Original Article

Is Newborn Hearing Screening Cost Effective? Economic Consideration for Policy Makers

Abstract

Backgroud: Hearing loss is the most common congenital disorder that appears as a unilateral or bilateral deafness. Early detection by screening and appropriate intervention lead to better oral communication and language development. The aim of this study was to evaluate the cost per new case identification of neonate hearing screening. **Methods:** The cost effectiveness of two stage hospital based newborn hearing loss screening was evaluated in this study. We gathered data for 11168 newborns born in 10 hospitals. We included a direct cost and new case identification as cost and outcome measures from health system prospective in our model. **Results:** We found 19 new cases with hearing loss from 11168 screened neonates (1.7 per 1000). The referral rates in the first and second stages were respectively 28% and 7%. The total cost of screening program was 132167 US\$. The main cost item is screening test (OAE). **Conclusions:** We concluded cost per new case detection is 6956 US\$ in Iranian neonate hearing screening program. Almost, it is equal to GDP per capita and it may be cost effective. Since there are many strategies to screening of newborn, it is suggested that all alternative screening strategies be analyzed by a cost effectiveness method to find the best strategy for hearing loss screening.

Keywords: Cost benefit analysis, hearing loss, infant, mass screening

Introduction

Hearing loss is the hidden disability to perceive sound and it's considered as a severe disability.^[1–3] It is estimated that significant hearing impairment incidence would be 1 to 3 per 1000 live births.^[4–10] The study of hearing loss in Iran showed bilateral hearing loss prevalence was 2.3 per 1000 in 2004.^[11]

Congenital hearing loss led to some disorders and dysfunctions such as behavioral problems, delay of speech and language development, decreased psychological well-being, and poor adaptive skills.[12] Early detection with appropriate intervention including the use of hearing aid and auditory rehabilitation lead to better oral communication and language development.^[13] Some evidence showed that appropriate procedures in the first 3-6 months, develop speech and language skills, decrease the psychological disorders,^[14,15] enhance the educational accomplishment, increase longevity, and decrease the learning costs.^[16]

Therefore, the early detection of hearing loss and use of appropriate interventions are essential steps in oral and social communication.[11] Countries use different methods and instruments such as growth questionnaires, risk factors registration, case history, physical examination, Auditory Brainstem Response (ABR), and Transient Evoked OtoAcoustic Emission (TEOAE) to screen the hearing loss in neonates.[3,16-22] They are different in sensitivity and specificity and expenditure along them. Therefore, some of them (such as TEOAE) are preferred in newborn screening, because it is a high-sensitive test and can perform faster than others; moreover, it has little false positive cases.^[16]

Neonatal hearing screening was programmed in Iran in 1999 and its pilot implementation is begun two years after. This program was piloted for three years and the countrywide implementation started in 2004.^[19,23] Iranian newborn hearing screening is hospital based that is conducted in two consecutive stages of OAE test and ABR diagnostic test. This is the new form of public private partnership in hearing screening and its successful

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Fatemeh Jafarlou^{1,2}, Behzad Najafi³, Seved Jalal Sameni²

¹Departments of Audiology, Faculty of Rehabilitation Sciences, Tabriz University of Medical Sciences, Tabriz, Iran, ²Department of Audiology, Faulty of Rehabilitation Sciences, Iran University of Medical Sciences, Tehran, Iran, ³Health Economics, Faculty of Management and Medical Informatics, Tabriz University of Medical Sciences, Tabriz, Iran

Address for correspondence: Dr. Behzad Najafi, Department of Health Economics, Tabriz University of Medical Sciences, Daneshgah St, Tabriz, Iran. E-mail: Bnadjafi59@gmail.com



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implementation depends on some important factors such as efficiency and cost effectiveness ratio. There are few studies on the cost effectiveness of hearing screening methods^[4,5] specially in the developing countries context. However, because of the differences in the country context, protocol, and method, it has been reported wide range of Incremental Cost Effectiveness Ratio (ICER) in the studies. For example, Kemper et al., reported US\$ 24000 additional cost for new case detection for Newborn Hearing Screening Programmed strategy compared with targeted newborn screening, whereas keren et al., estimate it US\$ 44000.^[4] So there are different ranges of ICER and cost per new case detection in the studies and this has made newborn hearing screening controversial. Therefore, the aim of this study was to analyze the cost and effectiveness of neonatal hearing screening in Iran from the health system perspective, in order to provide some evidence for decision makers.

