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Late Newborn Hearing Screening, Late Follow-up, and Multiple Follow-Ups Increase the Risk of Incomplete Audiologic Diagnosis Evaluation

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Abstract

This study aimed to determine whether the following factors were associated with an incomplete audiologic diagnosis evaluation (IAD): age at newborn screening, length of time between newborn hearing screening (NHS) and first follow-up, and total number of follow-ups. 2011-2013 linked Louisiana Early Hearing Detection and Intervention data and birth records were analyzed. Logistic regression models were used to evaluate different effects of the predictors on IAD among birth weight groups. In very low birth weight newborns, there were no statistical associations of IAD with age at NHS or length of time between NHS and first follow-up, but there was with the number of follow-up appointments. Among low birth weight or normal weight newborns, risk of IAD was significantly increased in babies with NHS > 30 days of age; length of time between NHS and first follow-up > 30 days; and having more than one follow-up. In order to reduce the number of infants who fail to complete the audiologic diagnosis evaluation, it is necessary to conduct NHS early, expedite follow-up, and decrease the number of follow-ups.

Key Words: Early Hearing Detection and Intervention, newborn hearing screening, audiologic diagnosis, lost to follow-up

Acronyms: ABR = auditory brainstem response, CDC = Centers for Disease Control and Prevention, EHDI = Early Hearing Detection and Intervention, IAD = incomplete audiologic diagnosis evaluation, JCIH = Joint Committee on Infant Hearing, LBW = low birth weight, LFU = lost to follow-up, LTD = lost to documentation, NICU = neonatal intensive care unit, NHS = newborn hearing screening, OAE = otoacoustic emissions, VLBW = very low birth weight

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Introduction

Hearing loss plays a crucial role in delayed development of speech, language, and cognition in early childhood (Bess, Dodd-Murphy, & Parker, 1998; Holt & Svirsky, 2008; Moeller, 2000; Nicholas & Geers, 2006). Previous studies showed that children with hearing loss who received intervention services before 6 months of age had significantly better academic achievement and language development than those who received them after 6 months of age (Yoshinaga-Itano, 2003, 2004). Early Hearing Detection and Intervention (EHDI) programs in the United States are designed to detect congenital and early acquired hearing loss and link infants and their families to appropriate intervention services. The Joint Committee on Infant Hearing (JCIH) recommends that all newborns be screened at no later than 1 month of age; diagnosis be completed at no later than 3 months of age for infants who do not pass screening; and appropriate intervention be received at no later than 6 months of age for infants identified with hearing loss (JCIH, 2007). Hearing screenings and diagnoses completed after recommended timelines are considered barriers to the effectiveness of EHDI programs (White & Blaiser, 2011). Although the programs have effectively identified many children with early childhood hearing loss in recent years (Centers for Disease Control and Prevention [CDC], 2010; Muñoz, Blaiser, & Barwick, 2013), results of diagnostic tests for

children who fail hearing screening are not consistently reported to the EHDI programs (Williams, Alam, & Gaffney, 2015). Based on 2013 National CDC EHDI data, the rate of undocumented audiologic diagnosis was 41.0% among infants who did not pass the newborn hearing screening (CDC, 2013).

