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Neonatal Hearing Loss in the Indigent

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Objective: **To compare the risk factor profile for neonatal hearing loss (HL), and the follow-up rate of those identified with HL in an indigent population with those in an insured population.** ***Study Design:*** **Retrospective review.** ***Methods:*** **We studied 4526 neonates from the high-risk nursery or neonatal intensive care unit from two adjacent hospitals in Houston, Texas. Ben Taub General Hospital (BTGH) is a county public hospital that serves mainly the indigent. Texas Children's Hospital (TCH) is a private tertiary care center that serves patients with private insurance and Medicaid.** ***Results:*** **Overall, 133 infants failed the screening test. Follow-up diagnostic testing identified 48 patients with definite HL. Although nearly twice as many patients at BTGH failed screening compared with TCH (88 vs. 45), four times as many patients at BTGH did not return for diagnostic testing (43 vs. 10). When a hearing aid was needed, there was a delay in getting one at BTGH (*P *<.05). There was a higher prevalence of dysmorphic facial features and central nervous system disease and a lower prevalence of long-term ventilatory support at BTGH (*P *<.05). There were no differences between BTGH and TCH in the prevalence of low birth weight, neonatal asphyxia, syndromic stigmata, neonatal infection, family history of HL, or neonatal transfusion (*P *>.1).** ***Conclusions:*** **Significant differences in the risk factor profile for neonatal HL exist between the indigent and the general population. A worrisome problem exists with the timely intervention in hearing-impaired indigent neonates.** ***Key Words:*** **Uninsured, Harris county, deafness, universal screening.**
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INTRODUCTION

The birth rate in the United States is 4,000,000 per year. From this population, there are estimated to be 4000 neonates with profound deafness and 37,000 with some type of hearing impairment.[1] Hearing impairment is 20 times more prevalent in neonates than other disorders that are routinely screened for, including phenylketonuria, sickle cell anemia, and hypothyroidism. The average age of identification of a congenital hearing impairment was 30 months in 1993.[1] Early auditory stimulation is necessary for the normal development of communication skills. The average deaf student graduates from high school with language and academic achievement levels below that of the average fourth-grade hearing student.[2,3] Maximal language function can be obtained if hearing is restored by 18 months of age.[4] More recent studies have shown that if hearing is restored by 6 months of age, hearing-impaired infants can catch up to normal-hearing infants in the development of speech and language.[5,6] Because early detection and intervention are critical in hearing-impaired infants, many states are adopting a policy of universal neonatal hearing screening.

Sufficient access to health care is of concern to the indigent population in the United States and to their health care providers. Access to health care by the indigent often includes substantial barriers.[7] It has been shown that in these situations, infants and children are among those who are most severely affected.[8] Although universal screening programs will detect those with hearing impairment, subsequent follow-up is required for definitive diagnosis and long-term intervention. The cost utility of such intervention has been proven in several studies.[9] There is a paucity of information about the delivery of these services and the follow-up of infants in the indigent population that have been identified with hearing loss (HL) by way of screening protocols.[10] Because all infants born in the United States deserve equal and quality care, the aim of this study was to compare the risk factor profile for neonatal HL, and the follow-up rate of those identified with HL in an indigent population with those in an insured, tertiary care population.

MATERIALS AND METHODS

This was a retrospective review of 4653 patients from either the neonatal intensive care unit or the high-risk nursery. Data were collected from two adjacent hospitals in Houston, Texas. Ben Taub General Hospital (BTGH) is a county hospital that has a birthing facility. This hospital serves an indigent, inner-city population. Seventy-five percent of all patients evaluated and treated by the otolaryngology service in this hospital are uninsured. The metropolitan area that this hospital serves has the

