Identification of Oral Clefts as a Risk Factor for Hearing Loss During Newborn Hearing Screening

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Abstract: Objective. This study assessed whether children with oral clefts are appropriately classified as at-risk for hearing loss at the time of newborn hearing screening and describes their screening and diagnostic results.

Design. Birth certificates were used to identify children with cleft lip and palate or isolated cleft palate born in Washington State from 2008–2013. These were cross-referenced with the state's Early Hearing Detection, Diagnosis and Intervention (EHDDI) database. Multivariate logistic regression was used to examine associations.

Results. Birth records identified 235 children with cleft lip and palate and 116 with isolated cleft palate. Six children were listed as having both diagnoses. Only 138 (39%) of these children were designated as having a craniofacial anomaly in the EHDDI database. Children who were misclassified were less likely to have referred on initial hearing screening, OR 0.3, 95% CI [0.2, 0.5]. Misclassification of risk factor status was also associated with delayed hearing screening past 30 days of age or unknown age at screening, OR 4.4, 95% CI [1.5, 13.3], $p = 0.008$. Of 50 children with diagnostic results; 25 (50%) had hearing loss: 18 conductive, 2 mixed, and 5 unspecified.

Conclusion. A majority of children with oral clefts were misclassified regarding risk factor for hearing loss in the EHDDI database.

Acronyms: EHDI = Early Hearing Detection and Intervention; EHDDI = Early Hearing-loss Detection, Diagnosis and Intervention; DHH = deaf or hard of hearing; JCIH = Joint Committee on Infant Hearing; NICU = neonatal intensive care unit

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Introduction

The Joint Committee on Infant Hearing (JCIH) has established the 1-3-6 guideline for detection, diagnosis, and intervention for congenital hearing loss (JCIH, 2007; Bower & St. John, 2014). All children should undergo hearing screening by 1 month of age, diagnostic audiology assessment by 3 months of age for those who do not pass screening, and enrollment in early intervention by 6 months of age for those with hearing loss. In addition,

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children with certain risk factors for hearing loss should undergo diagnostic audiology assessment by 24 to 30 months of age, even if they pass their initial hearing screen (JCIH, 2007; Beswick, Driscoll, & Kei, 2012). To promote adherence to these guidelines, states have created Early Hearing Detection and Intervention (EHDI) programs; in Washington State, the program has been titled, "Early Hearing Detection, Diagnosis and Intervention" (EHDDI). These programs can monitor follow-up for children who do not pass hearing screening or who have risk factors for hearing loss (White, 2014; Gracey, 2003).

The EHDDI program relies on newborn hearing screeners to report hearing screening results and indicate which children have risk factors for delayed onset or progressive hearing loss. In Washington State, newborn hearing screenings can be performed by nursing staff, obstetric technicians, audiologists, midwives, and staff from contracted newborn hearing screening companies. Individuals do not need certification to conduct newborn hearing screenings, but EHDDI program staff provide training and outreach. This training includes information about risk factors for delayed onset and progressive hearing loss and how to report risk factors to the EHDDI program. Newborn hearing screeners may also receive training from the hospital, clinic, or company where they work. Washington State does not have legislation that mandates newborn hearing screening or reporting results to the EHDDI program. Although screening is voluntary, it is estimated that 96% of all infants born in Washington State receive a newborn hearing screening (Weisman, 2014).

There are 29 audiology clinics throughout the state that provide comprehensive diagnostic audiological assessment for infants. These clinics meet Washington State's Protocol for Diagnostic Audiological Assessment: Follow-up for Newborn Hearing Screening. This protocol is based on the JCIH 2007 position statement and was created by a workgroup of 22 audiologists with expertise in the screening and diagnosis of hearing loss in newborns and infants (Washington State Department of Health, 2011).

