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# Timeliness of EHDI Benchmarks in Infants with a NICU Admission Greater than Five Days: Analysis from a Retrospective Cohort

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**Abstract**

The purpose of this study was to examine the timeline of early hearing healthcare in infants with a history of lengthy (> 5 days) admission to a neonatal intensive care unit (NICU) compared to non-NICU peers. We compiled four years of state Early Hearing Detection and Intervention (EHDI) records from 156,335 infants using a statewide administrative database. We compared age at the time of newborn hearing screening, diagnostic audiological evaluation, and entry into early intervention in NICU infants and non-NICU infants. We also compared the proportion of NICU and non-NICU infants meeting prescriptive EHDI timing benchmarks based on the Joint Committee on Infant Hearing (2019) position statement. Results indicated that NICU infants experienced delayed newborn hearing screening and diagnostic evaluation compared to non-NICU peers and reached both benchmarks in lower proportions. NICU and non-NICU infants entered early intervention at equivalent ages and met the early intervention benchmark in similar proportions. Considering the important medical factors that drive lengthy NICU admissions, our results suggest that specific clinical guidelines for the timing of early hearing healthcare in NICU infants may be warranted.

**Acronyms:** ECMO = extracorporeal membrane oxygenation; EHDI = early hearing development and intervention; EI = early intervention; IDPH = Iowa Department of Public Health; JCIH = Joint Committee for Infant Hearing; LTFU/D = loss to follow up/documentation; NICU = neonatal intensive care unit; PCHL = permanent childhood hearing loss

**Keywords:** EHDI, pediatric audiology, screening, NICU

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The Joint Committee on Infant Hearing (JCIH) position statement is a broad clinical practice guideline for providers and policy-makers about the screening, diagnosis, medical management, intervention, and surveillance of infants with hearing loss (or infants at risk for developing hearing loss; JCIH, 2019). In the United States, individual state early hearing detection and intervention (EHDI) programs integrate JCIH recommendations at the state level. A key feature of EHDI program quality has been the establishment of a timeline for three primary benchmarks: hearing loss screen no later than one month of age, diagnosis no later than three months, and enrollment in early intervention no later than six months. In the most recent JCIH position statement published in 2019, the committee advocated for states that regularly meet the 1-3-6 timeline to now pursue a 1-2-3 timeline. In both cases, meeting timing recommendations may be more challenging for families when infants have additional medical needs in the newborn period and spend time in the neonatal intensive care unit (NICU).

## NICU Trends and EHDI Status

Infants spend time in a NICU after delivery for a variety of reasons (e.g., low birth weight, preterm delivery). The rate of admission to the NICU increased 23% from 2007 to 2012 (Harrison & Goodman, 2015) and although long term survival for preterm infants has improved in the past 20 years, the likelihood of additional disabilities is high for preterm, low, and very-low birth weight infants (Chan et al., 2001; Kilbride et al., 2004; Stoll et al., 2015). Program planning for newborn hearing screening must account for an increasing burden of infants with a history of NICU stay.

Across NICU and non-NICU birth settings, hearing loss is the most common medical condition that is currently identified via newborn screening, at 16 infants per 10,000 live births (Williams et al., 2015). For comparison, recent incidence estimates for other serious congenital conditions in the United States were 14.85 cases of Trisomy 21 and 10.25 cases of cleft lip (with and without cleft palate) per 10,000 births (Mai et al., 2019). Other factors suggest that the congenital hearing loss rate of 16/10,000 may

underestimate the true number of infants who are born with developmentally significant hearing loss. Many newborn hearing screening programs experience high rates of loss to follow up or documentation (LTFU/D; or cases where the outcome of a failed screening cannot be confirmed). Across studies, the rates of LTFU/D for diagnostic audiological evaluation after the newborn hearing screening ranged from 9% to 41% (see review in Ravi et al., 2016). This rate does not account for infants who are born with slight and mild hearing loss and may not be detected with current screening approaches.

In NICU infants, incidence rates of hearing loss are higher than in non-NICU infants (Hille et al., 2007; Veen et al., 1993; White et al., 1994). In a 2007 study of early hearing outcomes in Dutch infants, 2.2% of study participants born at less than 32 weeks' gestation exhibited permanent childhood hearing loss (PCHL; van Dommelen et al., 2015). For comparison, similar population-level infant research on PCHL has revealed an overall rate of 0.16% (Williams et al., 2015). Younger gestational ages were associated with higher rates of hearing loss. Among the very earliest preterm births (24–25 weeks' gestation) the observed rate of hearing loss was 7.5% (van Dommelen et al., 2015). Xoinis et al. (2007) reported on both sensorineural hearing loss and auditory neuropathy spectrum disorder in NICU infants and found incidence rates of 2.2% and 0.56%, respectively.

