

Implementing a Hearing Screening Readiness Assessment Tool for Preterm and Term Neonates in the Newborn Intensive Care Unit: A Pilot Project

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Abstract

Newborn intensive care unit (NICU) patients are at risk for hearing loss. Early detection mitigates consequences of speech and language delay. The Joint Committee on Infant Hearing (JCIH, 2019) recommends hearing screening (HS) on all infants by 1 month of age. Routinely, hearing screening is performed around time of NICU discharge, oftentimes beyond JCIH recommendations. Automated auditory brainstem response (AABR) screening can be performed once an infant reaches 32 to 34 weeks corrected gestational age. Our project aimed to reduce HS delay among NICU infants. We created and implemented a HS assessment tool defining gestational age (GA) and medical stability parameters for initial HS. Data were compared between 100 infants pre-implementation and 325 infants post-implementation. After implementation, infants had HS performed 4 days earlier in days of life ($p = 0.28$) and 4 days earlier, prior to discharge ($p < 0.0001$). Infants born before 34 weeks GA had HS performed 11 days of life earlier ($p = 0.02$) and 14 days earlier prior to discharge ($p < 0.0001$). More preterm infants completed HS at less than 1 month of age (34% vs 61%, $p = 0.06$). Earlier HS is associated with a 6.3% increase in false positive screens among premature neonates, requiring repeat screening. Because evidence suggests the tool may promote earlier HS for preterm infants, additional work on optimizing the HS technique is needed to lower false positive results.

Keywords: hearing screening, preterm, neonates, neonatal intensive care unit, assessment tool

Acronyms: AAP = American Academy of Pediatrics; AABR = automated auditory brainstem response; EHDI = Early Hearing Detection and Intervention; GA = gestational age; HS = hearing screening; NICU = Newborn Intensive Care Unit; QI = quality improvement

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Hearing loss is one of the most common congenital birth conditions. The occurrence rate for sensorineural hearing loss is about 1.7 per 1,000 (0.17%) live births in the United States (Centers for Disease Control and Prevention, 2021) and up to 24% among high-risk neonates admitted to the Newborn Intensive Care Unit (NICU; Berg et al., 2005). NICU patients are at higher risk for hearing loss due to prematurity, sepsis, ototoxic medication exposure, congenital viral infections, genetic syndromes, congenital craniofacial anomalies, hyperbilirubinemia, hypoxia, and noise exposure from life-saving medical support (Wroblewska-Seniuk et al., 2016). Early detection and intervention may greatly improve long-term neurodevelopmental outcomes for deaf/hard of hearing infants (Joint Committee on Infant Hearing, 2019).

The Joint Committee on Infant Hearing (JCIH), which includes representation from the American Academy of Pediatrics (AAP), supports Early Hearing Detection and Intervention (EHDI) guidelines (JCIH, 2019). The EHDI 1-3-6 goals are to have all infants receive initial hearing screening by 1 month of age, a diagnostic evaluation no later than 3 months (if initial HS warrants additional testing), and early intervention no later than 6 months of age (if otologic evaluation confirms a deaf/hard of hearing outcome). JCIH 2019 guidelines support intervention as early as 3 months if possible.

Meeting EHDI guidelines can be challenging in the NICU population. It may not be practical to perform initial HS on NICU infants by 1 month of age. In addition to prematurity, NICU infants may be too critically ill to tolerate a HS

or the life-supporting medical equipment may interfere with the instrumentation required for hearing screening. The JCIH recommends HS for NICU infants as soon as they are medically stable (JCIH, 2019). However, JCIH does not define medical stability in respect to the NICU population. There are no specific recommendations on what parameters define medical stability, and there is no consensus on how to determine optimal timing for newborn HS in regard to NICU infants. Evidence shows that the automated auditory brainstem response (AABR) screen can be reliably performed at 32 to 34 weeks corrected GA (Wroblewska-Seniuk et al., 2016), yet HS is often performed immediately before NICU discharge and greater than 30 days of age (Chung et al., 2019; Patel et al., 2018).

The goal of this quality improvement (QI) project was to develop a HS readiness assessment tool aimed at improving the timing of initial HS for NICU patients. The specific aim was to reduce the age at which initial HS is performed.

Method

Setting/Population

Our QI project was conducted at a 52-bed level III NICU located in Salt Lake City, Utah. This NICU is a major birthing hospital for the Salt Lake region, as well as a referral center for four neighboring states. This unit provides care for critically ill newborns with gestational ages ranging from approximately 23 weeks to over 40 weeks, with more than 600 NICU admissions annually. The study population included all preterm and term NICU patients who required hearing screening. The study excluded infants who were being discharged home on comfort care/hospice, died prior to HS eligibility, or who were being transferred to a different facility or lower level of care.