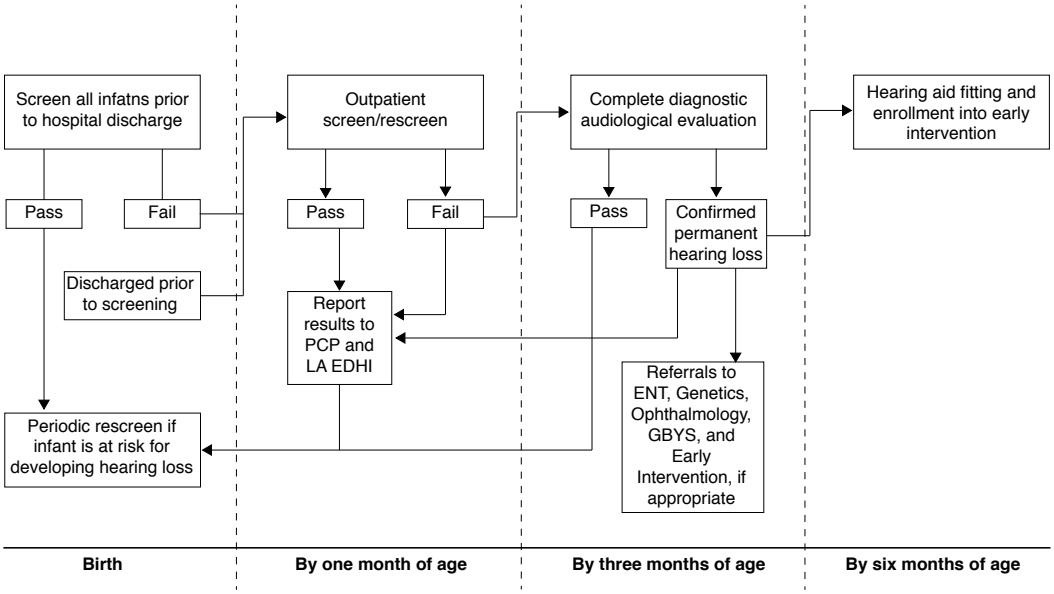
In Louisiana, all children who do not pass the final newborn hearing screening before hospital discharge are referred for hearing rescreening by audiologists or physicians at outpatient clinics. Of those who do not pass hearing rescreening, a referral is made for further evaluations to complete the audiologic diagnosis. Figure 1 presents a detailed process of newborn hearing screening, diagnosis, and intervention in Louisiana. In this study, a timeline for an audiologic diagnostic evaluation was defined as the time from the hearing rescreening at an outpatient clinic to the time when the audiologic diagnosis was completed. In Louisiana, in fact, many children undergo a prolonged and incomplete audiologic diagnosis process— in particular children with late newborn hearing screening, late follow-up, and many follow-ups. With a hypothesis that late newborn hearing screening, late follow-up, and multiple follow-up visits may increase the risk of incomplete audiologic diagnosis, this study was conducted to identify associations between incomplete audiologic diagnosis and (a) age at final newborn hearing screening prior to discharge, (b) length of time between final newborn hearing screening prior to discharge and first follow-up, and (c) total number of follow-ups among newborns who failed newborn hearing screening prior to hospital discharge. To our knowledge there are no published studies evaluating these associations.

Method

Study population

The study included children who were born in Louisiana between 2011 and 2013; had newborn hearing screening prior to hospital discharge, but did not pass; and completed at least one follow- up at an outpatient clinic. All follow-ups mentioned in the study were conducted at outpatient clinics by audiologists or physicians if the follow-up was for rescreening, and only by audiologists for audiologic diagnosis. The first follow-up was always for the hearing rescreening. The term screening in the study refers to hearing screening conducted before hospital discharge.

As mentioned previously, the timeline for an audiologic diagnostic evaluation was defined as the time from the hearing rescreening at an outpatient clinic to the time when the audiologic diagnosis was completed. The following children were excluded from the study: children whose mothers were not Louisiana residents at birth, children who died after hearing screening regardless of receiving any follow-up, or children who were reported as lost to follow-up (LTF; i.e., testing providers reported children did not show up at the time of the scheduled follow-up appointment, the family was unable to be contacted, or was contacted but unresponsive) or lost to documentation (LTD; i.e., the Louisiana [LA] EHDI program did not receive any report or documentation of follow-up or LTF from audiologists or physicians).



Note. ENT = Ear Nose Throat; GBYS = Guide By Your Side (a program providing family support services); LA EHDI = Louisiana early hearing detection and intervention; PCP = Primary Care Physician.

Figure 1: Louisiana Early Hearing Detection and Intervention (LA EHDI) Process of Screening, Diagnosis, and Intervention

Data sources and linkages
Three data sets were linked and used for analyses including: 2011-2013 birth certificates, 2011– 2013 newborn hearing screening (NHS), and 2011–Sept 2014 hearing screening follow-up (after NHS). NHS data provided screening status, screening date, and screening methods. Screenings defined as failed or passed were dependent upon results of final tests before hospital discharge using either auditory brainstem response (ABR) or otoacoustic emissions (OAE). Follow-up data provided follow-up status, time at follow-up, and number of follow-ups.