There are many reasons for clinicians and researchers to have special concern regarding the early hearing healthcare of NICU infants. First, their risk of hearing loss is more acute. A NICU stay of greater than five days has been identified as a risk factor for late-onset hearing loss and is sufficient motivation for a follow-up hearing evaluation no later than 9 months of age (JCIH, 2019). Low APGAR scores are associated with both the need for NICU admission (Chu, 2003; Weinberger et al., 2000) and increased risk of infant hearing loss (Hille et al., 2007; Vohr et al., 2000). Infants in NICUs routinely require medical interventions that are associated with increased risk of permanent hearing loss including broad-spectrum IV antibiotics, mechanical ventilation, and extracorporeal membrane oxygenation (ECMO; Coenraad et al., 2010). Second, NICU parents may balance competing health priorities during the neonatal period. Using qualitative research methods with parents of infants with auditory neuropathy spectrum disorder, researchers found that hearing status was a low priority at the point of diagnosis amidst more urgent medical needs in the newborn period (Uus, 2012). Third, many NICU infants who pass the newborn hearing screening before discharge have risk factors that put them at significant risk for developing hearing loss (Dumanch et al., 2017).

Older age at newborn hearing screening has been associated with late follow up and incomplete audiological diagnosis among low birth weight and normal weight infants (Tran et al., 2016). Measuring EHDI follow up in NICU infants is challenging due to their heterogeneous health and developmental outcomes, and there are mixed findings about the impact of NICU status on audiological follow-up. Awad and colleagues (2019) reported ages at diagnosis and hearing aid fitting for ten NICU infants in their analysis of adherence to JCIH benchmarks among infants with bilateral hearing loss in a large metropolitan children's hospital. Of the nine surviving infants, four were diagnosed and fit with hearing aids beyond the 1-3-6 timeline in unadjusted age. However, among their collapsed study cohort of children with PCHL, NICU stay was not associated with an increased risk of delays between diagnosis and hearing aid fitting or age at diagnostic assessment. They did not report the timing of JCIH benchmarks for NICU infants who were ultimately diagnosed as normal hearing after not having passed the newborn hearing screening. In Crouch et al. (2017), investigators found that although low birth weight infants with hearing loss were less likely to access early diagnostic services, they were more likely to be enrolled in early intervention. They did not report the NICU status of their sample, however, we expect that many were NICU graduates based on their low birth weight.

In other studies, NICU status was associated with greater challenges meeting the recommended EHDI timeline. High intensity of neonatal care needs has been associated with lower rates of follow up for diagnostic testing at 3 and 6 months of age (Deem et al., 2012). In that analysis of quality metrics in the Buffalo, New York area newborn hearing screening programs, the highest observed rates of LTFU/D occurred in the region's only level IIIB (more acute) nursery. Others have found that a NICU stay does not contribute to increased risk of LTFU/D among infants who do not pass the initial screening (Spivak et al., 2009). Lieu and colleagues (2006) showed that although follow up in NICU infants has improved over time, it falls behind the recommended EHDI timeline. That investigation followed NICU infants who did not pass the newborn hearing screening between 1999–2002. Researchers followed families for up to four years after a failed newborn hearing screening, but they did not report the timing of follow up services. The authors classified children as having received follow up if parents reported that a hearing evaluation took place at any point in the intervening years, and did not report the timing of follow up.

The challenges that a long-term NICU stay poses for accessing early hearing services on time (diagnosis, fitting of appropriate technology, and enrollment in EI) have not been well characterized in a population-level group of infants. Given the increased risk for hearing loss in this group and the barriers that NICU infants may face, an important first step is to identify practice patterns related to the timing of their early hearing care. Significant public health resources are allocated to EHDI tracking and data management systems and these systems have been identified as the strongest tool to improve rates of follow up (Ravi et al., 2016). The administrative dataset that EHDI tracking programs generate provides a valuable opportunity to assess program quality and ascertain if states are meeting the recommendations laid out in the JCIH (2019) position statement. In the present study, we

use state-level EHDI program data to examine hearing healthcare trajectories in NICU and non-NICU infants.

### Research Questions

This study utilizes a large public health dataset to analyze the timeliness of EHDI benchmarks for infants in the state of Iowa between 2014–2017. It is motivated by the need to establish the baseline characteristics of service delivery to NICU infants in light of expected challenges to meeting benchmarks (e.g., later ages at discharge driving later ages at diagnosis and early intervention, competing health priorities). Infants who had lengthy admissions to a NICU (> 5 days) are compared with non-NICU peers. We designed our research question to make a comparison in terms of their absolute ages at each of three hearing benchmarks and with reference to exogeneous timing benchmarks prescribed by state and national EHDI programs. Our research addresses the following questions:

1. How does the timing of EHDI benchmarks in infants with lengthy NICU stays compare to the timing of EHDI benchmarks in non-NICU infants? *We hypothesize that NICU infants will achieve EHDI benchmarks at later ages than non-NICU peers.*
2. Do lower proportions of NICU infants meet EHDI timing benchmarks compared to non-NICU infants? *We hypothesize that a lower proportion of NICU infants will meet EHDI benchmarks by the recommended ages compared to non-NICU infants.*

