

ORIGINAL STUDY

Universal neonatal hearing screening: Six years of experience in Qatar

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INTRODUCTION

Hearing loss is one of the most common congenital anomalies observed at birth. It is reported to occur in approximately 2 infants per 1000, and in approximately 3 per 1000 infants in intensive care.¹ Early identification and intervention can prevent severe psychosocial, educational and linguistic repercussions as hearing loss can result in a language delay if not addressed early—infants who are not identified before 6 months of age have delays in speech and language development. Intervention at or before 6 months of age allows a child with impaired hearing to develop normal speech and language.

Prior to implementation of universal newborn screening, testing was conducted only on infants who met the criteria of the high-risk register (HRR). Recently it was found that the HRR was not enough, given that as many as 50% of infants born with hearing loss have no known risk factors.²

A retrospective study conducted by Connolly et al. in 2005 found that 1 of every 811 infants without risk factors and 1 of every 75 infants with risk factors have hearing loss.³ The prevalence of hearing loss may continue to change as more data become available from universal newborn hearing screening programs.³

Data from newborn hearing screening programs in Rhode Island, Colorado, and Texas, USA, showed that approximately 3 of every 1000 neonates suffer hearing loss.⁴

The American Joint Committee on Infant Hearing (JCIH 2007),⁵ The American Academy of Pediatrics (1999), and The National Institute of Health (NIH 1993) suggested in their positional statement on developing and maintaining UNHS programs that by screening they should be able to identify hearing loss by one month of age, confirmation of hearing loss by three months of age and enrollment in family centered intervention program by six months of age.⁶

In 1994, Bess and Paradise⁷ challenged the need for earlier identification and intervention, stating that there is no evidence to support the notion that "outcome in children with congenital results of hearing loss are more favorable if treatment is begun early in infancy rather than later in childhood".⁸

In 1995, Apuzzo and Yoshinaga-Itano⁹ found that infants identified when they were younger than two months had significantly higher language scores than those identified when they were older, despite similar interventions in both groups. In 1995, Robinshaw reported that children who were identified and who wore hearing aids by the age of six months acquired age-appropriate vocal communicative and linguistic skills well before children who were identified at a later age.¹⁰

The JCIH services expanded the target hearing loss as permanent bilateral, unilateral sensory or permanent conductive hearing loss to include neural hearing loss (e.g., "auditory neuropathy/desynchrony"). It also established separate screening and rescreening protocols for well baby and neonatal intensive care units (NICU), specifying that babies in the NICU for 5 days or more should be screened for Auditory Brainstem Response (ABR).⁵

Liu et al. in 2008¹¹ stated that 97% of 4 million infants born in the USA were screened for hearing loss, and we know that the success of newborn hearing programs depends on far more than the initial step of screening, as nearly one half of infants screened for hearing loss at birth are not followed up on and are lost in documentation. Yet, only close to one third of babies diagnosed with hearing loss are documented as having entered intervention. This dead zone between diagnosis and follow up threatens the success of screening programs. To enhance the performance of EHDI in meeting with screening, diagnosis and intervention goals at one, three, and six months of life, respectively, the project will work with hospitals, primary care providers, audiologists, ENT interventionists and families to reduce the lack of follow up and help states reach their EHDI goals. So far, reports on NHSP in Qatar had been read in different neonatal hearing screening conferences held locally in Doha,¹² Italy¹³ and Australia.^{14,15} A report on NIHSP was presented; however, so far, no six-year report has been presented or published, internationally.

AIMS OF STUDY

The objective of this study was to investigate the results of the newborn hearing screening program run by Hamad Hospital, the main tertiary general hospital in Doha, Qatar, and the maternity department in Al Khor General Hospital, Qatar, over a six year period. The primary focal points of the study follow: (1) the prevalence of hearing

impairment; (2) the impact of universal hearing screening programs according to the age at which the diagnosis of hearing loss is defined; (3) the cost effectiveness of the programs; (4) the outcomes in terms of age of hearing rehabilitation; (5) to assess the impact of UNHS on early identification and prevention of any possible hearing disorder among infants.

