Assessment of Factors Involved in Non-Adherence to Infant Hearing Diagnostic Testing

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Abstract

Introduction: Delayed diagnosis of pediatric hearing loss can cause delays in cognitive and social development. This study described the sociodemographic factors associated with delayed timing of a final hearing diagnosis after an abnormal newborn hearing screening (NBHS). **Method**: Parent-infant dyads were recruited after being referred for further audiologic testing on an abnormal result from the NBHS. **Results**: Of the 53 participants, 55% ($n = 29$) did not receive a final diagnosis by the recommended 3 months of age. Of those with a delayed diagnosis, 45% ($n = 13$) had their first appointment within 3 months, but a delay was caused by an inconclusive or abnormal auditory brainstem response (ABR), middle ear pathology, or the presence of risk factors requiring additional testing. In a univariate analysis, older parental age ($OR = 0.90$, 95% CI: [0.82, 0.99]) and more total children in the household ([$OR = 0.66$, 95% CI: {0.18, 2.49}] for 1 child vs. 2 and [$OR = 0.14$, 95% CI: {0.03, 0.69}] for 1 children vs. 3 or more) were shown to significantly increase the odds of a delayed diagnosis, whereas younger infant age at first appointment (OR: 0.95, 95% CI: [0.92, 0.99]) was shown to significantly decrease the odds of a delayed diagnosis. In multivariate analyses, delayed diagnosis was also decreased by younger infant age at the initial appointment ($OR = 0.94$, 95% CI: [0.90, 0.99]). **Conclusion**: Parental age, number of total children in the household, and timing of first appointment may predict delayed diagnosis. Because many patients with a delayed diagnosis attended an appointment within 3 months, further standardization of the process and targeted interventions for families could improve chances of achieving a diagnosis within the first appointment.

Acronyms: ABR = auditory brainstem response; CDC = Centers for Disease Control and Prevention; EHDI = Early Hearing Detection and Intervention; JCIH = Joint Committee on Infant Hearing; NBHS = newborn hearing screening; OAE = otoacoustic emissions; OCSHCN = Office for Children with Special Health Care Needs

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Pediatric hearing loss is one of the most common neonatal sensory disorders in the United States with a prevalence of 1.7 per 1,000 babies screened in 2016 (Centers for Disease Control and Prevention [CDC], 2016). According to national Early Hearing Detection and Intervention (EHDI) data, the number of infants who are deaf or hard- of-hearing in the United States has increased significantly, which coincided with the adoption of the Newborn and Infant Hearing Screening and Intervention Act of 1999. There were 855 documented cases of deaf or hard-of-hearing infants in the United States in the year 2000. However, in 2014, 6,163 infants were identified as deaf or hard-of-hearing (CDC, 2016). This illustrates the impact of universal newborn hearing screening (NBHS) on early identification.

The Joint Committee on Infant Hearing (JCIH; 2007) recommends three timing milestones for screening, diagnosis, and treatment. This statement has been nationally recognized as the "1-3-6" rule: All newborns should be screened before leaving the hospital or before 1 month of age, diagnosed with normal or abnormal hearing before 3 months, and treated no later than 6 months of age. These recommendations intend to mitigate the

consequences of delayed diagnosis and intervention, which include significant delays in cognitive and social development, as well as language and long-term literacy impairment (Ching, 2015; Pimperton et al., 2016; Tomblin et al., 2015).

However, in 2016, only 47.6% of infants received a final diagnosis by 3 months of age after their NBHS reported an abnormal finding (CDC, 2016). This low level of compliance with the national recommendation highlights the importance of investigating factors associated with delays in diagnosis. To date, few studies have looked at this particular association. Location of residence may impact adherence to EHDI recommendations as rural children have significant delays in their diagnosis as compared with children from urban areas (Bush, Bianchi, et al., 2014; Bush, Osetinsky, et al., 2014). The findings in such studies may be due to parental factors specific to impoverished communities relating to insurance, socioeconomic data, and education status, as well as barriers to accessing specialist and primary care providers who feel comfortable addressing pediatric hearing healthcare (Bush, Alexander, Noblitt, Lester, & Shinn 2015; Bush, Kaufman, & McNulty, 2017). Furthermore, other research has investigated factors associated with delay in diagnosis and found that developmental or medical complications, such as middle ear disorders, can prolong the diagnostic workup of infant hearing (Fitzpatrick, Dos Santos, Grandpierre, & Whittingham, 2017).