Methods

We apprised the two-stage screening protocol and process in Iran [Figure 1]. In this protocol all newborns are assessed with TEOAE and those who fail or pass with risk, should be referred to the hospitals to be reassessed with OAE in the period of two weeks after first evaluation. Infants, who fail in second assessment, are referred to another site to follow up with diagnostic test by ABR. They are assessed with ABR and the result of this test is used as final finding and diagnosis. Infants who have hearing loss are referred to rehabilitation and auditory training centers and the normal infants will be reassessed after 6 months.

Screeners used the OAE Accuscreen device. The devices set on click stimulus in intensity level of 75 dBSPL. The hearing loss threshold in ABR is 35dB nHL.

Newborn screening is done in public, private, and charity hospitals and we collected the data of 11168 infants, from10 hospitals. Screening data was filled in specific form that have been designed by State Welfare Organization of Iran (SWO). This form includes age, sex, risk factors, result of initial and repeat OAE, recommendations (i.e., pass or outpatient rescreen) and date. Screeners were responsible for completing the data forms based on their contract at each site. The forms were filled in the hospitals and aggregated in another form for reporting to SWO.

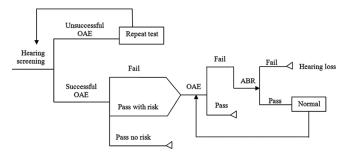


Figure 1: Decision tree for two-steps hearing screening

Resource utilization data was collected retrospectively from 10 hospitals and ABR sites. Some expenditure (such as screening equipment) was financed by SWO and some variable expenditure was funded by users. Screeners were from private sector and provided the screening services to neonates based on SWO tariff for five years; instead, SWO provided a free of charge TEOAE for each screener based on their contract. We estimated screening tests expenditure based on the real cost that was paid by patients.

We estimated two main categories of cost: (a) Fixed expenditures including screening equipment (TEOAE and ABR sets), and (b) Variable expenditures including screening tests expenditure by OAE, diagnostic expenditure, transporting and overhead cost. We used the patient user fees and other payers' data base such as Wellbeing Organization and hospitals data to estimate costs. We used the real cost which paid by different organizations. In the screening protocol there is no payment for screener other personals. Because of screener's salary, maintenance and other disposable costs are included in the OAE and ABR tariff, so we did not separate it. On the other hand, in the hospital-based screening, transportation expenditure in the first stage was zero and only it was paid for rescreening and ABR testing. Therefore, we ignored it in costing and long-term expenditures were not included. All costs were calculated from health system prospective and measured in IR Rial and change to US dollar based on International Monetary Fund (IMF) exchange rate¹.

Equipment costs were calculated based on their free market prices and depreciated in 5 years and overhead costs were computed based on spaces occupied by screening room.

Outcome measurement

The true identification of hearing loss cases was selected as the final measure of the outcome of newborn hearing screening program. Therefore, we reported the cost per new cases as final results in this study. We assume that those who failed in the two stage OAE test and ABR diagnostic test are true new cases and they have hearing loss.

Sensitivity analysis

In order to assess the robustness of the modeling results as recommended in guidelines for conducting cost effectiveness studies,^[20] we used one-way sensitivity analysis in this cost effectiveness study. Some parameters can vary in both costs and outcome sides based on modeling assumptions or program necessities. We used $\pm 20\%$ variation in the selected variables and estimated cost per identification again.

¹International Monetary Fund: 1 USD equal to 10361 IR Rial and 1\$ Int equal to 4558 IR Rial in the year of screening.

Results

This paper was focused on the costs of newborn hearing loss identification of universal newborn hearing screening in Iran. The results are presented in three sections. First, the process of screening and hearing impairment identification is presented. Second, we focus on the costs. Finally, we present the cost per new case identification.