MATERIAL AND METHODS

Qatar conducts universal, country-wide neonatal hearing screening. The program began in the fall of 2003 at Hamad Medical Corporation with the deployment of the first stage of neonatal screening based on the Otoacoustic Emission tests (OAEs) GSI 70 Distortion Product Otoacoustic Emission (DPOAE), followed by the introduction for use of Biologic's Audex screener in the following year combined with application of the automated ABR Biologic Automated Auditory Brainstem Evoked Response (ABEAR) instrument.

Newborn hearing screening protocols/well baby protocol

The screening center in Qatar is located in HMC Women's Hospital, which is the main and largest maternity hospital in Doha, and chosen because it is the hospital where most mothers deliver. In addition, it is located within Hamad General Hospital, where the Audiology and ENT departments are located.

All well baby (WB) hearing is examined before discharge from the hospital [usually 24 hours after delivery]. If the baby passes ("pass") through the first stage of examination using DPOAE, then it's referred to the second stage, to be performed two to three months later [at the date of first vaccination], again using DPOAE. If the baby passes, it will be referred to the third stage of screening, to be conducted when it enters school at age six.

If the newborn fails the first test, then it will be sent to have another test based on DPOAE over the following two weeks at the screening unit. Passing results will shift the newborn onto the aforementioned track of screening. If the result is still questionable ("refer"), the baby should undergo another screening using automated ABR. Passing results at this phased mean the baby will enter the aforementioned process. If following ABR, the result is refer, then the baby will undergo another automated ABR screening test. Passing places the baby into the pass protocol aforementioned; however, if the result of the ABR is refer, then the baby will be sent to the audiologist for diagnostic audiometry to be conducted within two to three weeks. If the baby displays hearing loss, the arrangement for a hearing aid fitting will be done before or within the following six months, as arrangements for AVT and speech therapy are made.

Well babies referred from hospitals outside the HMC Women's Hospital or country, who usually arrive within few weeks following delivery, should follow the second stage stream [above].

NICU baby and risk baby protocols

National recommendations state that NICU babies (babies who have spent 48 hours or more in the Neonatal Intensive Care Unit or Special Care Baby Unit) should be screened using both OAEs and AABR, to exclude the possibility of auditory dyssynchrony.

Babies who stayed less than 48 hours stay in the NICU would start their second stage DPOAE screening, and if results were passing, they should follow the main stream. If the baby stayed for more 48 hours in one of the specialized units, then both DPOAE and AABR should be performed according to the first stage; another DPOAE and AABR should be performed within the second stage (within 3 months).

Risk factors for hearing loss include post-meningitis, hyperbilirubinemia requiring exchange transfusion, low birth weight, respiratory distress, prolonged mechanical ventilation, low Apgar scores, exposure to ototoxic medications such as gentamicin and loop diuretics, and a family history of childhood hearing impairment.¹⁶

Hearing loss is a relatively common congenital disorder, where the number has increased to one in every 40 infants. Those who require care in the neonatal intensive care unit (NICU) for more than 48 hours should follow stage two screening to be completed within two months. A stage three should be done with DPOAE and AABR by one year's time, and a stage four to be done at school age using diagnostic ABR and supplemented with

an audiologist's opinion. All babies' results are collected, analyzed and saved to a database and can be referenced anytime.

Equipment and method used in screening

According to JCIH recommendations, two screening devices are used in our screening program, namely the Distortion Product Otoacoustic Emission (DPOAE) and Automated Auditory Brainstem Evoked Response (AABR) machines.

Otoacoustic emissions are used to assess cochlear integrity and are physiologic measurements of the response of the outer hair cells to acoustic stimuli. They serve as a fast, objective screening tests for normal pre-neural cochlear function. To measure OAEs, a probe assembly is placed in the ear canal, tonal or click stimuli are delivered, and the OAE generated by the cochlea is measured with a highly sensitive microphone. Provided that the patient's middle ear function is normal, these measurements can be used to assess cochlear function for the 500 – 6000 Hz frequency range. The presence of evoked OAE responses indicates hearing sensitivity in the normal to near-normal range.