The data linkages were conducted using SAS 9.3 and LinkPro 3.0. First, failed NHS data were linked to birth certificates by child's date of birth, first name, and last name with Soundex codes (codes of names based on the phonetic spelling of the name). Linked records were reviewed manually to define true matches using linking variables and the following variables: mother's last name, first name, maiden name, address of residence at birth or most updated address of residence, and birthing hospital. Second, the NHS-birth records matched data were linked to follow-up data by a unique identification number assigned by the LA EHDI database system. The match rate was 99.2% and 100% for the first and second linkage, respectively.

Analysis variables
Outcome variable. The outcome variable was classified as incomplete or complete audiologic diagnosis. An incomplete audiologic diagnosis (IAD) was defined if an infant (a) failed all newborn hearing screenings before hospital discharge, (b) completed at least one follow-up conducted by an audiologist or physician, and (c) did not have a conclusive audiologic diagnosis confirmed by an audiologist at the time of the study. As mentioned before, all follow-ups mentioned in the study were conducted at outpatient clinics by audiologists or physicians if the follow-up was for rescreening, and only by audiologists for audiological diagnosis. The first follow-up was always a hearing rescreening which marked the beginning of the diagnostic evaluation in this study. A complete audiologic diagnosis was defined, at the last follow-up, if an infant passed both ears with rescreening tests, if diagnostic findings indicated hearing threshold levels within normal limits, or if permanent hearing loss was confirmed. There was no specific time limit applied in definition of the study outcome variable. The LA EHDI program follows hearing status from birth to five years of age. At the time the study was conducted (September 2014), children with IAD were still in process of hearing loss diagnosis but had not yet had a conclusive audiologic diagnosis from the last follow-up between 2011 and 2014.

Predictor variables. There were three predictor variables used: age at NHS, time between NHS and first follow-up, and total number of follow-ups.

Age at NHS. Age in days was calculated using date of birth and date of newborn hearing screening prior to hospital discharge. If there was more than one screening, the date of the last screening was used for calculation. Age was categorized into < 30 days and > 30 days.

Time between NHS and first follow-up. The length of time between date of NHS and date of first follow-up was calculated. If there was more than one screening prior to hospital discharge, the date of the last screening was used for calculation. The time was grouped into < 30 days and > 30 days.

Total number of follow-ups. A sum of all follow-ups that an infant completed. The variable was grouped into one and more than one follow-up.

Covariates
All covariates were derived from birth certificates and defined as categorical variables. Birth weight was categorized into very low birth weight (VLBW, < 1,500 g), low birth weight (LBW, 1,500 g–2,499 g), and normal birth weight (> 2,500 g). Child's neonatal intensive care unit (NICU) admission was not included as another covariate in the study. NICU admission was considered underreported and the length of time of stay was not reported in birth certificates. However, this variable had a strong collinear relationship with birth weight because

Table 1. Population Characteristics (%) by Age at Newborn Hearing Screening (NHS), Time Between NHS and First Follow-up, and Total Number of Follow-ups