## Method

### Iowa Department of Public Health EHDI Data

To complete this retrospective cohort study on EHDI timing benchmarks in NICU and non-NICU infants, we accessed newborn hearing screening and follow-up records from the state of Iowa gathered between 2014–2017. The Iowa EHDI program tracks screening and follow up using *e-Screener Plus*™ (eSP™) software developed by OZ Systems. As of August 2020, although Iowa has begun educating providers about the 1-2-3 EHDI timeline, its goal remains meeting the 1-3-6 timeline. All EHDI records were extracted from eSP by the Iowa Department of Public Health (IDPH) at the end of March 2019, de-identified, and shared via a secure data transfer. Table 1 lists the variables we extracted from individual records. Iowa's EHDI Coordinator shared the dates of enrollment in early intervention for a sub-set of infants with confirmed hearing loss and linked them with the eSP dataset prior to data transfer. This study was approved by the University of Iowa Institutional Review Board under a data-sharing agreement with IDPH. It was determined that this study did not meet the criteria to be considered human subjects research.

**Table 1**

*List of Extracted Variables from the Oz Database for Infants Included in this Study*

| |
|---|
| Date of Birth |
| Gender |
| Race/Ethnicity |
| City |
| State |
| Zip Code |
| Nursery (well-baby, NICU) |
| Place of Birth (Hospital/Home/Other) |
| Birthing Facility |
| Birth Screen Provider |
| Outpatient Screen Provider |
| Assessment Provider |
| Patient Outcome (e.g., deceased, moved out of state, complete in process) |
| Hearing Outcome (e.g., bilateral hearing loss complete, unilateral hearing loss-in process, normal hearing) |
| Birth Screen Date |
| Birth Screen Outcome (e.g., Bilateral Pass, Unilateral Pass) |
| Outpatient Screen Outcome (e.g., Bilateral Pass, Unilateral Pass) |
| Audiological Assessment Outcome (e.g., bilateral hearing loss complete, unilateral hearing loss-in process, normal hearing) |
| First Test Type |
| First Diagnostic Session Date |
| Right and Left Ear Outcomes (e.g., sensorineural, mixed, auditory neuropathy, normal) |
| Date of HL Confirmation |
| Date of Early Intervention referral |
| Risk Factors (e.g., Cranio-facial anomalies, transfusion for elevated bilirubin, assisted ventilation) |
| Family history of childhood hearing loss |
| NICU > 5 days |
| Assisted Ventilation |
| Bacterial or Viral Meningitis |
| Congenital CMV confirmed in baby |

***Note.*** CMV = cytomegalovirus; HL = hearing loss; NICU = neonatal intensive care unit.

### NICU and Non-NICU groups

The initial dataset included records for 156,335 infants. We classified infant records according to their NICU status: Infants with a NICU stay greater than five days (NICU group, *n* = 8,149) and infants without lengthy NICU stays (non-NICU group, *n* = 143,888). Thus, the non-NICU group includes infants with very short NICU admissions in addition to infants with no NICU stay. Given the focus of this investigation on timing aspects, we did not expect shorter stays than 5 days to impact a family's ability to meet EHDI 1-3-6 goals. Iowa tracks infants with a NICU stay of greater than five days to monitor for delayed-onset hearing loss as recommended by JCIH (2019), and newborn hearing screeners check a separate box to indicate that an infant met this criteria. Therefore the five day cutoff was a reliable method for separating our groups. We approached incomplete records (for example, infants whose nursery was reported as the NICU but for whom the hospital screener did not include risk factors) in two ways. If risk factor information was missing, but newborn nursery location was reported as "Well-baby," infants were classified in the non-NICU group. If records were so incomplete that no determination could be made with relation to nursery status, we excluded those infants from further analysis. Table 2 provides demographic characteristics of both groups and sample sizes available during analysis for each of the EHDI benchmarks. Figure 1 illustrates how the data were reduced. If infants were classified as deceased, we did not include their records in any analyses.

**Table 2**

*Summary Statistics and Demographic Characteristics in Infants with Lengthy Stays in the Neonatal Intensive Care Unit (NICU) and Infants without (Non-NICU)*

| | Non-NICU | NICU | Between Group |
|---|---|---|---|
| **Age at screen (days)**<br>Median<br>Mean (*SD*)<br>Range<br>(*n*) | <br>1<br>1.75 (3.58)<br>0–249<br>(*n* = 143,888) | <br>11<br>22.01 (28.48)<br>0–353<br>(*n* = 8,149) | *p*-value < 0.001* |
| **Age at diagnostic test (days)**<br>Median<br>Mean (*SD*)<br>Range<br>(*n*) | <br>42<br>73 (85.89)<br>0–673<br>(*n* = 1,167) | <br>89<br>107.7 (79.59)<br>8–537<br>(*n* = 227) | *p*-value < 0.001* |
| **Age at EI enrollment (days)**<br>Median<br>Mean (*SD*)<br>Range<br>(*n*) | <br>118<br>173.5 (149.97)<br>35–749<br>(*n* = 111) | <br>155.5<br>188.58 (150.6)<br>54–629<br>(*n* = 38) | *p*-value = 0.6 |
| **Maternal race: White (*n*; %)** | 121,752; 84.6% | 6,606: 81.1% | *p*-value < 0.001* |
| **Maternal race: Black (*n*; %)** | 10,196; 7.1% | 790; 9.7% | *p*-value < 0.001* |
| **Race: Other/Multi-race (*n*; %)** | 11,940; 8.3% | 753; 9.2% | *p*-value < 0.001* |
| **Lost Contact (*n*; rate)** | 457; 0.3% | | n/a |
| **Moved out of state (*n*; rate)** | 203; 0.1% | | n/a |