These emissions are fast, efficient, and frequency-specific measurements of peripheral auditory sensitivity. However, the effectiveness of the test is reduced by contamination with low-frequency ambient noise in a busy nursery, vernix in the ear canal, or any middle ear pathology. This is not a sufficient screening tool in infants who are at risk for neural hearing loss (e.g., auditory neuropathy/dyssynchrony). Any infant in the NICU or in the hospital for more than two days should undergo an ABR screening so that the presence of

Table 1. Number of deliveries and screened subjects.

Calendar year		2005	2006	2007	2008	2009	2010	Total
Total delivery	HMC	13076	13570	14108	14714	15354	16188	87010
	A.Khor*			479	568	598		1645
Live births	HMC	13362	13851	14434	15022	15660	16416	88745
	A.Khor			484	568	602	751	2405
Still births	HMC	66	65	81	83	64	81	440
	A.Khor			19	28	26		73
Total infants	100%	13362	13851	14918	15590	16262	17167	91150
No. screened		13085	13241	14248	15018	15952	16312	87856
% screened		97.93	95.59	95.5	96.33	98.09	92.01	
No. not scr.		277	610	670	472	310	855	3194
% not scr.		2.07	4.4	4.49	3.66	1.9	4.89	

WH: Infants registered at HMC's Women's Hospital.
 A Khor: Infants registered at Al Khor hospital.
 *No Recorded infant from this item – Al Khor hospital was not yet opened.

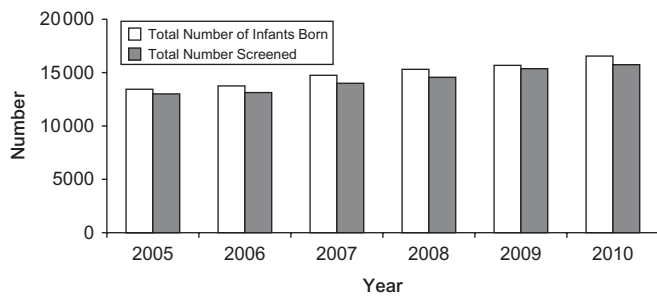


Figure 1. Total number of infants born and the screened over the year 2005 – 2010.

auditory neuropathy is not missed (recommendation of the JCHI 2007 and 2000), as children with this type of hearing loss have normal cochlear function and therefore normal OAE measurement results.

Automated auditory brainstem response (AABR) is an electrophysiological measurement that is used to assess auditory function from the eighth nerve through the auditory brainstem. These measurements are generally obtained by placing disposable surface electrodes high on the forehead, on the mastoid, and on the nape of the neck. The click stimulus (usually set at 35 dB hearing level [HL]) is delivered to the infant's ear via small disposable earphones designed to attenuate background noise. Most AABR systems compare an infant's waveform with that of a template developed from normative ABR infant data. A pass or fail response is determined from this comparison. Most commercially available systems can be used as an effective screening tool in infants younger than 6 months. The true sensitivity and specificity of newborn hearing screening is difficult to estimate from most screening programs. One large, good-quality study measured the sensitivity and specificity of OAE and ABR using an independent "gold standard," visual reinforcement audiometry, performed at 8 to 12 months.¹⁷

RESULTS

Table 1 shows the Newborn Hearing Screening Programme (NHSP) coverage over a six-year study period, revealing total number of deliveries, total live born infants delivered in HMC hospital and in Alkhor hospital, maternity unit. This covers almost most of the deliveries in Qatar, 91,150 newborns represent 100% of the infants that require screening over the six years of research. It shows the percentage of screened babies and the number and percentage of unscreened babies. It also reveals that an average of 96.4% of infants were screened and an average of 3.56% of infants were not (Figures 1 and 2).

Table 2 showed passing results [in first and second stages] of infant screening divided into WB number and

the number and percentages of those born and admitted more than two days in the NICU, as well as number and percentages of those WBs and NICU babies with refer results in the first two stages.

Table 3 showed results of screening tests over 6 years involving 86,865 newborns, and featuring 59754 [68%] pass results and 27103 [30.8%] refer results among infants screened. Both results were based on screening results of the first and second stages. Table 3 also features the final screening results, including the discovery of 1622 newborns with hearing impairment; specifically, 172 cases of bilateral permanent hearing loss and 1450 infants with unilateral sensorineural hearing loss and mixed loss, including syndromal conditions. It also features intervention results, including fitting of hearing aids with provision of AVT as well as speech therapy and cochlear implant surgery for 72 infants over 6 years.

