screening programmes for hearing in newborns across the country. The training and use of community extension workers and non-specialists is a proved short-term strategy in developing countries for scaling up health interventions, and may be equally valuable for the provision of screening, for hearing in infants.[34 35] In fact, OAE and ABR screening instruments are automated simple to use and require no audiological expertise. With minimal training, anyone with a basic education, besides nurses and midwives, can be trained in a few weeks to conduct the screening tests, whereas specially trained personnel such as audiologists may be required to conduct the diagnostic tests at designated referral centres.

Lately, the World Health Organization has issued guidelines for the development of audiological services to facilitate capacity-building at different levels of healthcare delivery to deal with the current resource gap.[36] Recognising the constraints owing to the high costs of hearing aids, the World Health Organization has also taken various steps to encourage the manufacture of affordable hearing aids for people in developing countries, besides other private-sector initiatives to produce solar-powered hearing aids at affordable running costs.[37 38] Consequently, failure to screen because of current temporary shortages in service delivery may be counterproductive for requisite capacity-building.

Before starting an early intervention programme, parents require time to adjust to the unpleasant news of a hidden abnormality with lifelong consequences in their child after the initial shock and denial. Any screening programme that entails diagnostic and therapeutic interventions that are associated with social stigmatisation, those that offend cultural norms or are inconvenient may prolong this period. Parents need to be reassured that their babies have normal abilities and are able to compensate for lack of sound during this period.[39] More importantly, many have intuitive skills to foster mutually satisfying and reciprocal early interactions with their babies. This stimulation and attachment are vital components of any effective family-oriented intervention programme.[33] Parents of children with PCEHI belong to all social classes[3] and as they long to establish meaningful relationships with their children, they (particularly the more affluent) are likely to drive the development of appropriate services that will enhance the prospects for mainstreaming.

For conditions such as phenylketonuria and congenital hypothyroidism, early detection through newborn screening is rarely worthwhile where intervention services are not immediately available, because untreated children may have progressive and severe neurological damage. For PCEHI, however, the early identification of the special needs of an apparently normal baby serves the best interest of the parents. For instance, such early awareness minimises the period of uncertainty regarding the condition of the child and the potential wild-goose chase in finding explanations for the early signs of PCEHI. Parents are often displeased and sometimes angry when such conditions are detected much later after prior consultations with health professionals.[28 29] In fact, not knowing what is wrong with a child early may

lead parents to take the wrong actions, especially in an environment given to superstitious beliefs and traditional medicine.[27] Consequently, we believe that early detection of hearing should be encouraged in developing countries, even where intervention services are limited but evolving.

Services for the education of the hearing impaired through sign language are relatively well-established in developing countries, because sign language was the only option available to parents for many years and this required enrolment of the child in a school for the hearing impaired. The literacy and educational achievements of hearing-impaired children restricted to sign language are, however, unable to offer them access to full and gainful employment in later life.[40] Sign language may therefore end up being a fall-back option when parents are unable to secure auditory–verbal intervention for their children.

CONCLUSIONS

From available evidence, screening for hearing in newborns satisfies the ethical standards for a public health intervention. No ethical priorities are served by further delay in implementing this programme in any developing country on account of limited and evolving intervention services. A systematic introduction of the programme, beginning with pilot schemes, should provide valuable lead-time in capacity building for the requisite support services.

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