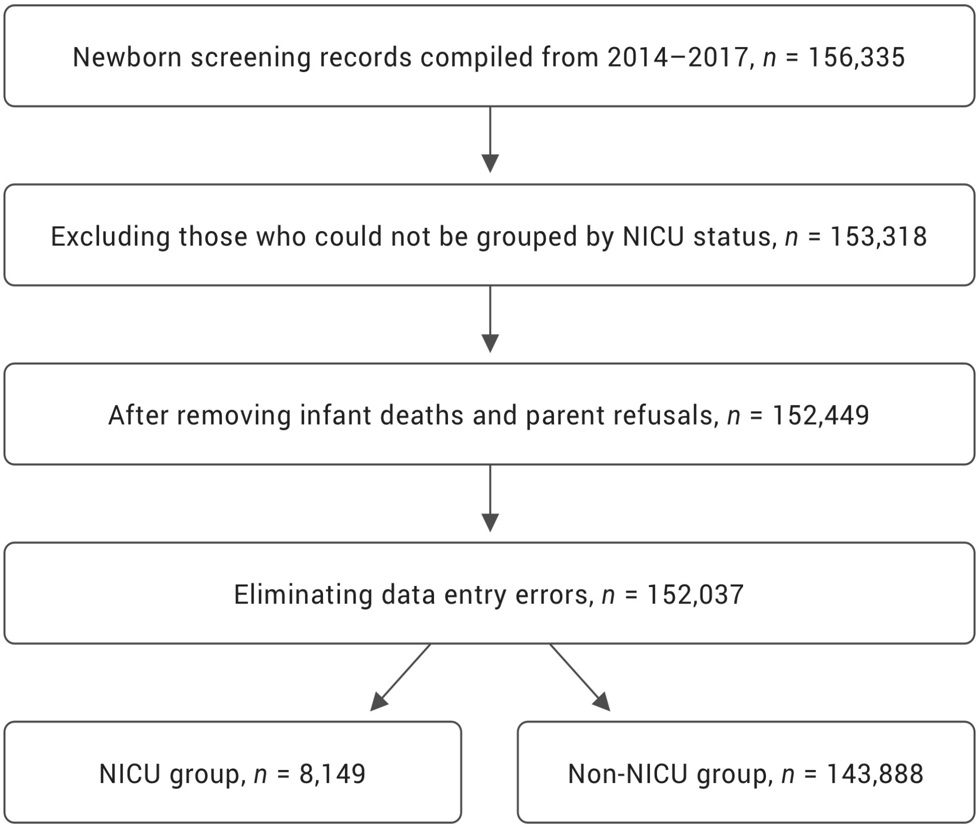
***Note.*** EI = early intervention

*indicates significance with alpha level = .05 level

To contribute to the larger body of research on hearing outcomes in NICU infants and characterize the representativeness of our dataset, we calculated group-specific incidence rates of hearing loss based on the full set of non-redacted data (with any length of NICU stay included in the NICU group, for incidence calculations only). We classified hearing losses as congenital if they were confirmed as a result of not having passed the newborn hearing screening. Across the four years examined here, the total incidence rate was found to be 1.91/1,000 births. Stratified by NICU status, the NICU-specific incidence rate was 5.27/1,000 births and the well-baby-specific incidence rate was 1.64/1,000 births.

**Figure 1**

*Data Filtering for Each Benchmark from Full 2014–2017 Dataset*




### Data Analysis

For statistical analysis, we narrowed the four years of data by stage in the EHDI process. We included all infants for the screening benchmark analysis, only infants who did not pass the screening for the diagnostic benchmark analysis, and only infants with confirmed hearing loss for the early intervention benchmark analysis. We performed all data manipulation and analyses in R 2.14.0, using the *epitools*, *dplyr*, *lubridate*, and *ggplot2* packages for analysis and data visualization (Aragon, 2020; Grolemund & Wickham, 2011; Wickham, 2016; Wickham et al., 2020). We generated new variables to represent an infant's age (in days) at each of the primary EHDI benchmarks by comparing appointment dates with the dates of birth. Finally, we created dichotomous variables to classify study participants as having *met* or *not met* timing recommendations. For all analyses, a month was treated as 30 days, three months as 90 days, and six months as 180 days to remain consistent throughout the four years of data. For this study, the early intervention benchmark represented enrollment into IDEA Part C Early Intervention programs, not the date of hearing aid fitting.