Intervention

To achieve this specific aim, the project underwent three phases. In Phase 1, a retrospective chart review was conducted on all NICU patients with HS done 3 months prior to study implementation. We evaluated the timing and GA of these NICU patients upon receiving initial hearing screening. In Phase 2, a multidisciplinary team including audiologists, developmental care specialists, nurses, and nurse practitioners was formed. The team developed an updated HS protocol based on current literature and expert opinion. The team considered criteria eligibility for HS, parameters of medical stability, and medical interventions or factors that may interfere with HS results.

The followings criteria were used to evaluate HS readiness. The neonate:

- 1) Is at least 34 weeks corrected gestational age
- 2) Maintains stable body temperature without external heat source
- 3) Requires minimal or no respiratory support
- 4) Has no critical self-supporting lines and/or drains; excluding feeding tubes

- 5) Has completed aminoglycoside treatment course (if needed)
- 6) Is not receiving medication treatment for neonatal abstinence syndrome
- 7) Tolerates routine care without decompensation

Initially, we attempted to integrate the above criteria as a function of the electronic health record; however due to constraints associated with the COVID-19 pandemic, the informatics department was unable to coordinate this in a timely manner. An alternative paper HS readiness assessment tool was created based on the above criteria (Appendix). This readiness tool was used to alert NICU providers when an infant met criteria for initial hearing screening.

In Phase 3, NICU providers received mandatory education on the new screening tool/guideline via an online presentation. Upon completion of training, the HS readiness assessment tool was implemented in the NICU and follow up outcome data were collected to evaluate tool effectiveness. The team set up a process to alert audiology to perform HS, as well as electronic documentation of HS results. Once a provider deemed an infant eligible for HS via the assessment tool, the form was placed in a pre-determined box for audiology. Audiology would then confirm readiness and perform HS per the new guideline, documenting HS results in the electronic health record. Weekly educational reminders were provided to promote continued awareness of the screening tool.

Measures

Outcome measures included the day of life at which initial HS was performed, the number of days HS was done prior to discharge, the proportion of HS performed prior to Day of Life 30, and the days between infants meeting HS eligibility and HS being performed. False positive rates were evaluated as a balancing measure in relation to performing earlier HS, particularly in the preterm population. Provider use of the HS readiness assessment tool was monitored as a process measure to determine if implementation improved outcomes.

Analysis

Data were compared between 3 months pre-implementation (July 2020–early October 2020) and 9 months (mid October 2020–July 2021) post-implementation. Descriptive statistics of median and interquartile ranges were used to describe demographic data including birthweight, GA, and length of stay. A subgroup comparative analysis of neonates who were born at less than 34 weeks GA was performed. Outcome measures were compared between the pre- and post-implementation period. The Mann-Whitney U-test, also known as a Wilcoxon rank-sum test, and Fisher's exact test were used for ordinal data or continuous data that were not normally distributed. A two-sided *p*-value of less than 0.05 was considered statistically significant. Statistical analysis was performed using GraphPad Prism Version 9.2.0 for MacOs (GraphPad Software, La Jolla, California, USA, <http://www.graphpad.com>).

Ethical Considerations

Institutional review board approval was obtained from the University of Utah. The committee approved a waiver of informed consent as it was a QI project with minimal risk to patients.

Results

Following implementation of the tool, informal feedback was gathered during weekly multidisciplinary rounds regarding the new HS process and the HS tool. Practitioners verbalized a better understanding of HS readiness and an increase in knowledge regarding current HS recommendations, although this feedback was not quantified.

Baseline data was gathered from 100 NICU patients who had HS performed within the 3-month period prior to QI project implementation. During the post-implementation period, 325 NICU patients had HS done within a 9-month period. The neonates' demographic characteristics were not significantly different between the two periods (Table 1). After study implementation, initial HS was performed 4 days earlier in age for all neonates, although this was not statistically significant (15 vs. 11 days, $p = 0.28$; Table 2). However, the number of days HS was performed prior to discharge improved from 3 days to 7 days earlier ($p < 0.0001$) with the length of stay not being significantly longer among the post-implementation population (Table 2).

Table 1

Demographic Characteristics of Neonates During the Pre- and Post-Implementation Periods

	Neonates of All Birth GA				Neonates of Birth GA < 34 Weeks			
	Pre (n = 100)	Post (n = 325)	Wilcoxon statistic	p value*	Pre (n = 29)	Post (n = 126)	Wilcoxon statistic	p value*
BW (kg), median (IQR)	2.5 (1.8–3.3)	2.4 (1.8–3.1)	15030	0.256	1.5 (1.2–1.9)	1.7 (1.4–2.0)	2065	0.277
GA (wk), median (IQR)	36 (33–38)	35 (33–38)	14727	0.156	32 (29–33)	32 (30–33)	1942	0.599
LOS (d), median (IQR)	19 (8–32)	21 (10–43)	17906	0.123	41 (30–74)	47 (30–67)	1839	0.958

Note. BW = birthweight; d = days; GA = gestational age; IQR = interquartile range; kg = kilogram; LOS = length of stay; n = number in category; Pre = pre-implementation period; Post = post-implementation period; wk = weeks. *Wilcoxon rank sum test.