		Age at Newborn hearing screening (days)			Time between NHS and first follow-up (days)			Total Number of follow-ups		
		<30	30+	*p* value*	<30	30+	*p* value*	One	Two+	*p* value*
Race/Ethnicity	NH White	45.9	30.6	<.0001	47.5	40.7	<.0001	45.4	44.9	0.1025
	NH Black	42.2	62.6		41.6	45.8		43.0	42.6	
	NH Other	4.0	2.3		3.7	4.5		3.7	5.3	
	Hispanic	7.9	4.5		7.2	9.0		7.9	7.2	
Maternal age	<20	11.6	9.4	0.1542	11.7	11.0	0.4800	11.6	11.0	0.6831
	20-34	79.1	78.1		79.0	79.2		79.1	78.9	
	>34	9.3	12.5		9.2	9.9		9.3	10.1	
Maternal Education	< High school (19%)	21.1	21.9	0.8407	19.9	23.6	<.0001	21.1	21.6	0.2611
	High school (33%)	31.5	29.8		30.8	32.7		31.1	33.3	
	>High school (48%)	47.4	48.3		49.3	43.7		47.8	45.1	
Married at birth	No	58.3	55.8	0.4198	57.2	60.7	0.0056	58.2	58.6	0.7892
	Yes	41.7	44.2		42.8	39.3		41.8	41.4	
Delivery method	Vaginal	66.3	30.6	<.0001	66.6	61.4	<.0001	66.2	57.2	<.0001
	C-Section	33.7	69.4		33.4	38.6		33.8	42.8	
Delivery payment method	Non-Medicaid	29.3	32.5	0.2631	30.1	27.8	0.0433	29.3	29.5	0.9072
	Medicaid	70.7	67.5		69.9	72.2		70.7	70.5	
Area of residence	Rural	35.3	34.0	0.6643	36.5	32.5	0.0011	34.9	37.3	0.1329
	Urban	64.7	66.0		63.5	67.5		65.1	62.7	
Previous live births	None	41.2	44.2	0.5729	43.2	37.7	<.0001	41.7	39.1	0.2691
	One	31.1	28.5		30.4	32.0		30.7	32.8	
	Two+	27.7	27.3		26.4	30.4		27.6	28.0	
Sex	Female	58.3	54.3	0.2056	58.6	57.1	0.2425	58.2	57.4	0.6411
	Male	41.7	45.7		41.4	42.9		41.8	42.6	
Plurality`	Singleton	97.2	82.6	<.0001	97.4	95.0	<.0001	96.8	95.1	0.0043
	Twin+	2.8	17.4		2.6	5.0		3.2	4.9	
Birth weight	VLBW	0.7	79.6	<.0001	2.4	6.3	<.0001	2.6	10.5	<.0001
	LBW	8.7	10.6		7.6	11.2		8.4	11.1	
	Normal Weight	90.7	9.8		90.1	82.5		89.1	78.4	
Age at newborn screening	<30 Days	-	-		97.6	93.6	<.0001	97.2	90.0	<.0001
	30+ Days	-	-		2.4	6.4		2.8	10.0	
Time between NHS and first follow-up	<30 Days	68.3	44.0	<.0001	-	-		69.3	55.9	<.0001
	30 Days	31.7	56.0		-	-		30.7	44.1	
Total number of follow-ups	One	86.9	63.4	<.0001	88.5	81.2	<.0001	-	-	
	Two+	13.1	36.6		11.5	18.8		-	-	

*Chi-square p value.
Note. NH: Non-Hispanic; VLBW = very low birth weight; LBW = low birth weight

all VLBW babies were admitted to NICU. Thus, presence of NICU admission in adjusted regressions of data analyses was not necessary. Table 1 shows distributions of all covariates by age at NHS, time between NHS and first follow-up, and total number of follow-ups.

Data analysis

Percentages and 95% confidence intervals of IAD by predictors were calculated. Confidence intervals were estimated by using the normal approximation method of the binomial confidence interval. Logistic regression models were used to determine associations between IAD and predictors. To address confounding in adjusted regression models all covariates were controlled. In fact, all VLBW babies are admitted into the NICU, typically for extended stays, and therefore have late newborn hearing screening. Because of VLBW newborns' long-term NICU stay and medical characteristics that are very different from other groups (low birth weight and normal birth weight), effects of predictors on IAD were evaluated in each group and also compared together among different groups of birth weight by including interaction terms between birth weight and predictors in models. Specifically, there were three analyses using logistic regression models to assess the associations described as follows:

Association of IAD with age at NHS. In the unadjusted model, the independent variables consisted of age at NHS, birth weight, and the interaction between age at NHS and birth weight. All covariates were added in adjusted model.

Association of IAD with length of time between NHS and first follow-up. In the unadjusted model, the independent variables consisted of time between NHS and first follow-up, birth weight, and the interaction between time between NHS and first follow-up and birth weight. All covariates plus age at NHS were added in the adjusted model.

Association of IAD with number of follow-up. In the unadjusted model, the independent variables consisted of number of follow-ups, birth weight, and the interaction between number of follow-ups and birth weight. All covariates plus age at NHS and time between NHS and first follow-up were added in the adjusted model.

All final adjusted models included only variables with p value < 0.05. Data analyses were conducted in SAS 9.3.