The purpose of this study is to assess factors related to adherence to diagnostic testing after an abnormal NBHS using a prospectively-recruited cohort of parent-infant dyads. By understanding these factors, interventions can be developed to reduce delayed diagnosis of hearing loss in infants.

Materials and Method

Ethical Approval

This study was approved by the Institutional Review Board of the Office of Research Integrity of the University of Kentucky (number 12-1059-P1H).

Design

This longitudinal study recruited participant dyads consisting of the parent and infant. Participants were recruited after the infant failed their hospital NBHS test (either auditory brainstem response [ABR] test or otoacoustic emissions [OAE] test) if they were discharged from the nursery within 1 week after birth and did not require a stay in the neonatal intensive care unit. Participants were referred to the University of Kentucky or the Office for Children with Special Health Care Needs (OCSHCN) for outpatient audiological evaluation and diagnosis confirmation. The OCSHCN provides comprehensive care to children with special healthcare needs who are Kentucky residents and who meet medical and financial eligibility.

At the time of enrollment, informed consent was obtained, and one parent of the infant was asked to fill out an entrance questionnaire with their sociodemographic data, which included: age, gender, employment status, marital status, ethnicity, educational level, county of residence of the individual parent who responded to the survey, yearly household income, child's insurance, and total number of children in the household.

The standard-of-care given to the participants was according to the statewide Kentucky EHDI standards: parents of children who fail the NBHS were given educational materials regarding infant hearing loss and services offered by EHDI. Follow-up appointments for audiological diagnostic testing were arranged by the birth hospital and the referral audiology clinics. From this point, the parent was able to self-initiate contact with the audiology office to discuss any questions or concerns. Confirmation of scheduled appointments and appointment reminder calls were arranged by the audiology clinics.

Data Collection and Analysis

Follow-up appointments and diagnostic testing results were recorded for each participant from the electronic medical record at the University of Kentucky Medical Center, the OCSHCN, and the Kentucky EHDI database. Medical records until 6 months after birth were used to assess whether a child received an evaluation and final diagnosis during that period. All information was recorded using the online data collection tool REDCap.

Values for Beale code, or the Rural-Urban Continuum Coding system, which ranges from 1 (most urban) to 9 (most rural) were found for each county of residence of the participants using the 2013 Rural-Urban Continuum Codes (United States Department of Agriculture, 2013). These codes were reclassified such that codes 1 through 3 represented urban areas, codes 4 through 6 represented suburban areas, and codes 7 through 9 represented rural areas (Fiorillo et al., 2018). Ethnicity, employment status, marital status, and education of the individual parent who filled out the survey were all recoded into the categorical variables as found in Table 1.

The outcome for each participant was determined by the notes in the electronic medical record. Having a final diagnosis was defined by a definitive normal or abnormal diagnosis in the medical record without additional appointments or hearing evaluations scheduled.

Exploratory analyses are included by using descriptive statistics in Table 1. Results were examined by diagnosis group (diagnosis before 3-months or after 3-months of age), which is the primary outcome of interest in this study. Continuous variables were summarized by means (standard deviations) and compared between the two groups using independent t-tests. Categorical variables are summarized by frequencies (percentages) and compared between the two groups by Chi-square or Fisher's exact tests, as appropriate. To determine each variable's association with diagnosis by 3 months, univariate and multivariate logistic regression modeling was used. Due to sparse data, household income and number of visits to the clinic could not be included within this model. C-statistics, or estimated areas under the