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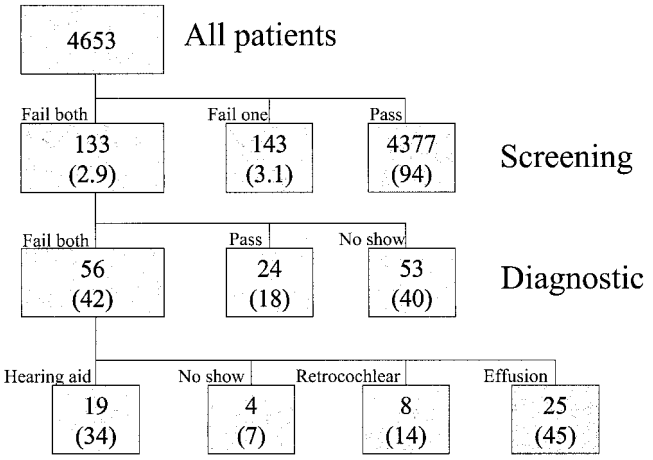

Fig. 1. Block diagram illustrating the results of neonatal hearing testing. Out of a total of 4653 patients screened, 133 failed the screening test in both ears. Of these patients, 56 patients failed the diagnostic test in both ears. Twenty-three patients were found to have HL severe enough to warrant a hearing aid: 19 patients followed through with this recommendation and 4 did not show up for hearing aid fitting. Eight patients were found to have retrocochlear HL, and 25 patients were found to have middle ear effusions.

highest rate of uninsured in the nation (approximately 30%), as well as a large recent immigrant population (including illegal aliens) that also has poor access to health care.[11] Analysis of the patients' racial distribution revealed a large minority population: 83% Hispanic, 12% black, 3% Asian-American, and 2% white (1999 data). A total of 2270 patients born between October 1994 and April 2000 were studied.

Texas Children's Hospital (TCH) is a tertiary care referral center with no birthing facilities. It serves both privately insured and Medicaid populations. A total of 2383 patients born between December 1997 and June 2000 were studied.

The audiologic testing protocol involved two stages. First, a screening test was performed during the initial hospitalization. This typically involved a screening bedside auditory brainstem evoked response using a stimulus intensity of 75 dB; however, during the later years of this study, transient evoked otoacoustic emission testing was performed. Patients who failed the screening test were referred back to the audiology department for diagnostic testing, including auditory brainstem evoked response using stepped stimuli to determine air and bone conduction thresholds. Otolaryngologic evaluation was performed on all patients found to have a HL by diagnostic testing. Additionally, infants who passed the screening test but were considered to be at high risk for delayed-onset HL (i.e., positive family history, in utero infection, neurofibromatosis type II, neurodegenerative disorders) were followed clinically with repeat testing every 6 months.

Intervention strategies varied depending on the pathology. Patients with middle ear effusions were treated initially with medical management; ventilating tubes were placed if this failed. Patients with congenital external or middle ear anomalies were initially fitted with hearing aids, with planning for later surgical correction as indicated. Patients with sensorineural HL were given a hearing aid trial as indicated by the degree of HL. If no benefit was obtained, consideration of a cochlear implant by 12 to 24 months was performed. Patients with retrocochlear HL (normal auditory brainstem evoked response wave 1, but delayed or malformed subsequent waves) were observed clinically with repeat testing every 6 months.

The data collected for each patient consisted of the screening and diagnostic test results, the age at the time of testing, and the risk factors for HL. Criteria for the risk factors are detailed in Table III. These data were entered into an MS Excel spreadsheet for statistical analysis. The data were analyzed using SPSS (SPSS Inc., Chicago, IL) version 8.0 using the χ^2 and analysis of variance tests to compare ordinate data (the risk factors and types of HL). The Fisher's exact test was used if the number of patients in a category was less than five. The Student t test was used to compare continuous variables (the gestational age and the age of testing). Multiple linear regression analysis was performed using a backward stepwise conditional mode protocol for risk factors with $P < .2$.