The prevalence of bilateral permanent sensorineural hearing loss in our series was found to be 0.0019 [1.9 per 1000 screened infants], and 0.0184 [1.84%] infants displayed unilateral sensorineural, and mixed hearing loss (Figure 3).

Cost effectiveness of NHSP in Qatar

Table 4 shows the cost estimate of equipment used for the screening program over the period (from 2005 to 2010), including cost of instruments used in the study and disposables. It documents a total of 1,162,000 QR spent. Table 5 shows the payroll estimate of personnel involved in the screening program over the study period [some of the results were averaged for different salary grades]. The table documents a total of 21,051,200 QR spent [NB: all numbers are the author's estimate]. Table 6 revealed costs of instruments and spare parts used for intervention as well as cost of disposables used to maintain their work [some of these prices are not included in the study estimate].

Table 7 featured baseline results for all newborns screened: 172 cases of bilateral permanent sensorineural hearing loss and 1622 infants with both unilateral and mixed hearing with hearing threshold (of > 40 dB)

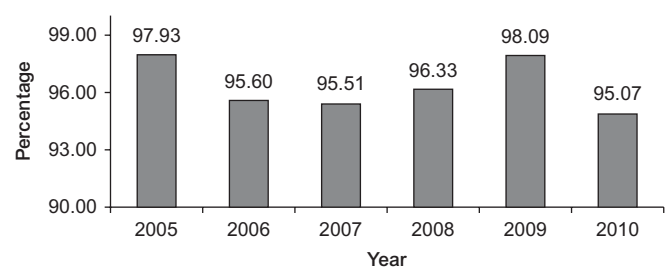


Figure 2. Percentage of screened subjects.

Table 2. Screened results.

Category	2005	2006	2007	2008	2009	2010	Total
Total infants screened	13085	13241	14248	15018	15952	16321	87865
No. of pass results stage 1 and 2	1122385.62%	954872.10%	907763.70%	920661.29%	961560.27%	1108567.95%	59754
No. of refer results stage 1 and 2	1863(14.23%)	2693(27.89%)	517136.29%	5812(38.7%)	633739.72%	5227(32.49%)	27103
Cases of HL diagnosed	186/13085 (1.42%)	253/13241 (1.91%)	463/14248 (3.24%)	304/15952 (1.92%)	163/15952 (1.02%)	253/16321 (1.5%)	
PBHL with CI intervention	5	11	5	18	21	7	67
PBHL with HA intervention	24	20	11	19	13	6	93
Earliest intervention start date	1/5/2006	7/9/2006	16/1/07	14/10/08	16/10/09	13/4/11	
Waiting for CI	1	0	0	3	2	6	12
Total PBHL infants	30/172	31/172	16/172	40/172	36/172	19/172	172
Total Uni PHL	156/1450	222/1450	447/1450	264/1450	127/1450	234/1450	1450

HL: Hearing loss.
 PBHL: Permanent hearing loss.
 Uni. PHL: Unilateral permanent hearing loss.

Table 3. Results of screening over the study period for all categories.

	2005	2006	2007	2008	2009	2010
Total WB screened	13085	13241	14248	15081	15952	16312
WB pass	10587[80.9%]	9330[70.46%]	9077[63.7%]	13884[92.06%]	9460[59.3%]	12194[74.74%]
WB referred	5205[39.77%]	4664[36.22%]	4738[33.25%]	5600[37.13%]	5401[33.85%]	4463[27.36%]
No. & % ref. WB	96[0.73%]	75[0.56%]	299[2.09%]	265[1.75%]	317[1.98%]	631[3.86%]
Total NICU	2666	1436	965	1340	1191	884
No. & % NICU pass	1262[47.35%]	1111[77.36%]	1563[61.74%]	1286[89.93%]	9688[1.27%]	500[56.56%]
No. & % NICU refer	59[2.61%]	180[12.63%]	52[3.32%]	61[4.55%]	68[5.7%]	36[7.2%]