For each of the three benchmarks, we first compared the un-adjusted ages at EHDI benchmarks using a Welch's adjusted *t*-test due to unequal variances between groups. We then performed a chi-squared test to assess proportions of each group that met specific EHDI timing benchmarks. Odds ratios and 95% confidence intervals were calculated to characterize the relationship between the exposure of interest (lengthy NICU admission) and the outcome of interest (successfully completing EHDI benchmarks on time).

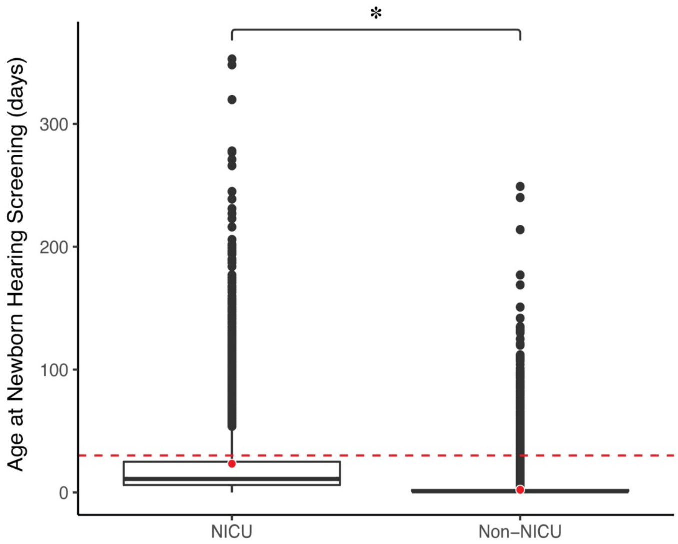
## Results

### Timing of EHDI Benchmarks in NICU Infants

Table 2 contains descriptive statistics for both groups, including the means, medians, standard deviations, and ranges of ages at each benchmark. Figures 2–4 show ages and distributions for both groups at each EHDI benchmark. On average, NICU infants received the initial screening at 22 days of life (compared to 1 day of life in non-NICU infants), had their first diagnostic assessment at 110 days (compared to 75 days), and enrolled in early intervention at 189 days (compared to 174 days). Although all infants in the NICU group were confirmed to have spent five or more days admitted, our data revealed that some infants in the NICU group received the newborn hearing screening on the first day of life. This could reflect late admission or re-admission to the NICU. We observed wide ranges for all three benchmarks across the full sample. NICU infants were significantly older at the time of hearing screening ($p$-value $< 0.001$) and diagnostic evaluation ($p$-value $< 0.001$) than non-NICU peers, but both groups enrolled in early intervention at equivalent ages. Fewer records were available for the early intervention benchmark due to both the lower numbers of confirmed hearing loss that required early intervention referral and incomplete records of referral for some cases of PCHL.

**Figure 2**

*Age at EHDI Benchmark for Newborn Hearing Screening in Infants with Lengthy NICU Stay and Non-NICU Infants*

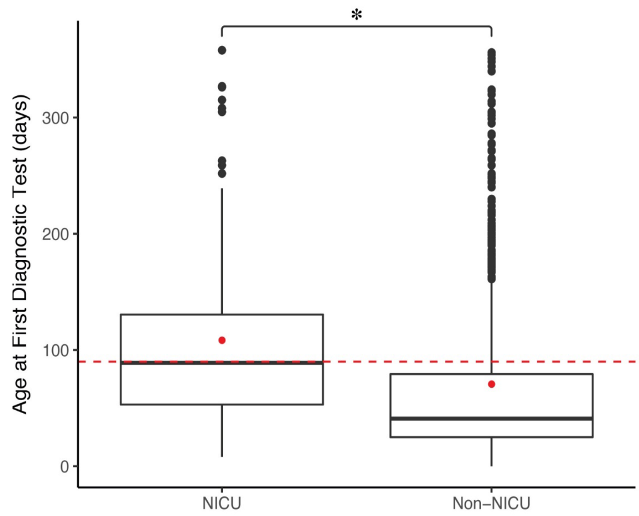



*Note*. Boxes show lower (Q1) and upper (Q3) quartiles and the median. Whiskers show data points within 1.5 times the interquartile range, and black circles show outliers. Means are plotted in red. For comparison, red hashed lines show the age recommendation in the Joint Committee on Infant Hearing (2019) position statement. EHDI = early hearing detection and intervention.