Table 1

Descriptive Statistics for the Sample by Diagnosis Status

Variables	Category	Diagnosis by 3 Months (*n* = 24)	No Diagnosis by 3 Months (*n* = 29)	*p*-value
Parental Age (years)	Mean (*SD*)	26 (5)	29 (6)	**0.043***
	(Min, Max)	(18, 38)	(19, 41)	
Child's Age at First Appointment (days)	Mean (*SD*)	52 (19)	90 (56)	**0.003***
	(Min, Max)	(28, 90)	(35, 247)	
Employment Status	Employed	8 (33.3%)	16 (55.2%)	0.112
	Unemployed	16 (66.7%)	13 (44.8%)	
Marital Status	Single/Never Married	12 (50.0%)	12 (41.4%)	0.530
	Married/Partnership	12 (50.0%)	17 (58.6%)	
Ethnicity	White/Caucasian	13 (54.2%)	19 (65.5%)	0.400
	Other	11 (45.8%)	10 (34.5%)	
Education Level	High School or Less	12 (50.0%)	9 (31.0%)	0.160
	College or more	12 (50.0%)	20 (69.0%)	
Household Income	Less than $10,000	8 (34.8%)	5 (18.5%)	0.709
	$10,000–$20,000	6 (26.0%)	8 (29.6%)	
	$20,000–$30,000	3 (13.0%)	4 (14.8%)	
	$30,000–$60,000	2 (8.7%)	2 (7.41%)	
	More than $60,000	4 (17.4%)	8 (29.6%)	
Beale Code	Urban (1–3)	13 (54.2%)	23 (79.3%)	0.130
	Suburban (4–6)	7 (29.2%)	3 (10.3%)	
	Rural (7–9)	4 (16.7%)	3 (10.3%)	
Child's Insurance	Private	7 (29.2%)	11 (37.9%)	0.502
	Medicaid	17 (70.8%)	18 (62.1%)	
Number of Children	1	10 (41.7%)	6 (20.7%)	**0.032***
	2	11 (45.8%)	10 (34.5%)	
	3 or more	3 (12.5%)	13 (44.8%)	
Number of visits within 6 months	0	1 (4.2%)	9 (31%)	**0.002***
	1	23 (95.8%)	11 (37.9%)	
	2	0 (0.0%)	5 (17.2%)	
	3 or more	0 (0.0%)	4 (13.8%)	

Note. Means and standard deviations (*SD*) are given for continuous variables, whereas frequencies and percentages are given for categorical variables. *n* = sample size, *SD* = standard deviation, min = minimum value, max = maximum value.

*significant at *p* < 0.05

receiver operating characteristic curve, were presented for each model as a predictive accuracy measure. In general, values of 0.5 or lower indicate a poor model, values over 0.7 indicate a good model and values over 0.8 indicate a strong model. Statistical significance was defined as $p \leq 0.05$. All data were analyzed using SAS version 9.4.

Results

Of the 53 parent-infant dyads who participated in this study, 55% ($n = 29$) did not obtain a hearing diagnosis by the recommended 3 months. In this sample, 64% ($n = 34$) of the dyads ended up being diagnosed with normal hearing and 8% ($n = 4$) with abnormal hearing (Figure 1). Of note, all the participants who were diagnosed with abnormal hearing received the diagnosis after the 3-month recommendation.
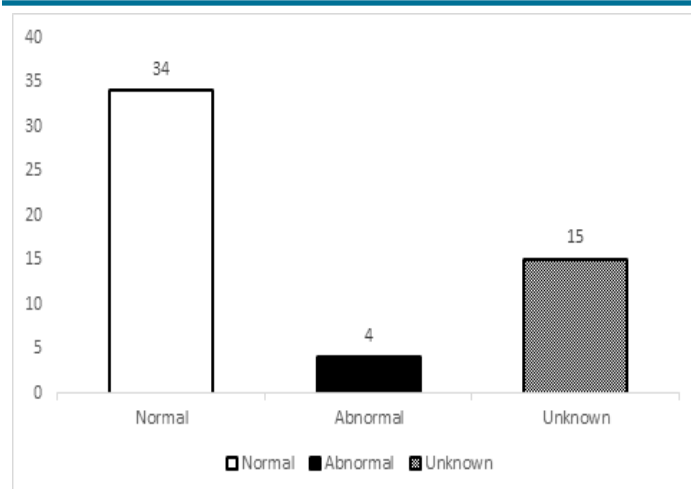

Figure 1. Final diagnosis classification.

Further, 72% ($n = 38$) of dyads attended at least one scheduled audiology appointment for diagnostic testing during the recommended 3-month period. Out of the 29 dyads whose final diagnosis was delayed beyond 3 months, 69% ($n = 20$) attended the audiology clinic at least once and 45% ($n = 13$) had their first appointment within 3 months. However, these individuals required a follow-up appointment because of inconclusive ABR testing (31% [$n = 4$]), middle ear pathology (31% [$n = 4$]), an abnormal ABR result needing additional confirmatory ABR (31% [$n = 4$]), or the presence of risk factors requiring additional ABR testing (7% [$n = 1$]).

