THE ECONOMIC IMPACT OF EARLY DIAGNOSIS OF CONGENITAL HEARING LOSS

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Abstract: The prevalence of newborn hearing disorders is 1-3/1000 and early diagnosis and treatment (preferably within the first 3 months) are crucial for normal intellectual, social, linguistic, cognitive, emotional and cultural development. For this purpose, universal screening is recommended but these tests are, unfortunately, not realized in most European health care systems. The current paper aims to present this issue not from a medical point of view but rather from an economic perspective. The fact that congenital deafness is the most frequent congenital condition that can be diagnosed immediately after birth offers the possibility of early treatment and considerable economic advantages by reducing the costs of special education, social integration and unemployment.

Keywords: congenital, deafness, economic, costs, integration

INTRODUCTION

Congenital deafness is the most common type of sensorineural hearing loss in developed countries (1/750 children develop a potentially debilitating sensorineural hearing loss).[15] In the general population, this affliction becomes more frequent with age, which can suggest the impact of genetic or environmental factors and their interaction. It can generate major difficulties of language development and social integration.

The disability is not always immediately apparent after birth. Since exposure to a normal acoustic environment is required for maturation, it follows that significant reduction of sensory input, induces both anatomical and physiological alteration of the auditory pathways.[1] Hattori et al., studied the speech recognition thresholds in children with moderately severe to profound hearing loss and found that speech recognition thresholds in children showed an increase of performance over time in both ears of binaurally aided and monaurally aided children. Nevertheless, the aided ear of monaurally fitted children showed a greater improvement in performance relative to that measured for the unaided ear. It was concluded that, in the aided ear, the benefit from amplification is added to natural maturation effect.[9,1] This means that early detection equals early treatment and therefore better chances for normal intellectual development. If congenital hearing disorders are detected and treated in time, most of the children are enabled to pass through a normal development of speech and no special education is necessary.[10] This would reduce drastically the expenses of any health care system. On the other hand, a low rate of early diagnosis and therapy has important socio-economic implications since more funds must be assigned for taking care of a deaf-mute child.

CAUSES OF CONGENITAL DEAFNESS

Various studies have shown that approximately 50% of all prelingual deafness in developed countries is caused by genetic factors whilst the rest of 50% is due to environmental factors such as pre- or postnatal infection, birth complications or ototoxic medication. (Table 1)

Although these figures must be regarded with caution, the number of genetic cases is expected to rise, especially in countries with a high level of healthcare although, environmental and birth-related risk factors must not be overlooked.

Table 1– Etiology of congenital deafness[16]

50% environmental		Cytomegalovirus infection(CMV) Meningitis Rubella Prematurity Neonatal jaundice Ototoxicity (genetic predisposition) Other infections
50% genetic	30% syndromic	Alport Norrie Usher Pendred Waardenburg Branchio-oto-renal syndrome Jervel & Lange-Nielsen
	70% non-syndromic	Autosomal dominant (DFNA1 – DFNA3) – 15% Autosomal recessive (DFNB1 – DFNB30) – 80% X-linked (DFN1 – DFN8) – 2-3% Mitocondrial – 2%

Our study from 2015 on 854 new-borns who underwent early screening (first week after birth) shows a statisticaly signifficant correlation between the risk of congenital hypoacusis and prematurity, gestational age, birth weight (Very Low Birth Weight -VLBW, Extremely Low Birth Weight - ELBW), Apgar score, congenital infection, ototoxic treatment, intraventricular hemorrhage, respiratory disstres, neonatal hypoxia, mechanical ventilation, NICU admission, neonatal hypotension, hypoxic-ischemic perinatal encephalopathy, prolongued jaundice, cranio-facial anomalies and other congenital anomalies.[14]

SCREENING METHODS

Otoacoustic emissions (OAE) are the most common method for early screening (Transient Evoked Otoacoustic Emissions -TE-OAE and Distortion Product Otoacoustic Emissions – DP-OAE). These are basically sounds generated by vibrations of the cells in the inner ear as a response to acoustic stimulation (clicks or tones) with a microphone in the external ear canal (EAC). They travel backwards, from the internal ear towards the external ear canal where they can be measured with a special probe. The lack of OAEs in a new-born is highly

suggestive of congenital sensorineural hearing loss and reflects the status of the peripheral auditory system. This test is extremely quick (1-3 minutes), practical and easy to perform (can be performed by nurses), needs no anesthesia, no special conditions and has no side effects. The equipment is also reasonably priced.

Auditory Brainstem Response (ABR) measures, in addition to the integrity of the inner ear, the auditory pathway. The electrophysiological response of the brainstem to auditory stimulation (microphone in the EAC) is measured by electrodes placed on the scalp. This test takes longer time (15-20 minutes) and sometimes requires sedation of the child.

The most common strategy is the so-called two step TE-OAE strategy with a single TE-OAE test in the first few days and another one a few days later if the at the first examination no OAEs are detected. However, different combinations of OAE and ABR testing are available.