*indicates significance with alpha level = .05 level

**Figure 3**

*Age at EHDI Benchmark of Diagnostic Evaluation for Infants with Lengthy NICU Stay and Non-NICU Infants*

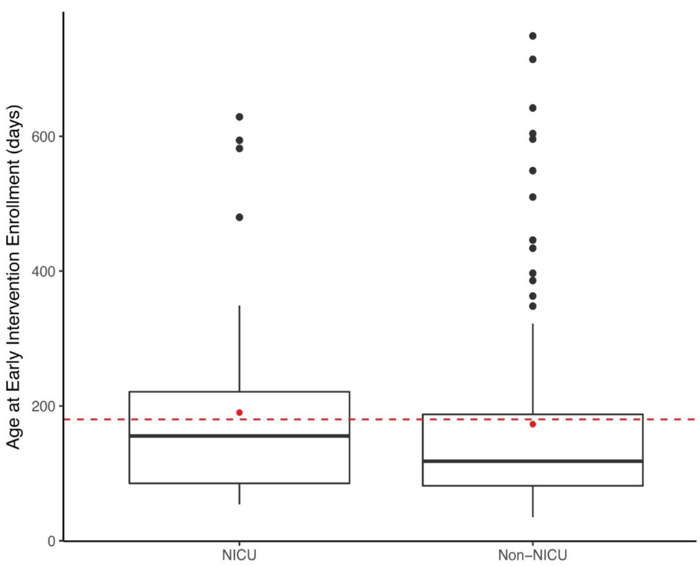



*Note*. Boxes show lower (Q1) and upper (Q3) quartiles and the median. Whiskers show data points within 1.5 times the interquartile range, and black circles show outliers. Means are plotted in red. For comparison, red hashed lines show the age recommendation in the Joint Committee on Infant Hearing (2019) position statement. EHDI = early hearing detection and intervention.

*indicates significance with alpha level = .05 level

**Figure 4**

*Age at EHDI Benchmark of Enrollment in Early Intervention for Infants with Lengthy NICU Stay and Non-NICU Infants*




*Note*. Boxes show lower (Q1) and upper (Q3) quartiles and the median. Whiskers show data points within 1.5 times the interquartile range, and black circles show outliers. Means are plotted in red. For comparison, red hashed lines show the age recommendation in the Joint Committee on Infant Hearing (2019) position statement. EHDI = early hearing detection and intervention.

**Table 3**

*Odds Ratios and Confidence Intervals Associated with Missing EHDI Timing Benchmarks in Infants with Lengthy Stays in the Neonatal Intensive Care Unit (NICU) and Infants without (Non-NICU)*

| EHDI Benchmark | Group | Not Met Benchmark | Met Benchmark | Odds Ratio | Confidence Interval (95%) |
|---|---|---|---|---|---|
| **Screening** | NICU (*n* = 8,149) | 1,623 | 6,526 | 96.47* | (85.9–108.3) |
| | Non-NICU (*n* = 143,888) | 370 | 143,518 | | |
| **Diagnosis** | NICU (*n* = 227) | 110 | 117 | 3.17* | (2.36–4.25) |
| | Non-NICU (*n* = 1,167) | 267 | 900 | | |
| **Early Intervention Enrollment** | NICU (*n* = 38) | 16 | 22 | 1.88 | (0.87–4.04) |
| | Non-NICU (*n* = 111) | 31 | 80 | | |

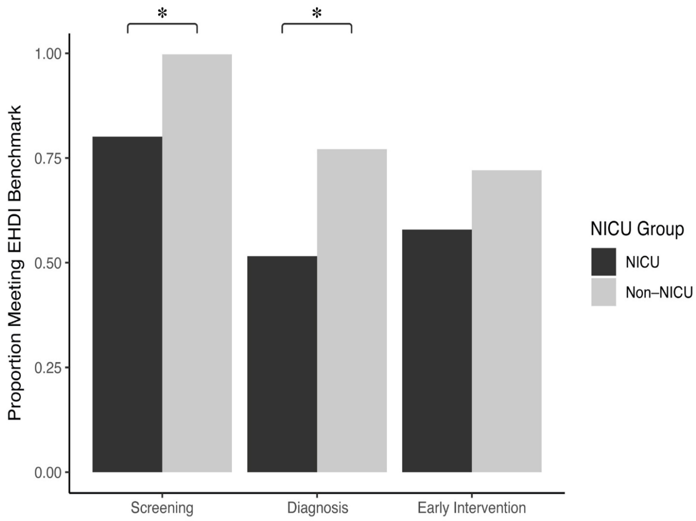
*Note*. EHDI = early hearing detection and intervention.
*indicates significance with alpha level = .05 level

### Benchmark Attainment by NICU Group

Our second research goal was to compare the proportion of infants who met EHDI age recommendations for NICU compared to non-NICU infants. Table 3 presents these results, including odds ratios and confidence intervals.

**Figure 5**

*Proportions Meeting Three EHDI benchmarks in Infants with Lengthy NICU Stays and Non-NICU Infants*




*Note*. EHDI = early hearing detection and intervention.
*indicates significance with alpha level = .05 level

Odds ratios express the likelihood of missing the recommended EHDI timeline for infants with lengthy NICU stays compared to non-NICU infants. For newborn hearing screening by one month of age, the odds of delay in NICU infants was 96.47 times that of non-NICU infants (CI = 85.9–108.3). For diagnostic evaluation by three months of age, the odds of delay in NICU infants was 3.17 times that of non-NICU infants (CI = 2.36–4.25). Both these differences were significant at the alpha = .05 level. There was no significant difference in the likelihood of enrolling in early intervention on time. Figure 5 displays these results.