RESULTS

Patient Population

From a total of 4653 patients, our screening protocol found 133 patients who failed in both ears (Fig. 1). Patients who passed the screening test in one ear but failed in the other were not included in this study. Only 60% (80 of 133) of the patients who were requested to return for diagnostic testing did indeed return. Of the 80 patients who returned for diagnostic testing 56 (42%) failed again, whereas 24 (18%) passed. The overall rate of hearing impairment based on failing the second diagnostic testing was 1.2%, but this is probably an underestimate because less than two thirds of those who failed the screening test chose to follow-up with the diagnostic testing. Of the 56

TABLE I.
Timing of Hearing Screening.

	TCH	BTGH	Total	*P* value
Gestational age (wk)	37.2 ± 4.1 (n = 2383)	35.3 ± 4.3 (n = 2149)	36.3 ± 4.3 (n = 4532)	<.001*
Gestational age at time of screening test (wk)	40.5 ± 5.0 (n = 2360)	39.6 ± 5.7 (n = 2166)	40.1 ± 5.3 (n = 4526)	<.001*
Postpartum age at time of diagnostic test (wk)	22.8 ± 23.7 (n = 35)	28.2 ± 38.9 (n = 43)	25.8 ± 32.8 (n = 78)	.477
Postpartum age at time of hearing aid fitting (wk)	28.2 ± 17.1 (n = 10)	49.6 ± 24.7 (n = 9)	38.3 ± 23.2 (n = 19)	.041*

All values are in mean ± standard deviation.
* $P < .05$, Students t test.
TCH = Texas Children's Hospital; BTGH = Ben Taub General Hospital.

TABLE II.
Diagnostic Test Results.

	TCH	BTGH	Total
Definite hearing loss	28	28	56
Conductive	*8*	*15*	*23*
Mixed	*5*	*2*	*7*
Sensorineural	*10*	*8*	*18*
Retrocochlear pathology	5	3	8
Normal hearing	7	17	24
Missed test	10	43	53
Total	45/2383	88/2270	133/4653

TCH = Texas Children's Hospital; BTGH = Ben Taub General Hospital.

patients who failed diagnostic testing in both ears, 25 were found to have conductive HL resulting from middle ear effusions and were treated for this as described in the *Methods*. Of the 23 patients who were referred for hearing aids, 19 followed through and received them. There were 8 patients with retrocochlear HL, who were followed expectantly. These patients tended to be those with severe neurologic deficits, such as cerebral palsy.

Timing of Hearing Screening

The timing of hearing screening was similar between the two hospitals (Table I). Patient age at birth and at the time of the screening test was calculated relative to the time of conception (gestational age). Patient age at the time of diagnostic testing and at the time of hearing aid fitting is relative to their date of birth (postpartum age). There were minor differences between the two hospitals regarding patient gestational age and the timing of the screening test. The fact that the differences were statistically significant reflects only the large number of patients that were studied, and we think they have little clinical relevance. There was no statistically significant difference in the patient age at the time of diagnostic testing between the two hospitals. When a hearing aid was needed, there was a statistically significant delay at BTGH. This critical delay in intervention is the result of lack of hearing aid fitting capacity at the public hospital; these patients need to be referred to an outside facility. In contrast, TCH does in-house fitting and follow-up for hearing aids.

Diagnostic Testing

The results of the diagnostic testing for both BTGH and TCH demonstrate some important differences between these populations (Table II). Although nearly twice as many patients at BTGH failed the screening test compared with that at TCH (88 vs. 45), four times as many patients at BTGH did not return for diagnostic testing (43 vs. 10). Among the 56 patients who were positively identified with a HL, the type of HL varied between the hos-

TABLE III.
Risk Factors for Hearing Loss: All Patients.