Screening results

Table 1 displays the total birth, total referral from out of the screening hospitals and other facilities, and total initial OAE in the hospitals. Initial OAE was done for all referral neonates; therefore, in the first stage, 11168 OAE tests were done in 10 hospitals. 10348 newborns referred from screening hospitals and 820 neonates from other sites. 170 neonates (1.6%) born in hospitals were referred to the screening site.

Figure 2 shows the referral rates, pass, and fail values for each steps of neonates hearing screening program. As mentioned in Table 1, 170 neonates have not referred to the screening room. Therefore, we had 11168 neonates for first OAE test.

The pass rate of screening in initial OAE was 72% that resulted in the normal discharge of 8043 infants without any follow up. 3125 neonates who failed in the initial OAE test was requested to follow up by second OAE test at 2 weeks. Unfortunately, most of them did not refer and only 1005 (32%) neonates returned to reassessment at deadline. The pass rate in the second stage of the screening was reasonable and 93% of neonates passed the OAE test. The 73 infants who failed in the second evaluation with OAE were referred to more evaluation with diagnostic ABR. From 73 cases assessed by ABR, normal response was detected in 54 cases and abnormal response was

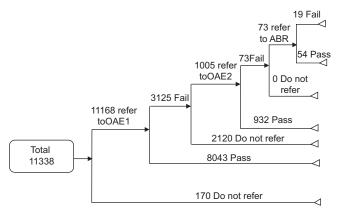


Figure 2: Flow chart and referral system for neonates hearing screening

obtained in 19 cases, so 19 neonates with hearing loss were diagnosed by ABR [Figure 2].

Finally, of the 11168 infants screened in the hearing screening program in the year, only 19 neonates (0.17%) did not pass the two-stage screen procedure with OAE and diagnostic evaluation with ABR. 2120 of high-risk neonates did not complete screening procedure.

Treatment and rehabilitation procedure were not included in this study and the screening procedure completed by conducting the diagnostic ABR. The newborn that has hearing disorder in the final step, referred to receive hearing services such as prescribing appropriate hearing aids and rehabilitation.

Costs

Since screening program was done in different hospitals by different OAE costs, screening cost per neonates differ. OAE prices varied from 54000 IR Rial (5.2 US\$) to240000 IR Rial (23.2 US\$). Table 2 displays the cost of the two stage OAE tests in national currency (IR Rial) and US\$, paid by patients in hospitals based on predefined prices. Total OAE cost in first stage of screening of 11168 neonates was 849932 thousand IR Rial (82032 US\$) that resulted in 3125 suspicious hearing loss newborn identification. Since the OAE prices varied by hospitals, the cost of the second stage was separately computed for every hospital based on referred cases. Total OAE cost in the second stage was 70086 thousand IR Rial (6764.4 US\$) for 1005 neonates screening. Therefore, we expensed 6764.4 US\$ to identify 73 newborns with suspicious hearing loss, who should have been referred to ABR. These costs are only for OAE test and other costs were computed following.

Totally 88796.3 US\$ was expensed in two stage OAE screening in order to identify 73 suspicious neonates. In other word, cost per suspicious case identification in these two stages was computed 1216.4 US\$. Suspicious cases were referred to test by diagnostic ABR. All 73 newborns referred from the previous stage, were evaluated by ABR in one screening site. Therefore, all of them had same costs. Based on SWO tariff, the ABR was 300000 IR Rial (28.95 US\$), so diagnostic test cost was computed 21900 thousand IR Rial (2113.7 \$) that resulted in 19 new cases with hearing loss identification.

Four TEOAE sets bought by SWO and were delivered to screeners. Its total cost was accumulated and depreciated in 5 years. So, we would have 134966470 IR Rial (13026 US\$) yearly. Overhead cost was computed by

	Table 1: Number of total birth and total referral to screening in hospitals									
	Total birth in screening hospitals	No. of who did not refer (Miss cases)	Percent	Referral from Other facilities	Total Birth	Total referred	Percent			
Total	10518	170	1.6	820	11338	11168	98.5			

their occupied spaces. This was computed 292500 thousand IR Rial (28231 US\$) for 2010.