Figure 2 displays the age of the infant at the time of final diagnosis. Of the 29 dyads with a delayed diagnosis, 48% ($n = 14$) did receive a diagnosis after 3 months; of the other 52% ($n = 15$), 9 individuals never appeared for an appointment during the 6-month study period and 6 were not diagnosed either due to loss to follow-up or inconclusive results.

The sociodemographic data for participants is presented in Table 1. Parents of infants who received a delayed diagnosis were significantly more likely to be older ($p = 0.043$) and to have more total children in the household ($p = 0.032$) than those who were diagnosed before 3 months (Table 1). Infants who received a delayed diagnosis were
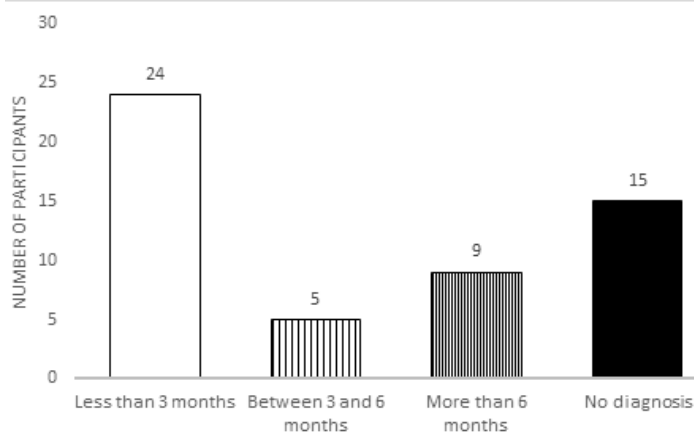

Figure 2. Age of infant at time of final diagnosis.

significantly older at the time of their first appointment ($p = 0.003$) than infants who were diagnosed before 3 months, with a mean difference of approximately 38 days.

Univariate logistic regression analyses are presented in Table 2. The odds of timely diagnosis increased with younger age at the first audiology appointment of both the parent ($OR = 0.90$, 95% CI: [0.82, 0.99]) and infant ($OR = 0.95$, 95% CI: [0.92, 0.99]), as well as with lower total number of children in the household ([$OR = 0.66$, 95% CI: {0.18, 2.49}] for 1 child vs. 2 and [$OR = 0.14$, 95% CI: {0.03, 0.69}] for 1 children vs. 3 or more). The largest predictive accuracy of diagnosis by 3 months was seen with the infant's age at first appointment (C-statistic = 0.786), followed by the total number of children in the household (C-statistic = 0.686) and parental age (C-statistic = 0.652). These values are indicative of a good model.

In multivariate analyses (Table 3), age of infant at the time of the initial appointment was the only variable that reached significance ($p = 0.016$). After controlling for all other variables, the odds in favor of receiving a final diagnosis by 3 months increased by 6% ($OR = 0.94$, 95% CI: [0.90, 0.99]) with younger infant age at the initial appointment. The predictive accuracy of diagnosis by 3 months for this model was 0.915, which indicates that the model has a good fit.

Discussion

Pediatric hearing loss requires a time-sensitive diagnosis in order to promote oral language development of the child and to improve social and academic outcomes (Armstrong et al., 2013; Bush, Osetinsky, et al., 2014). It was demonstrated in our study that the 1-3-6 rule established by the JCIH was not achieved in more than half of the patients referred on an abnormal NBHS. Our study demonstrates concerning rates of nonadherence that are consistent with the national rates (CDC, 2016).

Previous studies have looked at the impact of rural versus urban residence regarding timing of hearing diagnostic care and definitive treatment (Bush, Bianchi, et al., 2014; Bush, Osetinsky, et al., 2014). However, there is limited literature that describes the parental sociodemographic

Table 2

Univariate Logistic Regression Analysis Using Outcome of Interest "Diagnosis by 3 Months" with Unadjusted Bivariate Associations