	TCH	BTGH	Total	*P* value
Long-term ventilatory support	540/2188	24/2270	564/4458	<.001*
(>10 days)	(24.7)	(1.1)	(12.7)	
Low birth weight	297/2188	456/2266	753/4454	<.001*
(<1500 g)	(13.6)	(20.1)	(16.9)	
Ototoxic medication exposure	142/2188	1162/2269	1304/4457	N/A†
(>10 days at TCH, >3 days at BTGH)	(6.5)	(51.2)	(29.3)	
Neonatal asphyxia	221/2187	148/2270	369/4457	<.001*
(Apgar 0–4 at 1 min or 0–6 at 5 min)	(10.1)	(6.5)	(8.3)	
Dysmorphic facial features	33/2188	189/2270	222/4458	<.001*
(aural atresia, ear tags, cleft lip/palate)	(1.5)	(8.3)	(5.0)	
Syndromic stigmata	58/2187	53/2270	111/4457	.497
	(2.7)	(2.3)	(2.5)	
Neonatal infection	32/2188	233/2270	265/4458	<.001*
(sepsis, meningitis, syphilis, herpes, HIV, CMV)	(1.5)	(10.3)	(5.9)	
Central nervous system disease	12/2188	83/2270	95/4458	<.001*
(hydrocephalus, microcephalus, seizures)	(0.5)	(3.7)	(2.1)	
Family history of hearing loss	7/2188	14/2265	21/4453	.147
	(0.3)	(0.6)	(0.5)	
Neonatal blood transfusion	43/2188	13/2270	56/4458	<.001*
	(2.0)	(0.6)	(1.3)	
Maternal drug abuse	0/0	79/2270	79/2270	N/A‡
(alcohol or other drugs)		(3.5)	(3.5)	

Numbers in parentheses are percentages.
Not all data were available for every patient, so the total number of patients from each hospital varies between the risk factors.
* $P < .05$, Chi-square and Fisher exact tests.
† These data cannot be compared because they were collected differently from the two hospitals.
‡ These data cannot be compared because no data were available on maternal drug abuse from TCH.
TCH = Texas Children's Hospital; BTGH = Ben Taub General Hospital; HIV = human immunodeficiency virus; CMV = cytomegalovirus; N/A = not available.

pitals. Patients at BTGH had a higher prevalence of conductive HL and lower prevalence of mixed, sensorineural, and retrocochlear HL; however, these differences were not statistically significant ($P = .169$).

Risk Factors for HL Among All Patients

Of the risk factors for HL examined, the following statistically significant differences between patients at BTGH and those at TCH were identified: long-term ventilatory support, low birth weight, neonatal asphyxia, dysmorphic facial features, neonatal infection, central nervous system disease, and neonatal blood transfusion ($P < .05$) (Table III). There were no statistically significant differences between the two hospital populations for the prevalence of syndromic stigmata or family history of HL ($P > .1$). Risk factors relating to exposure to ototoxic medications and maternal drug use/abuse could not be compared because of incomplete data and/or different definitions of what constituted such a risk factor (see notes, Table III). Overall, these differences reflect the varied patient populations. Patients at TCH tended to be those with perinatal respiratory difficulties (such as bronchopulmonary dysplasia), whereas patients at BTGH are more typically those with maternal factors complicating their pregnancy (such as drug use, maternal syphilis, or HIV).

Risk Factors for HL Among Patients With HL

The 56 patients who failed both the screening and the diagnostic testing in both ears were considered to have a neonatal hearing impairment. The 8 patients with retrocochlear HL were excluded from further analysis, leaving 48 patients with conductive, mixed, or sensorineural HL. Risk factor data were available on 44 of these patients (Table IV). Statistical analysis revealed a significant difference between the prevalence of long-term ventilatory support, dysmorphic facial features, and central nervous system disease ($P < .05$). There was no statistically significant difference between BTGH and TCH in the prevalence of low birth weight, neonatal asphyxia, syndromic stigmata, neonatal infection, family history of HL, or neonatal transfusion ($P > .1$).

Multiple linear regression analysis was performed using the three risk factors with P values less than .2 (long-term mechanical ventilation, dysmorphic facial features, and central nervous system disease). Using a backward stepwise conditional mode, none of the terms could be removed from the equation without significantly affect-

TABLE IV.
Risk Factors for Hearing Loss: Hearing-Impaired Patients.