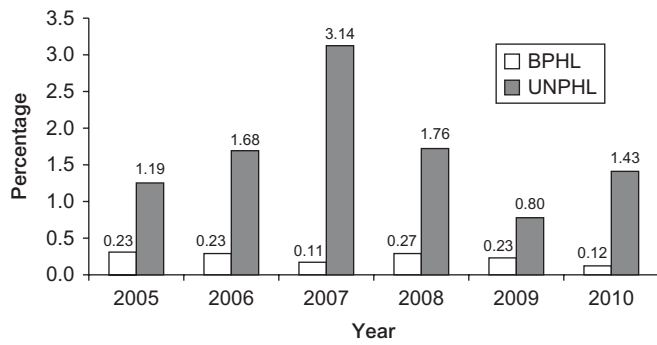


Figure 3. Percentage of unilateral and bilateral permanent hearing loss across year 2005 – 2010.

were identified. Cost estimate was 20218.98 QR for each case of hearing loss, whether bilateral or unilateral loss. The estimated cost for bilateral permanent sensorineural hearing loss (172 infants) was 190669 QR, (the equivalent of US \$52,381) for each case detected. The cost estimate for the whole procedure is shown in Table 5.

DISCUSSION

Each year, an estimated 5000 infants are born in USA with moderate, severe or profound bilateral hearing loss, with Bartter syndrome/sensorineural deafness (BSND) estimated in 1.5 per 1000 newborns. But this may be 10 to 20 times higher among infants in NICU vs. WBs. Prevalence of BSND also increases with other selected risk factors¹⁵ when compared with our study of the prevalence of permanent sensorineural hearing impairment in Qatar—1.9 per 1000 newborn babies at the end of 2010 and a total of 172 with BSNHL with HTL worse than > 40 dB. Infants were included in our intervention and rehabilitation program from 2005 to 2010. In addition, 1450 infants born with unilateral SNHL, congenital and conductive mixed loss as well as

auditory neuropathy/dyssynchrony attracted concern of health organizers in the country and required urgent management, because children with hearing loss experience delayed development in language, hearing and speech as early as the age three. Those affected face consequences throughout life, including lower reading abilities, poor social performance and under or unemployment.¹⁵ Thus it is rather imperative to accept the Newborn Hearing Screening Programme (NHSP) to be applied to all newborn children universally to prevent problems related to speech, language, social life and schooling. Qatar's NHSP involves as much as 97% of newborn infants in Qatar and is followed by children screening programs later in life, which involve early identification of hearing loss among children involved in primary and early school programs. Hearing loss detection programs are essential and aimed at early detection of hearing loss to avoid further repercussions of permanent hearing loss. These programs are considered essential among health care organizers.

The coverage range of newborns screened in our study from 2005 to 2010 range between 91.01% and 98.09%, which is high enough to achieve the quality indicator for screening established by the JICH [2007] which is more than 95%. This could help Qatar advance beyond missing many cases of infant hearing loss. Data on screening is frequently checked by more than one supervisor working in the program and by the program director.

Table 3 showed results of first and second stage screening in the WB infant group. The initial pass range screening in our study depended on screening results from 2005 through 2010, ranging from 27.23% to 39.77% with a mean of 34.43%, which is higher than what is found in the literature. The rate depends on lot of possible factors, among these are obstruction of the external auditory canal by vernix and debris or amniotic fluid, similar to that suggested by Vohr et al. 2001¹⁸ This can be overcome by one, second-day re-screening.

Table 4. Cost estimate of equipment used in screening over the study period.

SN	Material	Price per item	Total price in QR over 6 years	Number of items
1	DPOAE – Audex	30000 QR	300000 QR	10
2	DPOAE - Echoscreen	30000 QR	120000 QR	4
3	AABR - GSI	60000 QR	120000 QR	2
4	AABR - Abeer	90000 QR	180000 QR	2
5	Diagnostic ABR	100000 QR	100000 QR	1
6	Diagnostic ABR Vivasonic	66000 QR	198000 QR	3
7	Disposable electrodes	24000 QR/year	144000 QR	
			1162000	

Table 5. Cost estimate of screening study involving personnel salary over 6 years, 2005 – 2010.