Children with craniofacial anomalies are at greater risk of hearing loss (Lieu, Ratnaraj, & Ead, 2013; Yelverton et al., 2013; Beswick, Driscoll, Kei, Khan, & Glennon, 2013). Most commonly, children with these anomalies will experience conductive hearing loss due to anatomic abnormalities affecting middle ear function, although sensorineural hearing loss can occur as well (Swibel Rosenthal, Caballero, & Drake, 2012). The most common craniofacial anomaly is cleft lip and palate, which occurs in approximately 10 per 10,000 live births; isolated cleft palate is also relatively common with an incidence of 6.5 cases per 10,000 births (National Birth Defects Prevention Network, 2010). These craniofacial anomalies can be grouped together under the term oral clefts. Children with oral clefts have high rates of conductive hearing loss, most commonly associated with Eustachian tube dysfunction (Kuo et al., 2014).

The objective of this study was to determine what proportion of children with cleft lip and palate or isolated cleft palate were correctly identified as having a craniofacial anomaly at the time of newborn hearing screening, and to determine if there was an association between correct identification of risk factor status and adherence to guidelines for newborn hearing screening.

Materials and Method

Prior to investigation, approval for this study was obtained from the Washington State Department of Social and Health Services' Human Research Review Section. In Washington State, the long form birth certificate records from January 1, 2008 to December 31, 2013 were electronically queried. The long form birth certificate includes a field for designating whether a child is born with an oral cleft, which includes either cleft lip and palate or isolated cleft palate.

Birth record numbers were then cross-referenced with the state's EHDDI database to identify the cohort. The EHDDI database was queried to obtain information regarding birth weight, race and ethnicity, maternal age, and presence of JCIH hearing loss risk factors. The Washington State newborn hearing screening card includes data regarding five risk factors for delayed onset or progressive hearing loss including the following: (a) neonatal intensive care unit (NICU) stay greater than 5 days, (b) syndrome with stigmata of hearing loss, (c) family history of hearing loss, (d) presence of craniofacial anomalies, and (e) in-utero infection. In addition, screeners can denote that a child has no risk factors present. Hearing screening and diagnostic audiologic test results were recorded. The birth hospital location and site of diagnostic audiology assessment were also recorded for each child. The demographic data were collected because factors such as birthweight, socioeconomic status, and geographic location may impact the potential association between risk factor misclassification and adherence to newborn hearing screening guidelines.

After data collection was completed, cases were reviewed to determine the number of children who underwent hearing screening and diagnostic testing, and at what age the testing occurred. Diagnostic testing results were then reviewed to determine the types and configuration of hearing loss.

The cohort was divided into two groups based on whether the child had been appropriately identified as having a craniofacial anomaly (risk factor 4) on EHDDI screening card. One group consisted of children with oral clefts correctly classified as having risk factor 4, while the other consisted of children with oral clefts who were misclassified by not having risk factor 4 noted at time of screening. Comparisons were made between the two groups regarding demographic characteristics, presence

of other risk factors, and adherence to 1-3-6 guidelines for screening and diagnostic testing.

All children who had completed newborn hearing screening by 30 days of age were considered to have met the guideline for screening. Children who did not have record of screening were noted, but excluded from further analysis. Among children who did not pass their hearing screen, those who completed diagnostic testing by 90 days of age were considered to have met the guideline for diagnostic testing.

Analysis

Based on the presence or absence of craniofacial risk factor designation, univariate analysis was performed to calculate descriptive statistics, including means and proportions, for the two groups within the cohort. For continuous variables, an unpaired t-test was used for inferential testing; for binary variables, chi-square testing was used to determine significance.

Multivariate logistic regression was then used to investigate association between risk factor status and adherence to screening and diagnostic guidelines. Risk estimates were expressed as odds ratios (OR) and 95% confidence intervals (CI). The following variables were considered as potential confounding covariates: maternal age, birth weight, race or ethnicity, and distance from birth hospital to site of diagnostic audiology assessment. Data that met $p < 0.05$ were considered statistically significant. Stata 13.1 (Stata Inc, College Station, TX) statistical software was used for all analyses.