	TCH	BTGH	Total	*P* value
Long-term ventilatory support	9/19	1/25	10/44	<.001*
(≥10 days)	(47.4)	(4.0)	(22.7)	
Low birth weight	7/19	5/25	12/44	.214
(≤1500 g)	(38.6)	(20.0)	(27.3)	
Ototoxic medication exposure	7/19	11/25	18/44	N/A†
(>10 days at TCH, >3 days at BTGH)	(36.8)	(44.0)	(40.9)	
Neonatal asphyxia	2/19	2/25	4/44	1.000
(Apgar 0–4 at 1 min or 0–6 at 5 min)	(10.5)	(8.0)	(9.1)	
Dysmorphic facial features	2/19	12/25	14/44	.010*
(aural atresia, ear tags, cleft lip/palate)	(10.5)	(48.0)	(31.8)	
Syndromic stigmata	4/19	4/25	8/44	.710
	(21.1)	(16.0)	(18.2)	
Neonatal infection	3/19	3/25	6/44	1.000
(sepsis, meningitis, syphilis, herpes, HIV, CMV)	(15.8)	(12.0)	(13.6)	
Central nervous system disease	0/19	7/25	7/44	.014*
(hydrocephalus, microcephalus, seizures)	(0.0)	(28.0)	(15.9)	
Family history of hearing loss	1/19	1/25	2/44	1.000
	(5.3)	(4.0)	(4.5)	
Neonatal blood transfusion	1/19	0/25	1/44	.432
	(5.3)	(0.0)	(2.3)	
Maternal drug abuse	0/0	1/25	1/25	N/A‡
(alcohol or other drugs)		(4.0)	(4.0)	

Numbers in parentheses are percentages.

Not all data were available for every patient, so the total number of patients from each hospital varies between the risk factors.

* $P < .05$, Chi-square and Fisher exact tests.

† These data cannot be compared because they were collected differently from the two hospitals.

‡ These data cannot be compared because no data were available on maternal drug abuse from TCH.

TCH = Texas Children's Hospital; BTGH = Ben Taub General Hospital; HIV = human immunodeficiency virus; CMV = cytomegalovirus; N/A = not available.

ing the probability of the likelihood–ratio statistic based on conditional parameter estimates. The fit values were long-term mechanical ventilation (B = 1.91, P = .107), dysmorphic facial features (B=-1.99, P = .034), and central nervous system disease (B=-9.30, P = .799). A constant value was included as well (B = 9.09, P = .804).

We compared the types of hearing impairment (conductive, mixed, and sensorineural) associated with the three risk factors that were different between the two hospitals (Table V). None of the risk factors were associated with a specific type of HL (ANOVA, $P > .1$). However, as might be expected, patients with dysmorphic facial features tended to have more conductive hearing losses than those without this risk factor. The types of dysmorphic facial features consisted of cleft lip and/or palate (5), generalized craniofacial anomalies (4), ear tags (2), and aural atresia (2). The types of central nervous system diseases present were hydrocephalus (4), microcephalus (1), intraventricular hemorrhage (1), and seizures of unknown origin (1).

DISCUSSION

Risk Factors

Our study indicates that the risk factor profile for neonatal HL in the Houston indigent population is similar to that in the tertiary care referral population. However, we found that the prevalence of dysmorphic facial features and central nervous system disease was higher and the prevalence of long-term ventilatory support was lower in the indigent. The reason that more patients required ventilatory support at TCH is understandable because they are referred a substantial number of premature infants (<32 wks) who often have bronchopulmonary dysplasia and require mechanical ventilation. In contrast, the reason why there were more patients with dysmorphic features and central nervous system disease at BTGH is unclear. Based on our statistical analysis, it does not appear that other comorbidities exist. Additionally, maternal drug abuse is probably not linked to these risk factors, because there was only one patient with HL at BTGH with a history of maternal drug use. It is possible that patients with non-life-threatening dysmorphic features are not referred to TCH from outlying hospitals, and are simply managed as outpatients. The difference between the two populations screened in the prevalence of patients with central nervous system deficits is surprising and difficult to explain, because one would expect most neonates born in the surrounding community with these severe conditions to be referred to a specialized children's hospital (Texas Children's Hospital) for evaluation.