Variables	OR	Lower CI (95%)	Upper CI (95%)	p-value	C statistic
Parental Age (years)	0.90	0.82	0.99	**0.049***	0.652
Infant's Age at First Appointment (days)	0.95	0.92	0.99	**0.010***	0.786
Employment Status	0.41	0.13	1.25	0.115	0.609
Marital Status	0.71	0.24	2.10	0.531	0.543
Ethnicity	1.61	0.53	4.88	0.402	0.557
Education Level	0.45	0.15	1.38	0.163	0.595
Household Income				0.720	0.612
Less than $10,000	Reference				
$10,000–$20,000	0.47	0.10	2.19	0.808	
$20,000–$30,000	0.47	0.07	3.04	0.849	
$30,000–$60,000	0.63	0.07	5.97	0.849	
More than $60,000	0.31	0.06	1.61	0.352	
Beale Code				0.146	0.632
Urban	Reference				
Suburban vs Urban	4.13	0.91	18.75	0.221	
Rural vs Urban	2.36	0.46	12.21	0.861	
Child's Insurance	1.48	0.47	4.72	0.503	0.544
Number of Children				**0.046***	0.686
1	Reference				
2	0.66	0.18	2.49	0.340	
3	0.14	0.03	0.69	**0.015***	

Note. OR = odds ratio, CI = confidence interval, C-statistic = estimated areas under the receiver operating characteristic curve.

*significant at p < 0.05

factors with regards to delayed diagnosis after an abnormal newborn hearing screening. In this study, potential influencing sociodemographic factors specific to the challenges within the appointments to assess hearing loss were identified, including parental age and number of children in the household. With regards to number of children and parental age, it is possible that older parents may have more children, which may confound the observed effect. It is also important to note that the age ranges of the groups compared were close together (Table 1), which may further limit the clinical significance of this finding.

Younger infant age at the initial appointment following an abnormal NBHS was found to be significant in both univariate and multivariate analysis. This seems to indicate that it may be easier to schedule additional follow-up within the recommended 3-month timeframe if the initial appointment were scheduled earlier in the infant's life. This may relate to other trends identified in the data: many patients who received a delayed diagnosis came to at least one appointment within 3 months of age and there was a statistically significant increase in delayed diagnosis when more appointments were needed to make the diagnosis.

Furthermore, needing more appointments prior to final diagnosis was found to be related to factors inherent to the current diagnostic process such as inconclusive ABR results due to middle ear pathology or sleeplessness of the infant, abnormal ABR results needing additional confirmatory ABR, and additional ABR testing required due the presence of risk factors. Fitzpatrick et al. (2017) similarly describes the difficulty and complexity involved in determining a final diagnosis due to many factors, including the nature of the diagnostic test itself.

Table 3
Adjusted Multivariate Logistic Regression Analysis Using Outcome of Interest "Diagnosis by 3 Months"

Variables in the mode	OR	Lower CI (95%)	Upper CI (95%)	p-value
(95%)	**(95%)**	0.65	1.13	**(95%)**
Infant's Age at First Appointment (days)	0.94	0.90	0.99	**0.016***
Employment Status	0.15	0.01	1.79	0.133
Marital Status	0.55	0.07	4.68	0.585
Ethnicity	2.19	0.14	34.81	0.578
Education Level	0.15	0.02	1.55	0.112
Beale Code				0.198
Suburban vs Urban	4.92	0.25	96.74	0.750
Rural vs Urban	9.56	0.19	486.37	0.428
Child's Insurance	0.06	0.01	2.80	0.149
Number of Children				0.085
2 vs 1	0.27	0.02	3.88	0.991
3 vs 1	0.08	0.002	2.83	0.192
2 vs 3	0.99	0.26	3.71	0.991

Note. OR = odds ratio, CI = confidence interval, C-statistic of the multivariate logistic regression model = 0.915.

*significant at $p < 0.05$

Diagnostic testing results can be influenced by external and middle ear pathology, such as stenosis, debris, or effusion. Therefore, if a child has evidence of abnormal hearing function on definitive audiological testing, many institutions require a follow-up confirmatory audiological test to confirm the presence and severity of the hearing loss (Rowe, Gan, Benton, & Daniel, 2016). Because existence of middle ear pathology is not predictable, scheduling the initial appointment earlier may be an effective strategy to allow adequate time for a second one to be scheduled in case a definitive diagnosis cannot be obtained during the first appointment. Alternatively, clinics can consider giving priority to follow-up appointments after failed ABRs to ensure that the infant can still be diagnosed within the recommended 3-month timeframe. This strategy helps mitigate unpredictable reasons for multiple appointments such as middle ear pathology.