ECONOMIC CONSIDERATIONS

A direct result of school-age hypoacusis is difficulty in properly receiving language in an educational environment followed by lack of attention, difficult learning and social integration. The costs of rehabilitation, special education and especially of not integrating deaf-mutes into the labour market have been estimated at $154-186 billion (approximately 3% of the internal gross income of the U.S.A.)[17]. In 2003, the averaged costs over the entire life span of a deaf patient were $383.000 whilst 30% of pupils repeated at least one grade.

A person suffering from hypoacusis is more likely to have poorer results than his colleagues in reading and writing, even though he can obtain a baccalaureat diploma. He will be less competitive on the labour market and will have smaller chances to complete higher education.

A 2007 German study concluded that most European countries, including Germany, have a mean diagnosis age for congenital deafness of 2 – 4 years and that treatment usualy starts 9 months later.[8,10] This data creates a grave discrepancy with international recommendations of diagnosis in the first 3 months and treatment in the first 6 months. This type of early detection can only be achieved through universal screening. In Germany universal screening is only available in some clinics since the costs are being reimbursed by insurance companies only if hypoacusis is diagnosed. Other countries use a risk screening which only tests newborns with documented risk factors (perinatal infections, birth complications, ototoxic treatment etc.). Romania practices none of these types of screening on an organized scale. Some maternity hospitals from the big cities have the necessary equipment and use it for universal screening, other clinics only employ risk screening but overall, most of the children have no type of hearing test once or ever.

Another conclusion of the German study by Hessel et al. is that a two stage universal screening using TE-OAE will diagnose 72% of all hypoacusis cases while risk screening only 43% and random screening 20%. In 2000, the estimated costs of universal screening for 100.000 neonates was €2 million whilst for risk screening €1 million and for random screening (no usual screening strategy) €0.6 million. This would mean a cost per capita of €20, €10 and €6 respectively.[10]

A 2001 Dutch study compares the costs of TE-OAE and ABR screening and concludes that the first method is cheaper with a cost of €25 per capita versus €39 for the second method.[2] A three stage screening tends to be more cost efficient than the two stage screening.

In 2007, Ciorba et al., at the University of Ferrara, Italy have had a cost of €9.20 for each tested child. For a rate of 1000 children tested every year, the initial investment for purchasing the OAE device was recovered in 2 years.[4]

It is recommandable that all children be tested in the maternity hospital, during the first week of life, thus avoiding the situation in which parents to not return for a scheduled appointment.[6,11] and also because all the necesary information regarding risk factors, pregnancy, birth and complications are readily there. On the other hand, there are some disadvantages to early OAE testing such as false negative results induced by amniotic fluid in the middle ear or the presence of vernix in the external ear canal of the newborn.[18,11] Some studies reported up to 20% testing errors in such cases.[3,5,12,13,18] For this reason a two step screening is absolutely necessary.

Our study from 2015 reports an alarmingly high percentage (33%) of subjects who did not return for a second test after initially failing the first TE-OAE testing[14]. These values are comparable to those reported in Iran, by Haghshenas et al., of 29.7% of patients „lost" during their three step testing.[7] This state of facts can be attributed to poor medical education in developing contries all over the world, lack of acces to health care or lack of equipment. We can only hope that some of these cases did not return because they seeked medical advice in other specialised clinics from Romania or abroad.

According to Hessel et al., some of the cost calculations for screening must include[10]:

Direct medical costs:

- Costs of implementation of screening programme
- Costs of screening tests
- Costs for tracking
- Costs for further diagnosis
- Treatment for detected cases (regular controls, hearing aids, batteries, check-ups, cochlear implantation and rehabilitation)

Direct non-medical costs:

- Transportation costs for diagnostic and treatment
- Caregiving time for brothers and sisters
- Additional education costs for special institutions

Indirect costs:

- Work time loss for parents
- Work time loss for grown-up persons with hearing disorders
- Income loss due to hearing disorders
- Productivity loss due to premature mortality

CONCLUSIONS

TE-OAE is easier to perform (no sedation required) and interpret, less time consuming (25-330 seconds) and cheaper but shows more false positive results. ABR is more time consuming (15-20 minutes), sometimes requires sedation but is considered a golden standard for diagnosis of hearing disorders.

At a cost of around €10 for each test, the initial investment of approximately €3000 for a TE-OAE device will be recovered in 2 years but the long term economic advantages are considerable and can amount to hundreds of thousands of Euros over the entire lifespan of a deaf

person. Under the socio-economic conditions of Romania we find the two step TE-OAE testing to be sufficient for an initial early screening that could raise a suspicion of congenital hearing loss. Failing this initial test can be followed by DP-OAE testing or ABR. The one step testing tends to give more false positive results.

The implementation of a national screening system would benefit the economy by reducing the rehabilitation and social integration costs. These children who are diagnosed within the first 3 months will be able to have a normal intellectual development and would function as normal individuals on the labour market.

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