Table 2

Outcomes for Neonates During the Pre- and Post-Implementation Periods

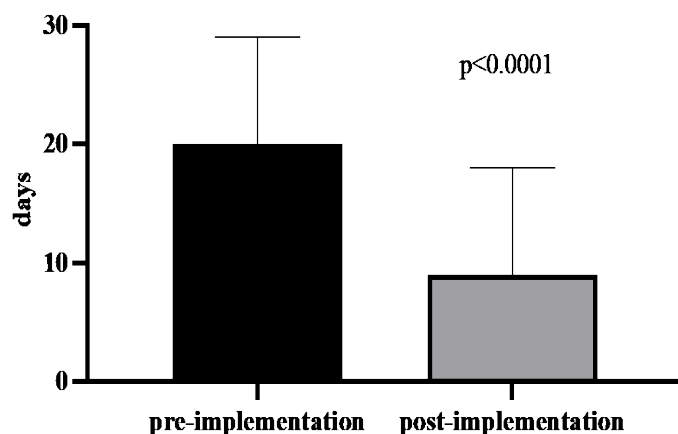
	Neonates of All Birth GA				Neonates of Birth GA < 34 Weeks			
	Pre (n = 100)	Post (n = 325)	Wilcoxon statistic	p value	Pre (n = 29)	Post (n = 126)	Wilcoxon statistic	p value
Percentage of HS was done at < 31 DOL (%)	80%	84%	N/A	0.45+	34%	61%	N/A	0.06+
DOL when HS was done (d), median (IQR)	15 (6–27)	11 (7–23)	15161	0.311*	37 (24–65)	26 (15–40)	1202	0.004*
Days between HS was done and NICU discharge (d), median (IQR)	3 (2–4)	7 (2–18)	22275	< 0.001*	4 (3–8)	18 (11–26)	3141	< 0.0001*

Note. d = day; DOL = days of life; GA = gestational age; HS = hearing screening; IQR = interquartile range; n = number in category; Pre = pre-implementation period; Post = post-implementation period. *Wilcoxon rank sum test; +Fisher's exact test.

The subgroup analysis showed significant impact among neonates who were born at less than 34 weeks GA. Compared to the pre-implementation period, there was an increased percentage of HS being done by 1 month of age, meeting the JCIH recommended goal, (34% vs. 61%, $p = 0.06$) in the post-implementation period. During the post-implementation period, preterm neonates had initial HS done 11 days earlier ($p = 0.02$) and 14 days earlier prior to discharge ($p < 0.0001$; Table 2). HS was also done 11 days sooner once corrected GA eligibility was met ($p < 0.0001$; Figure 1).

Performing HS at an earlier GA increased the risk of false positive results. During the pre-implementation period, 3 out of 100 neonates (3%) had an abnormal AABR hearing screen. They all had confirmed hearing loss and were all born at over 34 weeks gestational age. The false positive rate was 0%. During the post-implementation period, 25 out of 325 neonates (7.7%) failed the first AABR, 15 of these infants had confirmed hearing loss on repeated testing. The prevalence of hearing loss in this cohort was 4.6%, which was not significantly different compared to the pre-implementation period ($p = 0.58$). Of the infants

Figure 1
Pre and Post Data



Note. Days between when hearing screen was performed after reaching corrected gestational age of 34 weeks, compared to the pre-implementation ($n = 29$) and post-implementation period ($n = 126$). 20 vs 9 days, $p < 0.0001$.

who failed the first AABR, ten passed the repeat HS. The false positive rate of the initial HS was 3% (10/325) higher than the pre-implementation period ($p = 0.08$). The HS sensitivity and specificity were 100% and 96.8% respectively during the post-implementation period. The majority of the false positive cases was found among neonates who were born at less than 34 weeks gestational age (8/126, 6.3%).

The paper HS assessment tool usage was only tracked for the first three months during the nine month post-implementation period. During this time period the paper HS assessment tool was only used for 55% of eligible patients. Paper tool usage was not tracked for the remainder of the post-implementation period due to low adoption rates among caregivers.

Discussion

NICU infants experience HS delay more often compared to their non-NICU peers. Previous studies suggest that creating specific clinical guidelines for the timing of early hearing screening in NICU infants may be warranted to improve the delay NICU patients experience in regards to initial HS (Sapp et al., 2020).