The project was deemed exempt by the Louisiana State University Institutional Review Board because it did not meet the federal definition of human subjects research.

Results

There were 6,970 children included in the study. A majority of children (96.2%) completed NHS before 30 days of age and completed one follow-up (86.1%). The percent of children who completed the first follow-up before 30 days after NHS was 67.4%.

The overall rate of IAD was 6.9% (CI: 6.3–7.5). The rate was very high among newborns with NHS at 30 days of age or older (25.7%) compared to those with NHS within 30 days of age (6.1%). Stratified by birth weight, this difference was also seen among newborns with low birth weight or normal weight (LBW: 8.1% for age at NHS < 30 days vs. 25.0% for age at NHS > 30 days; normal weight: 5.8% for age at NHS < 30 days vs. 26.9% for age at NHS > 30 days). However, among newborns with VLBW, the rate was very high in both age groups and was not statistically different (20.5% for age at NHS < 30 days vs. 25.6% for age at NHS > 30 days; $t(6959) = 0.72$, $p = 0.4734$).

For the length of time between NHS and first follow-up, the rate of IAD was 5.0% with the length < 30 days and it doubled with the length > 30 days (10.5%). Stratified by birth weight, this difference was seen among newborns with LBW and normal weight (LBW: 6.8% for the length < 30 days vs. 11.9% for the length > 30 days; normal weight: 4.3% for the length < 30 days vs. 9.4% for the length > 30 days). Similar to age at NHS, among babies with VLBW the rate was very high in both groups and was not statistically different (26.4% for the length < 30 days vs. 23.2% for the length > 30 days; $t(6,929) = -0.57$, $p = 0.5682$).

For the number of follow-ups, the rate of IAD was 4.8% among newborns with one follow-up and it was almost four times higher among those who had more than one follow-up (19.6%). Stratified by birth weight, the rate was high and statistically different between groups among newborns with VLBW, LBW, and normal weight: VLBW: 20.1% for one vs.

Table 2. Percentage of Incomplete Audiologic Diagnosis by Age at Newborn Hearing Screening (NHS), Time between NHS and First Follow-up, and Total Number of Follow-ups Stratified by Birth Weight.

	Age at newborn hearing screening (days)			Time between NHS and first follow-up (days)			Total number of follow-ups		
Birth Weight		*n*	Percent, 95% CI		*n*	Percent, 95% CI		*n*	Percent, 95% CI
VLBW	<30	9	20.5, 8.5–32.4	<30	29	26.4, 18.1–34.6	One	31	20.1, 13.8–26.5
	30+	54	25.6, 19.7–31.5	30+	33	23.2, 16.3—30.2	Two+	32	31.4, 22.4–40.4
	Total	63	24.7, 19.4–30.0	Total	62	24.6, 19.3–29.9	Total	63	24.6, 19.3–29.9
LBW	<30	47	8.1, 5.9–10.3	<30	24	6.8, 4.2–9.4	One	34	6.8, 4.6–9.0
	30+	7	25.0, 9.0–41.0	30+	30	11.9, 7.9–15.8	Two+	20	18.5, 11.2–25.8
	Total	54	8.9, 6.6–11.1	Total	54	8.9, 6.6–11.2	Total	54	8.9, 6.6–11.1
Normal Weight	<30	354	5.8, 5.2–6.4	<30	181	4.3, 3.7–4.9	One	223	4.2, 3.6–4.7
	30+	7	26.9, 9.9–44.0	30+	175	9.4, 8.1–10.7	Two+	138	18.1, 15.4–20.9
	Total	361	5.9, 5.3–6.5	Total	356	5.9, 5.3–6.4	Total	361	5.9, 5.3–6.5

Note. VLBW = very low birth weight; LBW = low birth weight.

31.4% for more than one follow-up ($t(6,962) = 2.03$, $p = 0.0423$); LBW: 6.8% for one vs. 18.5% for more than one follow-up ($t(6,962) = 3.74$, $p = 0.0002$); and normal weight: 4.2% for one vs. 8.1% for more than one follow-up ($t(6,962) = 14.0$, $p < 0.0001$). Table 2 presents percentage of IAD by age at NHS, time between NHS and first follow-up, and total number of follow-ups stratified by birth weight.