SN	Job Description	Number of staff	Average Salary QR/month	Total Salary QR/year	Total salary for 6 years
1	Screening technicians	12	3800	45600	
2	Technician	6	8000	48000	
3	Senior technicians	5	6000	30000	
4	AVT& speech therapist	5	9600	48000	
5	Audiological physicians	5	50000	250000	
6	Administration staff	4	3400	136000	
Total				557600	3355600 QR

Referral rate results for WB ranged from 1.9% to 4.98%, with an average of 3.56%, which seemed within acceptable range.

Results also showed that the NICU referral rate percentile for NICU and risk babies ranged from 2.61% to 12.53% with an average of 5.98%, and the pass rate of the NICU subjects ranged from 47.35% to 89.93% with an average of 69.07%. Refer result for WB and NICU are within acceptable international averages of 7%.

False positive screening results produced inevitable worry among parents—this lead to the recommendation that special attention must be paid to improving the sensitivity and specificity of equipment in use.¹⁸

It seems probable that within five years at least two-thirds of all cases of PCHI in childhood will be diagnosed before the age of six months and that the profile of service provision to support children with such impairments will have undergone radical change compared to today.

Other objectives of NHSP include identifying babies who have hearing loss and providing necessary intervention as soon as possible. In our study, the age of hearing impairment was detected using diagnostic ABR and visual reinforcement audiometry and found to range across three different [hearing aid] fitting levels, from

that appropriate for a few weeks to that fitting a user 5 years of age. Newborns ranging from a few weeks to six months represented 12% of the total; the next group ranged from six months to 2.5 years and represented 69% of the total number, and the final group ranged above 2.5 years and represented around 16% of the population studied. It appears that most of hearing aids fitted and cochlear implant surgeries performed were provided at early and good time.

CONCLUSIONS

We have successfully screened more than 95% of infants over a six-year study period, with a resulting prevalence of permanent sensorineural hearing loss of around 1.9 per 1000 screened infants. The results are within the range of those internationally documented. Yet the prevalence of 16.5 per 1000 screened infants with unilateral severe sensorineural hearing impairment and congenital conductive HL needs to be reduced to universally accepted levels.

The identification of all newborns with hearing loss before the age of six months has now become an attainable and realistic goal. In the past, parents and pediatricians often did not suspect hearing loss until the age two to three years, after important speech and language milestones

Table 6. Cost of equipment involved in intervention.

	Item[intervention equipment]	Cost/year	Cost for 6 years[study period]	No. of items
8	Hearing aids	5000 QR	800000 QR	93
9	CI	120000 QR	2040000 QR	77
10	Disposables for HAs	5000 QR	30000 QR	
11	Spare parts for CI	100000 QR	600000 QR	
12	Hearing aids Lab	100000 QR		
13	HA lab disposables	2000 QR	12000 QR	
			3482000 QR	

Table 7. Total number of infants screened having bilateral permanent hearing loss divided for gender, nationality and rehabilitation services.

Years of birth	Number of Infants	Gender Female	Gender Males	Rehabilitation choice CI	Rehabilitation choice Hearing aid
2010	18	8	10	7	11
2009	24	8	16	11	13
2008	29	13	16	9	20
2007	20	5	15	9	10
2006	28	12	16	8	18
2005	53	20	33	19	34
Total number	172	66	106	63	107

have not been met. By the time these milestones are missed, the hearing impaired child had already experienced delay in speech and language development.

Technology allows for the development of screening tools that are reliable and that can be used to identify infants with hearing loss. Otoacoustic Emission and ABR testing can provide the audiologist with valuable information about an infant's level of hearing loss. There is a need for the implementation of hearing screening programs all over that globe, and there is a need apply easier and more sensitive methodology in testing for hearing loss in newborns.

Although the prevalence of BPHL in Qatar seems comparable with international figures, more effort is required to reduce it to minor levels. This effort will be enhanced mostly by the emerging work on genetics and chromosomal studies of the local population in Qatar. Specifically, there exists a need to reduce the percentage of HL development through reduction plans aimed at reduction of consanguineous marriages.¹⁵ It is worth noting that the screening program is proceeding well relative to the most established of similar international programs. Looking at the effectiveness of programs so far, continued efforts to improve screening along these lines in Qatar is highly worthwhile.

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