Results

Total number of resident births in Washington State from January 2008 to December 2013 was 526,774. Birth certificate records identified 357 children with oral cleft malformations born during this time period. There were 235 children with cleft lip and palate and 116 with isolated cleft palate for a birth prevalence of 4.5 per 10,000 births and 2.2 per 10,000 births, respectively. There were 6 children who had been erroneously designated as having both diagnoses. Of 357 children, only 138 (39%) were designated as having a craniofacial anomaly in EHDDI database, while 130 (36.4%) were erroneously designated as having no risk factor for hearing loss. The other 89 children were not provided with risk factor classification at time of screening.

Table 1 contains the characteristics of the cohort based on whether or not they were designated as having craniofacial risk factor. Sixty-six percent of children with isolated cleft palate were misclassified compared to 59% of children with cleft lip and palate. Children who were correctly classified had a mean birthweight of 3318.9 g ($SD = 600.7$ g), which was higher than the mean birthweight among

children who were misclassified 3189.4 g ($SD = 670.5$ g), but this finding did not achieve significance with unpaired t-test, p-value = 0.07.

Table 1
Characteristics of Children with Oral Clefts by Craniofacial Risk Factor Identification

	Craniofacial risk factor identified at hearing screening	Craniofacial risk factor not identified at hearing screening
Total number (%)	138 (38.7%)	219 (61.3%)
Craniofacial anomaly*		
Cleft lip & palate (%) ($n = 235$)	96 (40.9%)	139 (59.1%)
Isolated cleft palate (%) ($n = 116$)	40 (34.5%)	76 (65.5%)
Mean birthweight in grams, (SD)	3318.9 (600.7)	3189.4 (670.5)
Racial/ethnic minority (%)** ($n = 118$)	56 (47.5%)	62 (52.5%)
Mean maternal age in years, (SD)	28.4 (6.2)	28.1 (5.7)
Presence of other risk factors for hearing loss (%):		
Neonatal ICU ($n = 59$)	21 (35.6%)	38 (64.4%)
Stigmata of syndrome ($n = 10$)	5 (50%)	5 (50%)
Family history ($n = 11$)	4 (36.4%)	7 (63.6%)
In-utero infection ($n = 0$)	--	--

* 6 children were listed as having both malformation types
**Not all children had information available regarding race or ethnicity

There were no significant differences between the groups based on race and ethnicity, maternal age, or presence of other risk factors. Of the 357 children, 59 children (16.5%) also had NICU risk factor. No child had a risk factor for in-utero infection, and only a small proportion of children had any of the other risk factors.

Mean age at first hearing screening was 5.4 days (range 0–135 days); 217 (60.1%) underwent screening by the second day of life. There were 25 children (7%) who did not undergo newborn hearing screening; none of these children were correctly classified as having a craniofacial risk factor. Twelve children (3.6%) underwent screening after 30 days of age, and 16 children were of unknown age at time of screening.