Adjusted regression models showed that associations of IAD with the predictors varied among birthweight groups. The interactions were significant between birthweight and age at NHS ($F(2, 6,863) = 3.13$, p = 0.0439); length of time between NHS and first follow-up ($F(2, 6,859) = 5.37$, p = 0.0047); and number of follow-ups ($F(2, 6,858) = 4.59$, p = 0.0101). Among VLBW newborns, there were no statistical associations of IAD with age at NHS (Odds Ratio [OR]: 1.3, CI: 0.6–3.0) or the length between NHS and first follow-up (OR: 0.8, CI: 0.5–1.2); however, the association of IAD with number of follow-ups was found (OR: 1.9, CI: 1.0–3.4). Among newborns with LBW, odds of IAD was significantly higher in babies with NHS > 30 days of age (OR: 3.8, CI: 1.5–9.4); the length of time between NHS and first follow-up > 30 days (OR: 1.8, CI: 1.0–3.2); and more than one follow-up (OR: 2.9, CI: 1.6–5.3). Among normal weight newborns, odds of IAD was also found statistically higher in babies with NHS > 30 days of age (OR: 6.0, CI: 2.5–14.3); the length of time between NHS and first follow-up > 30 days (OR: 2.3, CI: 1.9–2.9); and more than one follow-up (OR: 4.7, CI: 3.4–6.0). See Table 3.

Table 3: Odds Ratio (OR) Estimates and 95% Confidence Interval (CI) for Associations between Incomplete Audiologic Diagnosis and Age at Newborn Hearing Screening (NHS), Time between NHS and First Follow-up, and Total Number of Follow-ups

Age at newborn hearing screening (days)

		Unadjusted			Adjusted		
		OR, 95%CI	*t* statistic value	*p* value	OR, 95%CI	*t* statistic value	*p* value
Very low birth weight	<30	1.0			1.0		
	30+	1.3, 0.6–3.0	0.72	0.4734	1.2, 0.6–2.8	0.54	0.5895
Low birth weight	< 30	1.0			1.0		
	30+	3.8, 1.5–9.4	4.00	0.0040	4.0, 1.6–10.1	2.99	0.0028
Normal birth weight	<30	1.0			1.0		
	30+	6.0, 2.5–14.3	2.88	<0.0001	5.3, 2.1–13.4	3.49	0.0005

Time between NHS and first follow-up (days)

		Unadjusted			Adjusted		
		OR, 95%CI	*t* statistic value	*p* value	OR, 95%CI	*t* statistic value	*p* value
Very low birth weight	<30	1.0			1.0		
	30+	0.8, 0.5–1.5	–0.57	0.5682	0.8, 0.4–1.5	–0.69	0.4916
Low birth weight	< 30	1.0			1.0		
	30+	1.9, 1.1–3.2	2.14	0.0323	1.8, 1.0–3.2	2.00	0.0460
Normal birth weight	<30	1.0			1.0		
	30+	2.3, 1.9–2.9	7.61	<0.0001	2.3, 1.9–2.9	7.54	<0.0001

Total number of follow-ups

		Unadjusted			Adjusted		
		OR, 95%CI	*t* statistic value	*p* value	OR, 95%CI	*t* statistic value	*p* value
Very low birth weight	One	1.0			1.0		
	Two+	1.8, 1.0–3.2	2.03	0.0423	1.9, 1.0–3.4	2.04	0.0414
Low birth weight	One	1.0			1.0		
	Two+	3.1, 1.7–5.7	3.74	0.0002	2.9, 1.6–5.3	3.39	0.0007
Normal birth weight	One	1.0			1.0		
	Two+	5.1, 4.0–6.4	14.00	<0.0001	4.7, 3.7–6.0	13.00	<0.0001

Discussion

This study showed that children with late NHS (> 30 days of age), late follow-up (> 30 days after NHS), and multiple follow-ups were more likely not to complete the audiologic diagnosis process. Effects of age at NHS, timing of follow-up, and number of follow-ups on IAD varied among birth weight groups. With the presence of VLBW, the rate of IAD was very high (> 20%) regardless of time when NHS and first follow-up were completed or number of follow-ups, and associations of IAD with age at NHS and length of time between NHS and first follow-up did not exist. However, the association was seen with number of follow-ups. For LBW or normal weight groups, the rate was consistent between two groups and higher among those who had late NHS, late follow-up, and multiple follow-ups. The risk difference of IAD between groups of the predictors was fairly similar among LBW and normal weight newborns and larger than one among VLBW newborns.