### NICU-Related Delays by Maternal Race

Based on the differences in maternal race between our NICU and non-NICU groups (Table 2), we performed follow-up analyses with racially stratified data for screening and diagnostic benchmarks. Table 4 contains stratified odds of missing EHDI benchmarks in white, black, and other/multiracial NICU infants. Wide, overlapping 95% confidence intervals revealed no large differences in NICU-associated odds of missing either EHDI benchmark among white, black, and other/multiracial infants in our sample. We did not perform a stratified analysis by race for the early intervention benchmark due to low numbers of infants with data for this benchmark.

**Table 4**

*Odds Ratios and Confidence Intervals Associated with Missing EHDI Timing Benchmarks in Infants with Lengthy Stays in the Neonatal Intensive Care Unit (NICU) and Infants Without (Non-NICU), Stratified by Maternal Race*

| | | White | | Black | | Other/Multirace | |
|---|---|---|---|---|---|---|---|
| Screening | | Not Met Benchmark | Met Benchmark | Not Met Benchmark | Met Benchmark | Not Met Benchmark | Met Benchmark |
| | NICU | 1268 | 5252 | 187 | 603 | 168 | 671 |
| | Non-NICU | 300 | 120,221 | 30 | 10,166 | 40 | 13,131 |
| | | OR: 96.75* (CI: 85.06–110.05) | | OR: 105.09* (CI: 70.86–155.86) | | OR: 82.19* (CI: 57.72–117.04 ) | |
| Diagno | NICU | 77 | 98 | 17 | 9 | 16 | 10 |
| | Non-NICU | 206 | 730 | 27 | 66 | 34 | 104 |
| | | OR: 2.78* (CI:1.99–3.9) | | OR: 4.62* (CI: 1.83–11.63) | | OR: 4.89* (CI: 2.03–11.8) | |

*Note*. EHDI = early hearing detection and intervention.
*indicates significance with alpha level = .05 level

### Discussion

The findings of this study revealed that infants with a history of lengthy NICU stays access newborn hearing screening and diagnostic evaluation at later ages than non-NICU infants. Further, NICU infants met EHDI benchmarks for newborn hearing screening and diagnostic evaluation in lower proportions than non-NICU infants. On average, NICU infants were screened and seen for diagnostic assessment within the recommended age ranges; however, marked variability was present. This partly confirms the previous findings in Crouch et al. (2017). A discrepancy between the early benchmarks (screening and diagnostic evaluation) and the later enrollment in early intervention benchmark may result from NICU infants being referred for EI services for reasons other than PCHL. This would be consistent with clinical practice patterns observed for NICU graduates with preterm delivery and extremely low birth weight (Verma et al., 2003; Kuppala et al., 2012). However, due to the low number of infants whose enrollment in EI could be confirmed, we had lower power to detect true differences for this benchmark compared to screening and diagnosis benchmarks. Because of the nature of research with administrative data, we were not able to collect additional information that may reveal primary EI referral diagnosis. Thus, while we may find overall age at enrollment and proportions meeting the EHDI goal are equivalent among NICU and non-NICU infants, it remains important to ensure that children with PCHL receive services that address their

auditory and language development needs even in the presence of other qualifying diagnoses.

Our work demonstrates that infants with lengthy NICU stays do not achieve EHDI benchmarks at the same rate as their non-NICU peers. Failure to meet even one benchmark is associated with poorer long-term outcomes for children with PCHL, even if other benchmarks are met (Yoshinaga-Itano et al., 2017). However, this has not yet been examined in NICU infants alone. If delays are caused by lengthy NICU admissions, they may not lead to the same adverse effects on long-term outcomes as delays that stem from LTFU/D and clinical undermanagement.

A strength of this population-based study is that it incorporates the screening and outcomes of a large number of infants who were born in Iowa hospitals, regardless of hearing outcomes. Rather than excluding infants with normal hearing, we have used a winnowing treatment of the dataset. Thus, we were able to include benchmark timing data for the full population of Iowa infants who required care, even if they later went on to receive a diagnosis of normal hearing. A shorter time-to-diagnosis for children with normal hearing means fewer state public health resources tracking progress, shorter windows of parent concern, and an increased likelihood that diagnostic assessment can be completed under natural sleep. In addition, our work documents that although the NICU group defined in our analysis exhibited greater racial diversity than our non-NICU group, the relationship between lengthy NICU admission and risk of missing EHDI benchmarks appeared consistent across racial categories.