Of the 332 children who underwent hearing screening, 60 (18%) did not pass their hearing screen (see Figure 1) and 36 (60%) underwent diagnostic testing. The mean age at diagnostic testing for these children was 74.8 days (range 8–232 days). Thirty of the 60 (50%) who referred underwent diagnostic testing by 90 days of age.
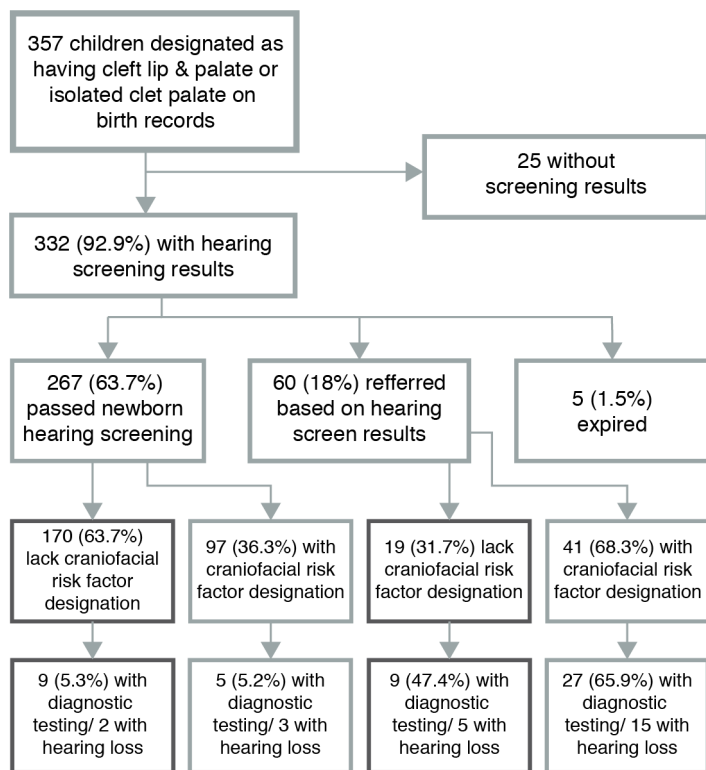
not, although this difference did not achieve statistical significance. Therefore, birth-weight was included in the model as a binary variable based on a child's birth weight being less than or greater than 2500 g. After adjustment for birth-weight status, children who were misclassified were less likely to have referred on hearing screening, OR 0.3, 95% CI [0.2, 0.5], *p* < 0.001, see Figure 2.
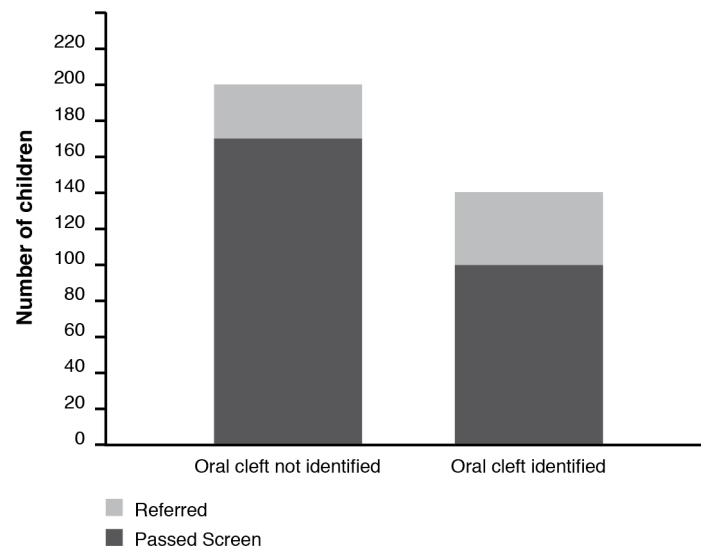

Figure 1. **Flowchart of screening and diagnostic characteristics of children with oral clefts.**

Diagnostic results were available for an additional 14 children who had passed their hearing screen, so that a total of 50 children had diagnostic results. Of these, 25 (50%) had a final diagnosis of hearing loss: 18 conductive, 2 mixed, and 5 unspecified.

Logistic regression was performed to investigate associations between risk factor status and not passing the newborn hearing screen. There were not significant differences in race/ethnicity or maternal age between children who were correctly classified compared with those who were not, so these covariates were not included in the regression model. Distance from a child's birth hospital to the audiology center of referral was calculated and explored as a covariate, but it was not significant.

Low birth-weight was found to be significantly associated with likelihood of delayed screening. Of 12 children who underwent screening after 30 days of age, 6 (50%) weighed less than 2500 g at birth, *p*-value = 0.001. In addition, as noted above, there was a difference in the mean birth weight between children who were correctly classified on risk factor status and those who were