Screening for HL

It should be noted that patients were typically in the hospital for 3 to 4 weeks after they were born before their hearing was screened. This may explain our lower rate of failed screening tests compared with that typically reported in the literature for a single modality screening protocol (6% vs. 9%–26%).[12–15] Presumably, debris in the ear canals or fluid in the middle ear cleft had time to resolve after birth and did not elevate the false-negative rate of hearing screening. The patients with conductive HL are clearly not otherwise normal infants with a middle ear effusion that might be expected to clear spontaneously. For the most part, these are infants with dysmorphic facial features and chronic effusions associated with a significant HL that need to be managed aggressively. It is important to identify these infants early.

High-risk stratification for the process of neonatal hearing screening distills the general population down to one with a higher prevalence of HL. Unfortunately, this is not too useful for hearing screening because 50% of hearing-impaired babies have no risk factors.[16–18] For

TABLE V.
Type of Hearing Loss by Risk Factor.

	Conductive	Mixed	Sensorineural	Total	*P* value
Long-term ventilatory support					
Yes	6	1	3	10	.768
	(60.0)	(10.0)	(30.0)	(100)	
No	16	5	13	34	
	(47.1)	(14.7)	(38.2)	(100)	
Dysmorphic facial features					
Yes	9	3	2	14	.105
	(64.3)	(21.4)	(14.3)	(100)	
No	13	3	14	30	
	(43.3)	(10.0)	(46.7)	(100)	
Central nervous system disease					
Yes	3	0	4	7	.331
	(42.9)	(0.0)	(57.1)	(100)	
No	19	6	12	37	
	(51.4)	(16.2)	(32.4)	(100)	

ANOVA test comparing conductive vs. mixed. vs. sensorineural hearing loss. There was no statistically significant difference for any of the risk factors.

this reason, universal hearing screening has been shown to be successful even when applied to an economically depressed, inner-city environment, when given adequate resources.[19] Although universal screening is definitely needed to permit the early detection of HL in a tertiary care hospital setting, we think the use of risk factors is also important. For our study, this strategy proved useful because it provided a higher yield of hearing-impaired patients.

Intervention

Early intervention is the ultimate goal of state governments that implement universal newborn screening programs. Approximately half of the indigent patients identified with hearing impairment at initial screening failed to return and were lost to subsequent follow-up. This is a dramatically higher rate, a fourfold difference, than that of the group examined at the tertiary care facility. Some factors that might be involved in this discrepancy for indigent care in general are: 1) parental misunderstanding of the gravity of the situation, 2) poor access to transportation, 3) financial burden to the family for lost time from work as a result of repeat visits, 4) inefficiency in the system requiring multiple visits for the parents and infant resulting in frustration, 5) significant barriers to obtaining health care benefits that will cover the cost of the intervention, and 6) insufficient resources at the institution to provide for the intervention.[11,20]

Once a HL is diagnosed, rapid treatment is needed. However, the indigent often does not get this. At BTGH, the time to fitting of a hearing aid was 22 weeks longer than that at TCH, mainly because of the logistics of getting the patients sent to an outside facility. These types of issues are critical to identify and resolve to achieve the full benefit of universal neonatal hearing screening.

CONCLUSION

In infants identified with a hearing impairment, significant differences between the prevalence of long-term ventilatory support, dysmorphic facial features, and central nervous system disease exist between the indigent and the general population. Screening methods can be adapted to take advantage of this knowledge to increase detection efficiency.

Although diagnosis of a neonatal hearing impairment can be implemented for the indigent, a significant and worrisome problem exists with the timely intervention and follow-up in those patients identified with a HL.

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