#### Limitations

The results from the first research question were meant to be descriptive in nature and capture the current clinical practice patterns regarding the timing of clinical activities. Our dichotomous categorization strategy pooled the data from infants with any length of NICU stay beyond five days and was not sensitive to discrepencies between intermediate term NICU stays and extended NICU stays. A major limitation of this investigation is the lack of access to gestational age that could be matched with infants in our two groups. Without gestational age, we are not able to characterize delays in NICU infants that stem from prematurity alone compared to infants with complex medical needs. Although the findings explored here are essential to characterize the current screening and follow-up timing trajectory for infants with lengthy NICU stays, a critical next step would be to consider delays in light of their gestationally adjusted age and comorbidities. Specific recommendations regarding gestational age adjustment would be a valuable addition to future JCIH position statements. Our analysis also excluded infants whose data concerning early benchmarks or NICU status could not be confirmed. These were the result of LTFU/D, incomplete data entry (such as missing information about risk factors), and parental withdrawal of consent to share detailed screening records with the IDPH.

A final limitation is that we calculated age at diagnosis using the first diagnostic assessment. Although we can safely assert that a confirmed diagnosis could not have preceded the first diagnostic appointment, we cannot exclude the possibility that this date represents a best-case scenario rather than a true age at confirmation of hearing loss. Holte et al. (2012) showed that, on average, families experienced delays between the initial diagnostic assessment and what they considered the confirmation of hearing loss. Recent EHDI literature suggests that some families go through up to five diagnostic evaluations before receiving a confident diagnosis of PCHL (Awad et al., 2019). If a transient conductive loss is suspected, the process of confirmation can be further delayed if families have long waits for ENT (Ear, Nose, and Throat specialist or otolaryngologist) appointments or if their physician prefers a wait-and-see approach for transient conductive loss. In the Outcomes of Children with Hearing Loss longitudinal study, parents reported reasons for delay included multiple re-screening, equivocal results, and protracted medical management (Holte et al., 2012; Walker et al., 2014). There is also the risk that results reported to EHDI as the first diagnostic assessment consist of repeated screening (i.e., OAEs only) instead of a true diagnostic evaluation. Concurrent quality checks at the IDPH during an overlapping period revealed that among children with hearing loss, in 87 of 299 cases the child's first evaluation with an audiologist consisted of a repeat screening despite being reported as a diagnostic evaluation (A. Hagerman, personal communication, August 12, 2020).

#### Future Directions

There are significant research opportunities in partnerships between researchers and state EHDI programs to improve service delivery in early hearing healthcare. Access to large public health databases of EHDI tracking results provides a unique opportunity to ask such questions and allows researchers to measure quality changes over time. Our work here examines one narrow piece of the JCIH clinical practice guideline. The data collected and tracked by state EHDI programs is rich with the level of detail necessary to examine other medical and audiological management patterns. Specific to NICU populations, future work should include a population-level assessment of the exclusive use of AABR screening technology. Using eSP records, we can track progress on this goal over time by comparing service dates with discharge dates and potentially address some of the delays revealed by the present research.

Our findings suggest that greater attention to timing benchmarks for NICU infants is needed within EHDI systems. Further research should assess the functional impact of these delays and whether a modified timeline or one executed with respect to gestationally adjusted age results in language and developmental outcomes on par with those of non-NICU peers. Research should also examine length of NICU stay with greater granularity (e.g., NICU stays of less than one month, six months, nine

months, 12+ months) and in the presence or absence of additional medical diagnoses. The JCIH now emphasizes the use of diagnostic ABR services prior to discharge for infants with lengthy admissions (JCIH, 2019), but we do not yet know how this update will change the care trajectories of NICU infants. Widespread access to inpatient diagnosis could remediate the NICU-related effects that we observed for the diagnostic benchmark (for infants born in hospitals with pediatric audiology services). It could also open the door for inpatient fitting of assistive devices when care teams confirm the presence of PCHL and the initiation of early intervention services. The heterogenous patient populations that require protracted NICU admission may not benefit from a one size fits all approach to improving EHDI delays. Expansion of inpatient diagnostic services and the development of other strategies to meet the needs of NICU infants should be family-centered to promote attention to and respect for a family's goals, ensure access to timely and evidence-based care, and provide coordinated services (Moeller et al., 2013). Care coordination would be especially important for families of NICU infants with complicated medical needs and who must balance competing concerns.

Finally, although we analyzed racially stratified odds ratios with respect to missing prescriptive EHDI benchmarks in a sub-set of NICU infants with longer admissions, significant gaps remain in our knowledge about EHDI benchmarks and racial disparities among both NICU and non-NICU infants. Future work may consider examining racial disparities among infants with any length of NICU stay, using more specific categorizations of racial background, including hearing outcomes, and integrating data on LTFU/D.

## Conclusion

This work contributes to the epidemiological literature about infant and early childhood hearing loss. Baseline characterization of the current EHDI trajectory for infants with lengthy NICU stays is a necessary step to refining recommendations for this population and if indicated, adapt JCIH recommendations in the future by accounting for gestational age. Our results reveal that overall, NICU babies achieve EHDI benchmarks at lower rates than non-NICU peers, including age at initial screening which has otherwise been considered fully-achieved in the literature. It may be appropriate to consider an alternate EHDI timeline based on gestationally adjusted age in formal clinical guidelines.

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