Figure 2. **Among children identified as having oral clefts on birth certificate, those who were not identified as having a craniofacial risk factor at time of hearing screening were less likely to have referred on initial hearing screening, OR 0.3, 95% CI [0.2, 0.5], *p* < 0.001.**
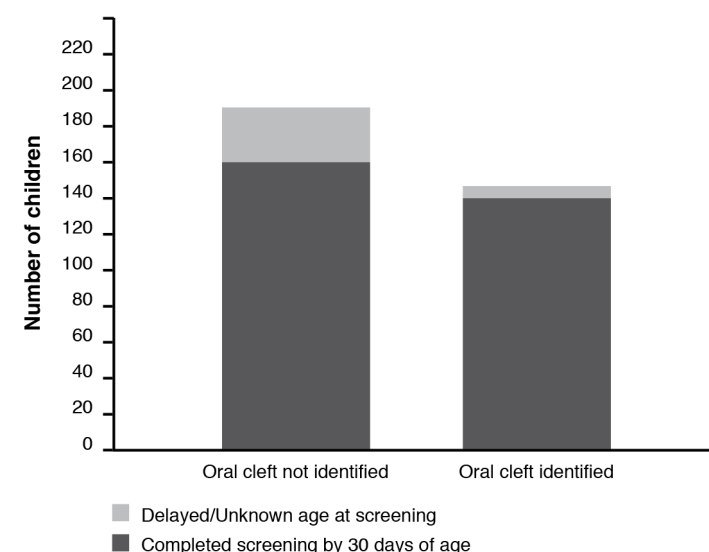

Figure 3. **Stacked bar chart compares adherence to hearing screening guidelines among children with oral clefts. Those who were not identified as having a craniofacial risk factor at time of hearing screening were more likely to also have delayed or unknown age at screening, OR 4.4, 95% CI [1.5, 13.3], *p* = 0.008.**

When association between risk factor status and adherence to 1-3-6 guidelines was investigated, misclassification of risk factor status was associated with delayed hearing screening past 30 days of age or unknown age at screening, OR 4.4, 95% CI [1.5, 13.3], p-value = 0.008, see Figure 3. This study also found that misclassification may be associated with delayed diagnostic testing past 90 days of age or unknown age at testing, OR 5.7, 95% CI [0.9, 38], p = 0.07, see Figure 4. However, this result did not reach statistical significance. The results of the logistic regression models are also summarized in Table 2.
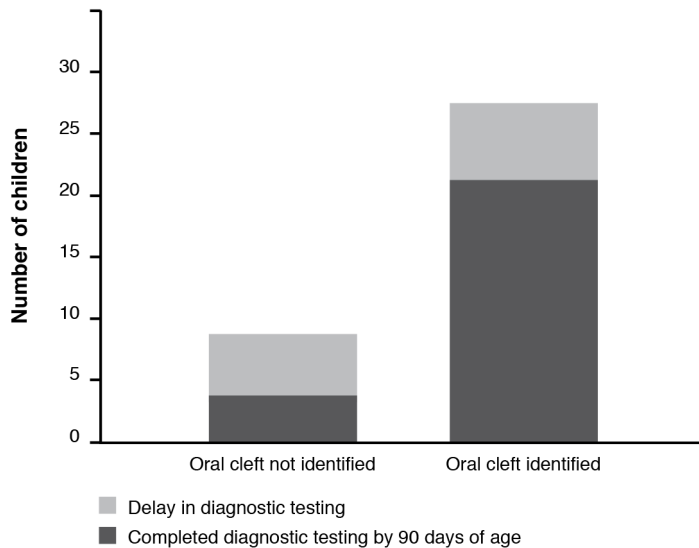

Figure 4. **Stacked bar chart compares adherence to diagnostic testing guidelines among children with oral clefts who did not pass their hearing screen. Those who were not identified as having a craniofacial risk factor at time of hearing screening may be more likely to also have delayed or unknown age at diagnostic testing, OR 5.7, 95% CI [0.9, 38], p = 0.07; however, this result did not reach statistical significance.**

Table 2
Logistic regression models of association between risk factor misclassification and newborn hearing outcome

Outcome	Coefficient	Coefficient Confidence Interval	Odds Ratio	Odds Ratio Confidence Interval	p-value
Child refers on 1st screen	-1.2	-1.7, -0.6	0.3	0.2, 0.5	<0.001
Child has delayed or unknown age at hearing screening	1.5	0.4, 2.6	4.4	1.5, 13.3	0.01
Child has delayed or unknown age at diagnostic assessment	1.4	0.5, 2.7	5.7	0.9, 38	0.07

Discussion

In 2013, JCIH issued a statement emphasizing the importance of accurate data management systems for newborn hearing screening, stating that such systems are critical to facilitate timely, well-coordinated entry into early intervention for all children who are deaf or hard of hearing (DHH). Other studies have come to similar conclusions (Shulman et al., 2010).