Cost per case detected

The 19 New case identification cost summarized below:

•	First stage screening cost:	82032 US\$
٠	Second stage screening cost:	6764.4 US\$
٠	Diagnostic test cost:	2113.7 US\$
٠	Equipment cost:	13026 US\$
•	Overhead cost:	28231 US\$
•	Total cost:	132167

US\$

Cost per new case = $\frac{132167}{19}$ = 6956 US\$

In total, the new case identification in Iranian newborn hearing screening program had 6956 US\$.

Sensitivity analysis

The results of the one-way sensitivity analysis were displayed in Table 3. We changed the selected variables in \pm 20% range and re-estimated cost per new case as shown in Table 3. 20% variation in all costs and referral rates do not have important effects on the result. But the incidence rate affected the cost per identification if it varied in \pm 20% range.

Discussion

The early detection and rehabilitation of hearing impairment

give the best chances for neonates to develop normally.^[21] Hereupon, universal newborn hearing screening has been approved by both the American Task Force on Newborn and Infant Hearing and the Joint Committee on Infant Hearing (JCIH).^[22,24]

There isn't unique program for hearing screening and most hearing screening programs use the OAE and Automatic Auditory Brainstem Response (AABR) with different combinations.^[25]

The Iranian newborn hearing screening uses OAE for the initial and rescreening that is followed by ABR as a diagnostic test. Screening of 11168 neonates in ten hospitals resulted in 19 hearing loss newborns. Although 68% of who failed in first stage did not return to rescreening, but referral rate from initial OAE to rescreening is very high and also this rate to diagnostic test is high (7%). Vohr and et al., reported pass rate in the 120 ongoing programs 92% for OAE and 96% for ABR-based programs.^[26] In other words, OAE-based programs had twice the refer rate at discharge compared with ABR-based programs. High referral rates and high false positive response led to high cost per identification. In this study, referral rate in initial OAE screen is 28%. Although the studies reported screening methods make significantly different referral rates in hospital based screening; for example, AABR had 3.21% referral rate compared with 4.67% and 6.49% for

Table 2: Expenditure by cost drivers																
Hospital OAE test		1th OAE Test		2th OAE Test		Total OAE		ABR Test		Devices		Overhead				
Code	Fee		No	o Expenditure		No	Expenditure		Expenditure		Expenditure					
-	IR	US\$		US\$	IR		US\$	IR	US\$	IR	US\$	IR Rial	US\$	IR Rial	US\$	IR Rial
	Rial				Rial			Rial		Rial		(1000)		(1000)		(1000)
	(1000)				(1000)			(1000)		(1000)						
330	100	9.7	223	2152	22300	11	106.2	1100	2258.5	23400						
240	60	5.8	1165	6746	69900	85	492.2	5100	7238.7	75000						
220	120	11.6	317	3671	38040	5	57.9	600	3729.4	38640						
210	65	6.3	1578	9900	102570	109	683.8	7085	10583.4	109655						
190	137	13.2	433	5725	59321	19	251.2	2603	5976.6	61924						
170	240	23.2	68	1575	16320	0	0	0	1575.1	16320						
160	60	5.8	4089	23679	245340	501	2901.3	30060	26580.4	275400						
150	144	13.9	452	6282	65088	20	278	2880	6560	67968						
140	95	9.2	1891	17339	179645	168	1540.4	15960	18879	195605						
130	54	5.2	952	4962	51408	87	453.4	4698	5415.1	56106						
Total	_	_	11168	82032	849932	1005	6764.4	70086	88796.3	920018	2113.7	21900000	13026	134966470	28231	292500000

Table 3: Sensitivity analysis of main variables in the model									
OAE Unit Cost	Baseline	-20%	+20%	Tota	l cost	Cost per identification			
				-20%	+20%	-20%	+20%		
OAE cost	88796.3	71037	106555.7	114408	149926	6021.5	7890.8		
ABR cost	2113.7	1691	2536.4	131744.4	132589.9	6933.9	6978.4		
Overhead cost	28231	22585	33877	126520.9	137813.3	6659	7253.3		
Equipment cost	13026	10421	15631	129561.9	134772.3	6819	7093.3		
Referral rate to OAE2	28% (1005)	804	1206	130814.2	133520	6885	7027		
Referral rate to ABR	7% (73)	58	88	131732.8	132601.4	6933.3	6979		
Hearing loss incidence rate	0.17%	0.136%	0.204%	-	-	8811	5746		

two-steps TEOAE, respectively.^[12,24,27-31] The studies which apprised the sensitivity and specificity of OAE found it high sensitive and specific.^[26,32,33] On the other hand, in spite of high risk screening, referral rate in second stage showed that the specificity and sensitivity of OAE is not low (7% referral rate). It seems that the financial motivation of health providers like screeners, led to supply induced demand and request to rescreening. The payment mechanism in the Iranian hearing screening is fee for service. Therefore, screeners have a tendency to provide more services and impose more financial burden to health system.