Our review of the electronic medical record on these patients revealed that one of the most common reasons for requiring a second appointment was that the infant was unable to sleep during the ABR test. Sleep-deprived ABR testing involves obtaining hearing testing results during sleep; this testing technique is rendered inconclusive or must be discontinued if the infant will not sleep. Parents were then counseled on proper preparation for a follow-up ABR, but, even then, some of the patients returned for a second appointment in a wakeful state and the testing could not be completed. Sedated ABR testing can be performed, but it requires the coordination of multiple disciplines to anesthetize the infant and may be a risky procedure for some infants. Although some practices only perform infant ABR under sedation, many practices perform sleep-deprived ABR as a first line approach with sedated ABR being a secondary option. These practices may consider the role of earlier sedated ABR testing to proactively prevent the possibility that the infant still will not sleep during the follow-up appointment. A sedated ABR is just as effective at comprehensively evaluating the hearing of an infant (Levit, Mandel, & Matot, 2018; Mühler, Rahne, Mentzel, & Verhey, 2014). Several of our participants, as we have demonstrated, required multiple appointments or, ultimately, sedation to obtain a complete and accurate ABR. However, sedated ABR is costlier than sleep-deprived options, has medical risks inherent to sedation and general anesthesia, and may present with scheduling problems because multiple departments, such as anesthesia, are involved. Moreover, some children with other medical problems may not be eligible for sedation

due to their comorbidities and these children may also be those who are most vulnerable to receiving a delayed diagnosis (Fitzpatrick et al., 2017).

Another strategy to mitigate delayed diagnosis due to infant wakefulness may include improved parental pre-appointment counseling. It is integral for parents to understand their role in the diagnostic process, especially since a second appointment may not be able to be scheduled within the recommended diagnostic timeframe.

A patient navigator model, in which a layperson or a healthcare professional advocates for early follow-up and adherence to appointment preparation, has been studied to address the problem of compliance following referral on the NBHS (Bush et al., 2017). As mentioned previously, Fitzpatrick et al. (2017) found that infants with developmental or medical problems were at higher risk to have delayed diagnosis of hearing loss. These families may require more personalized attention when navigating the healthcare system because of the complexity of their child's medical needs. Interventional strategies that promote personalized pre-appointment counseling such as a patient navigator may reduce delayed diagnosis by guiding families with complex care needs and by effectively counseling parents prior to the ABR testing to reduce the number of appointments needed from wakefulness.

Another area for intervention to address access barriers is in service-system capacity. There is some evidence pointing toward shortages in screening equipment and pediatric audiologists, lack of provider knowledge, lack of standardized protocols for screening and presenting screening results, and challenges to families in obtaining services, such as transportation issues, as well as information and communication gaps (Shulman et al., 2010). Many primary care providers lack confidence in counseling and leading a family through the EHDI process (Bush et al., 2015). Targeted interventions that tackle these specific gaps in the system could improve timely diagnosis and, therefore, the language development outcomes of the child.

Limitations

This study has a small sample size, which adds difficulty to computing adjusted associations and identifying potentially confounding variables such as increased parental age and increased number of total children in the household. Moreover, the small sample size may not allow for a clear picture of the importance of variables that may otherwise have been significant.

Conclusion

This study assesses parental sociodemographic factors involved in delayed diagnosis after referral on the NBHS. Universal newborn hearing screening, which began in the United States almost twenty years ago, has been improving, which is evidenced by the larger number of infants being screened. However, since more than half of all children screened do not receive a diagnosis within the recommended timeframe, there is still much work needed to ensure that children who have hearing deficits receive adequate and timely services to ensure normal social development, academic performance, and speech intelligibility. It was identified in this study that certain parental factors may play a role in delayed diagnosis, which may be able to be reduced with a patient navigator model or improved pre-appointment counseling with regards to sleeping during the ABR testing. The problem of infant wakefulness during the ABR may also be mitigated with earlier timing of sedated ABR, but that is costlier and puts the patient at increased risk of adverse effects from the anesthesia. It was also identified that factors intrinsic to the diagnostic process may impact the risk an infant has of receiving a delayed diagnosis of hearing loss, such as total number of appointments needed to achieve a diagnosis and infant's age at the initial appointment. Therefore, earlier scheduling of initial and follow-up audiology appointments may decrease incidence of delayed diagnosis. Research should continue to address this topic to move toward a stronger model that identifies both individual and systematic factors that contribute to delay in diagnosis.

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