States have dedicated substantial resources to early identification of hearing loss through the establishment of EHDI programs; however, debate remains as to the role that risk factor designation should play in hearing screening and surveillance. Not all states track risk factors for hearing loss as part of a hearing screening program. In a recent national survey, about 65% of states had databases containing information about risk factors for hearing loss (Houston, Behl, White, & Forsman, 2010).

Evidence supports universal screening as preferable to screening based upon risk factors. Universal newborn hearing screening is associated with improved language and literacy outcomes among children who are DHH because it facilitates their enrollment in early intervention services (Yoshinaga-Itano, 2003). Lack of intervention at a critical period in development may have long-lasting impact. In a prospective cohort study of children who were DHH, Pimperton and colleagues found that disparity in reading comprehension between those enrolled in early intervention and those who were not continued to widen with age (Pimperton et al., 2016). Wake and colleagues (2016) recently performed a population-based investigation of Australian states with similar demographic characteristics, finding that universal screening was associated with greater improvement in language outcomes than risk-factor based screening. Other studies have raised questions as to which risk factors for hearing loss are important to monitor. For example, a retrospective study of risk factor registry in Queensland, Australia, found two risk factors, family history and craniofacial anomalies, to predict the occurrence of postnatal hearing loss (Beswick et al., 2013). However, a subsequent study found family history of hearing loss to be of low yield in predicting development of hearing loss (Driscoll, Beswick, Doherty, D'Silva, & Cross, 2015), and it is no longer monitored as a risk factor in the United Kingdom (Sutton et al., 2012). A more recent study found that approximately 10% of children have a risk factor for delayed onset or progressive hearing loss, and of those children, 2.3% develop a permanent hearing loss by age 3 (Dumanch et al., 2017). This study also found the presence of craniofacial anomalies to be among the factors that placed a child at the highest risk for permanent postnatal hearing loss.

However, risk factor identification may be a useful adjunct to universal screening in that it could help target limited resources to those at greatest risk. EHDI programs across the nation continue to face challenges,

including incomplete data reporting and lack of follow-up (Nikolopoulos, 2015). Using a nationwide survey, Gaffney, Green, & Gaffney (2010) found that two-thirds of children who did not pass their final hearing screening in 2005 did not have documentation of a diagnostic test result, primarily due to loss to follow-up. In 2014, a similar study found that more than one-third of children referred lacked a diagnostic result (Alam, Gaffney, & Eichwald, 2014). A recent nationwide survey of parents found that more than 35% could not recall whether their child had undergone screening, and many remained unsure about guidelines for follow-up (Pynnonen et al., 2016). If we acknowledge there are limited resources available for counseling families regarding screening, testing, and surveillance; then perhaps there is a role for proper identification of risk factors as a way to assist efforts for targeted counseling.

The current study used birth certificate records to determine that only 39% of children with oral clefts were correctly classified as having a craniofacial risk factor at the time of newborn hearing screening. Children who were misclassified were less likely to have met 1-3-6 guidelines for screening and diagnosis. Unfortunately, early intervention data were not available at the time of this study. JCIH guidelines call for all children with craniofacial anomalies to complete one diagnostic assessment by 24 to 30 months of age. However, this study found only 50 (14%) of 357 children with oral clefts to have diagnostic results in the state's EHDDI database. Of the children who did not pass screening, 60% had a diagnostic test result, which is consistent with the national percentage (Alam et al., 2014).

Oral clefts are associated with conductive hearing loss (Schönmeyr & Sadhu, 2014); this increased risk is thought to be due to abnormality of the tensor veli palatini muscle, which inserts onto the membranous portion of the Eustachian tube to equilibrate the middle ear space. Children with cleft palate, or even submucous cleft, will often have persistent middle ear effusion requiring tympanostomy tube placement (Reiter, Brosch, Wefel, Schlömer, & Haase, 2011; Smillie, Robertson, Yule, Wynne, & Russell, 2014; Szabo 2010). Of the 25 children found to have hearing loss in this study, most had a conductive hearing loss; these findings appear similar to previous studies (Viswanathan, Vidler, & Richard, 2008).