It was clear that VLBW had a strong effect on IAD as well as late NHS, late follow-up, and multiple follow-ups. The main reason of late NHS was VLBW. The data showed that more than 80% of VLBW babies had NHS after 30 days. VLBW babies who normally have severe medical conditions often have long-term hospital stays, particularly in the NICU where procedures of medical stabilization are required (Berry, Shah, Brouillette, & Hellmann, 2008), thus the NHS is delayed until just prior to initial discharge. For parents of those babies, appointments for medical conditions may take priority over hearing follow-up appointments. Thus, hearing follow-up appointments could be missed, ignored, or delayed which leads to IAD or late follow-up, respectively. In fact, VLBW newborns are more likely to get many follow-ups. Those babies are harder to test due to very small ear canals. In addition, a very small head sometimes makes it harder to obtain results on bone conduction testing. Thus, more tests are needed before confirming the diagnosis. Table 2 presents the difference of late NHS, late follow-up, and number of follow-ups by birth weight.

The study indicated that when stratified by birth weight, age at NHS, timing of follow-up, and number of follow-ups had different effects on IAD. Specifically, LBW or normal weight babies with late NHS, late follow-up, and multiple follow-ups had a higher risk of IAD. Although the associations were well defined in LBW and normal weight babies, their underlying mechanisms were not clear. Therefore, further research is needed to understand the

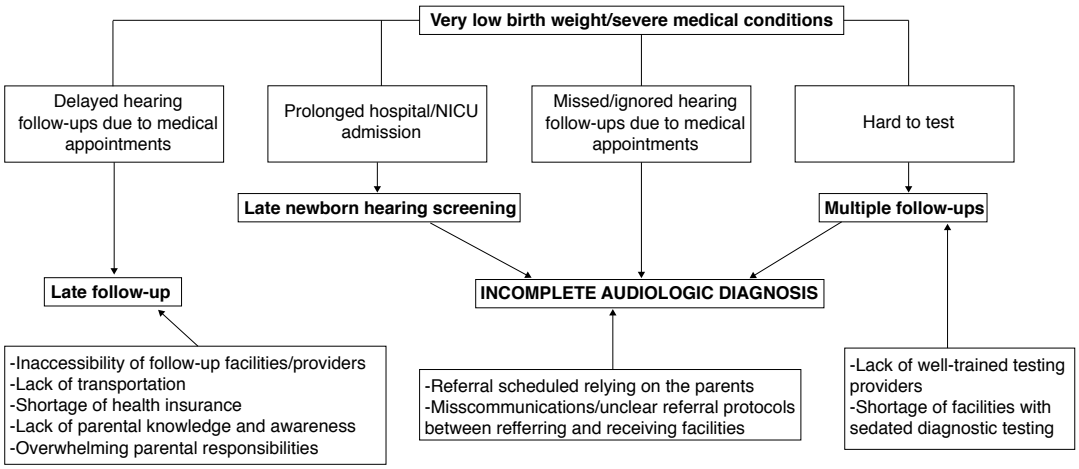


Figure 2: Possible Reasons of Late Newborn Hearing Screening, Late Follow-ups, and Multiple Follow-ups and Pathway Leading to Incomplete Audiologic Diagnosis