However, the American Academy of Pediatrics Task Force on Newborn and Infant Hearing in the revised recommendations of the newborn hearing screening, provide two following concepts: (1) all infants with significant bilateral hearing loss should be identified in the screening program, and (2) the referral rate after screening should not exceed 4%. It is important to bear in mind, that although lower referral rate decreases screening cost, more emphasis on lower referral rate could cause in greater missing in identification of infants who have significant hearing loss.^[34]

There are few cost effectiveness studies on universal newborn hearing screening in the world. Most of them analyzed the cost per new case identification. We found the 11.8 US\$ per screened infants in this study. Other studies reported the cost per screened newborn 13 to 33 US\$ and the main component of cost was salary.^[27,30,35,36] Nguyen estimated 21 US\$ per infant screened by TEOAE.^[10]

Universal screening, high risk or targeted screening has different cost per identification. We found 6956 US\$ per new case identification in our study. The new cases that are identified have large effect on cost per identification. In our study high fall out rate could explain the low new cases identification. 68% of those who failed in the initial test did not return to rescreening. Low follow up rate is seen in similar program in other countries; for example, Ria and *et al.* reported 44.4 percent fall out in their study. This high falls out maybe due to lack of parents' confidence on screening test because of child's responses to sounds in the next months. Another cause possibly due to distance and the children's parent may be referred from distant location and they could not return to first location.^[37]

The literature showed that high risk screening can miss 50% cases.^[37] On the other hand, universal screening has large cost. Kemper and *et al.* compared the cost and benefit of universal and risk group screening via health system prospective. They reported that new cases are 40% more in universal screening compared with targeted screening. While incremental cost effectiveness of universal hearing screening compared targeted screening is 24000 US\$.^[13,8] Hessel and *et al.* analyzed the cost effectiveness of three screening strategies. They reported universal screening have 13395€ cost per child detected in comparison with 6715€ in risk screening and 4125€ for no screening.^[8]

There are no thresholds to find the cost effectiveness interventions in our country. However, based on the World Health Organization criteria, the acceptable criteria of an intervention, is based on GDP per capita. If incremental cost effectiveness ratio of an intervention is less than three times of GDP per capita, that intervention is cost effectiveness. Indeed, it is only acceptable when we use the DALY as outcome and report the excessive cost of each DALY gained. We found the cost per new detection is 6956 US\$ and, neonate hearing screening compared with no screening maybe cost effective, but it should be noted that this does not means it is the best strategy for hearing screening.

There is some limitation in our study that should be considered when its results are interpreted. There were some other costs that we did not include in the cost estimation. First, we analyzed the ongoing screening programs. Therefore, we did not include the establishment and running the program cost. Second, we use tests prices as a proxy of personal cost. However, personnel expenditures are included in tariff. Third, the equipment was annualized (amortizing over 5 years) and, therefore, doing not represent actual startup cash requirements. Finally, we included the short-term expenditures, but the long-term cost was not included.

Conclusions

In this study we screened 11168 neonates and found 19 (1.7/1000) new cases as hearing loss. There is very wide variation in referral rate from first OAE testing to the second one and second OAE to ABR. The results showed that the referral rate is high when there is self-refer.

The total diagnostic cost of screening in this study was estimated 132167 that resulted in 19 hearing loss cases. Therefore, cost per new case identification was estimated 6956 US\$. We also found the 11.8 US\$ per screened infants in this study.

Finally, we found that the newborn hearing screening in Iran is feasible and may be cost effective, but to find the best strategy for heating screening, we suggest all alternative screening strategies would be analyzed by cost effectiveness methods.

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Conflicts of interest

There are no conflicts of interest.

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