Tympanostomy tube placement is frequently performed at the same time as cleft palate repair, often around 1 year of age (Kosowski, Weathers, Wolfswinkel, & Ridgway, 2012). Craniofacial centers typically offer pediatric audiology services and diagnostic assessment, especially in the setting of tympanostomy tube placement. Therefore, we need to investigate how well these results are being reported to EHDDI. A next step for this investigation would be to longitudinally track clinical records for the children within this cohort, perhaps at the state's largest tertiary care facility, Seattle Children's Hospital, to determine how many children have records of evaluation and intervention.

Given the frequency of middle ear effusion among patients with cleft palate, recent studies have also questioned whether children with oral clefts should undergo diagnostic audiology testing prior to tympanostomy tube placement (Jordan & Sidman, 2014). Perhaps provider uncertainty regarding the utility of current guidelines could also be contributing to lack of adherence to 1-3-6 guidelines among children with oral clefts. More qualitative data gathering from pediatric otolaryngologists and audiologists might help to clarify this point further.

As an observational cohort study, this investigation had several limitations. It was a population-based study, but the frequency of certain events was quite low, which can make it difficult to determine statistical significance. In addition, a number of children had missing data. There is also the potential for additional confounding by unmeasured factors. Previous studies found that children from rural settings are more likely to have delayed screening or diagnosis (Bush et al., 2015), but this dataset did not include home address information. Distance from birth hospital to audiology center was explored as a covariate, but it was found to not be significant.

If states choose to allocate resources for documentation of risk factors, it is important for this process to be accurate. This study found oral clefts, the most common craniofacial risk factor, to be widely under-reported. If craniofacial risk factors are being under-reported, it is likely that other risk factors are as well. These findings prompt the question of what can be done to improve the system.

Nationwide, efforts are underway to improve integration of electronic health records among clinical providers. Perhaps integration could expand to include public health programs, such as EHDI, although privacy concerns would have to be addressed (Uhler, Thomson, Cyr, Gabbard, & Yoshinaga-Itano, C., 2014). To our knowledge, efforts are underway to implement linkage models in Utah and Nebraska between birth certificate and other vital records and newborn screening registries (McVicar, 2014; Northrop, 2014). It will be important to monitor how effective these systems are at correctly identifying risk factors and whether this change has any effect on rate of enrollment in early intervention programs.

In the meantime, additional education could be provided to those who perform newborn hearing screening regarding the importance of correctly identifying an infant's risk factor status. Newborn hearing screeners are often clinical staff or technicians who receive training by either hospitals or contractor companies. One commonly used curriculum is provided through the National Center for Hearing Assessment and Management (NCHAM), but there are not standardized methods for assessment of screener competencies in the vast majority of states.

In Washington State, EHDDI staff conduct site visits and hold annual meetings to train screeners. About half of hospital newborn hearing screening programs

in Washington require that newborn hearing screeners achieve annual competencies in screening. It might be helpful to develop a training curriculum for hearing screeners that includes education related to identification of risk factors for hearing loss, including oral clefts. Regulations could be set that require hospital screeners to participate in training or meet certain competencies in order for institutions to receive certification from states. For example, California is now requiring that inpatient hearing screening be administered by certified facilities, and certification is required for reimbursement for hearing screening services provided to MediCal eligible infants (California Newborn Hearing Screening Program, 2016). If more states had a certification process that required particular training elements, there may be improvement in screening and reporting of risk factors.

Conclusion

Using birth certificate records, this population-based study found that a majority of children with oral clefts were misclassified regarding hearing loss risk in the Washington state EHDDI database. Children who were misclassified may be less likely to meet 1-3-6 guidelines for screening and diagnostic audiology assessments. As states take steps to improve data systems and standards for hearing screening certification, it is important to investigate accuracy and effectiveness of newborn hearing screening systems to improve care and services for children who are DHH.

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