mechanisms of associations as well as reasons of late NHS, late follow-up, and multiple follow-ups among non-VLBW babies, particularly among normal weight babies which consisted of about 90% of the total study population. A large reduction of late follow-up and multiple follow-ups in normal weight babies would have a significant impact on a decrease of late follow-up and multiple follow-ups as well as an improvement of IAD in the whole study population. The following factors may be some of the possible reasons of late follow-up in normal weight babies who have fewer medical conditions: inaccessibility to follow-up facilities or providers; lack of transportation, particularly in rural areas; lack of health insurance; lack of parents' knowledge and awareness of the importance of early diagnosis of hearing loss (Shulman et al., 2010); and overwhelming parental responsibilities (Folsom et al., 2000; Lui, Farrell, MacNeil, Stone, & Barfield, 2008). Some of the main reasons for multiple follow-ups may be a lack of well-trained pediatric audiologists and physicians who provide follow-up testing or a lack of facilities that provide sedated diagnostic testing (Shulman et al., 2010). In fact, the sleeping or quiet state of the infant, particularly for those younger than three months old, is necessary in the early stages of testing and diagnosis to avoid the need for sedation (National Center for Hearing Assessment and Management, 2012). If the sleeping or quiet state is not attained sufficiently, untrained or inexperienced providers may recommend rescheduling another visit or referring to another facility that can conduct sedated hearing testing for diagnosis. Parents of babies with many follow-ups may become frustrated with the continual re-testing with no conclusion and lose confidence in the follow-up provider and the facility. This assumption may explain why some parents did not follow through with subsequent appointments and the audiologic diagnosis evaluation was not completed. Figure 2 summarizes possible reasons of late newborn hearing screening, late follow- ups, and multiple follow-ups as pathways leading to incomplete audiologic diagnosis.

Strengths and Limitations

This study had three major strengths: First, covariates including mother and child characteristics collected in birth certificates were captured through data linkages. These characteristics were controlled for in adjusted regression models to evaluate independent effects of predictors. Second, the study displayed an important role of VLBW, a strong confounder, in contributing to late NHS, late follow-up, and multiple follow-up appointments as well as IAD. Last, independent effects of the predictors were evaluated among different birth weight categories, which excluded a direct effect of VLBW on predictors in evaluating associations among LBW and normal weight newborns.

The study included two major limitations: First, underlying mechanisms to explain associations were limited, particularly among LBW and normal weight newborns; therefore, more studies are needed. Second, the incomplete audiologic diagnosis status was not verified through contacting parents or follow-up facilities. Verification may improve underreporting problems and avoid misclassification of the study outcome as well as bias of study results.

Conclusions

In order to reduce IAD, it may be necessary to conduct NHS early, expedite follow-up, and decrease the number of follow-up visits. Severe medical conditions, particularly VLBW, majorly contributed to late NHS that increased risk of IAD. Efforts to reduce severe medical conditions by enhancing the quality of prenatal and obstetrical care could help prevent both NICU admission and prolonged hospitalization, and thus reduce late NHS prior to hospital discharge (Gregory, Jackson, Korst, & Fridman, 2012; Lu, Kotelchuck, Hogan, Johnson, & Reyes, 2010; Newnham et al., 2014; Sakala, Yang, & Corry, 2013). To reduce the risk of IAD due to late NHS, screening should be conducted as early as possible during the NICU stay as medical conditions allow instead of waiting until hospital discharge.

Currently, based on the JCIH Position Statement 2007, all infants admitted to the NICU should be screened for hearing loss before hospital discharge. Although only 10-15% of the newborn population spends time in the NICU, this population has a higher risk of hearing loss and in particular, neural hearing loss (auditory neuropathy spectrum disorder; D'Agostino & Austin, 2004; Starr, Sininger, & Pratt, 2000). Therefore, not only screening but also the diagnostic process should be completed prior to discharge for newborns with severe medical conditions or those with prolonged hospitalizations, particularly in NICU, if at all possible. An increased number of sedated hearing diagnostic testing facilities and follow-up providers with significant pediatric experience, may reduce referral to other facilities and the number of follow-up appointments.

To understand mechanisms of the associations and reasons of late NHS, late follow-up, and multiple follow-ups, particularly among non-VLBW newborns, further in-depth quality improvement studies are needed. Through such studies, both parents and follow-up facilities should be contacted. Specifically, the studies might target the following: parents' knowledge and awareness of the importance of early diagnosis of hearing loss, providers and audiologists' experience or skill in screening young infants, referral scheduling relying on the parents instead of staff of referring facilities, and miscommunications and unclear referral protocols between referring and receiving facilities.

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