An extensive literature review was performed prior to project implementation; to our knowledge this is the first study in which a HS readiness assessment tool was developed to define GA and medical stability criteria for HS readiness in the NICU population. Our QI project promoted HS to be done earlier to meet the JCIH recommended HS goal. The major impact was seen among neonates born before 34 weeks GA by significantly reducing the age at which initial HS was performed. HS was also done sooner once GA eligibility was met. Earlier initial HS is the most important outcome of this project. Earlier screening creates more time between HS and NICU discharge. The benefit of extra time allows for repeat assessment and adequate referral set up as needed. The extra time also allows for the infant's caregiver(s) to process a new diagnosis,

begin education regarding hearing loss, and further develop a relationship with the audiologist/audiology team, hopefully reducing loss to follow-up post discharge. These benefits may directly and/or indirectly translate into improved linguistic and developmental outcomes. Longitudinal studies have shown that timely referral to early intervention systems improves spoken and signed language development of deaf/hard of hearing newborns (Yoshinaga-Itano, 2014).

False positive rates were found to be increased in the post-implementation group, especially for those born at less than 34 weeks ($p = 0.08$). Prior studies in full-term infants have shown that the false positive rate of initial HS was 3.9% and repeated HS prior to discharge decreased the false positive rate to less than 1% (Clemens & Davis, 2001). The initial HS false positive was 6.3% among preterm neonates. More preterm neonates required repeat HS prior to discharge and passed the test subsequently. The common reason of the failed initial screening may be contributed to middle ear issues (Clemens et al., 2000). Middle ear effusion is even more prevalent in NICU patients, as they tend to be in the supine position for long periods of time. They may be receiving nasogastric tube feedings and/or humidified respiratory support. Another reason for our higher rate of false positive results may be due to neonatal prematurity. The peripheral hearing system matures with gestational age (Pujol et al., 1991). There is a risk of introducing false results by performing HS earlier in gestational age. Prior studies have shown that the hearing threshold decreased with increased gestational age postnatally (Pujol et al., 1991). The hearing threshold of extreme preterm neonates decreased from 28 dB at 28 weeks corrected GA to 13 dB at 42 weeks corrected GA, a rate of 1 dB/week (Jiang et al., 2015). When initial HS is performed earlier in GA, it becomes more likely that a preterm neonate may fail. Van Straaten and colleagues (2001) have shown that AABR screening can be reliably performed at 32 to 34 weeks corrected gestational age with a threshold setting of 35 dB, as adapted by our unit protocol. Because the risk of false positive HS results exists with screening at an earlier gestational age, we chose to perform initial screening at 34 weeks corrected GA, as opposed to 32 weeks corrected GA, in hopes of reducing the amount of false positive results.

A false positive HS result may increase parental anxiety and process costs; however a survey has shown no significant long-term or detrimental emotional impact on parents of infants with false positive HS (Clemens et al., 2000). Parental anxiety could be reduced with improved understanding regarding the infant hearing screening process (Clemens et al., 2000). We feel the benefit of earlier screening likely outweighs the risk of false positive HS as it allows the audiologist more time to properly support parents, repeat HS prior to discharge, and coordinate referral/offer interventions as needed (Clemens et al., 2000).

Implementation success was attributed to the providers' participation and education provided. Similar to prior literature, timing of initial HS greatly improved after

implementing an updated HS process and educating providers to identify patients eligible for early screening (Patel et al., 2018). The HS readiness assessment tool also improved workflow of the audiologists' by alerting them to eligible neonates, allowing the team to better prioritize NICU neonates for HS. The cost of this project was minimal; most of that cost surrounding the project was attributed to creating time for staff education.

Limitations

Major barriers in implementing an updated HS guideline were communication and education. Specifically, there was a lack of understanding that AABR screening can be reliably performed at approximately 32 to 34 weeks corrected gestation (Van Straaten et al., 2001). Additionally, the HS readiness assessment tool was unable to be integrated into the electronic health record in a timely manner due to constraints associated with the COVID-19 pandemic. A paper screening tool was developed as an alternative, but use was poor. These barriers were addressed with education regarding the new HS guideline via PowerPoint presentation and weekly educational reminders provided to promote continued awareness of the new protocol. Providers were verbally or electronically reminded via email to use the HS readiness assessment tool on a weekly basis. Despite consistent reminders for using the paper tool, the usage rate remained low. We speculate that our improved outcomes were due to verbal communication and education rather than the paper tool usage. Integration of an electronic version of the HS assessment tool would likely increase use and decrease dependence on the project team leader's verbal reminder for long-term sustainability.

Conclusion

Timely identification of hearing loss in NICU patients is important to improve long-term neurodevelopmental outcomes. The project was likely the first to itemize HS eligibility. The HS readiness assessment tool improved timeliness of initial HS in the NICU, particularly for the preterm population. The project would likely be more sustainable by integrating the HS assessment tool into the electronic medical record system. Although NICU HS readiness guidelines may benefit preterm neonates, further study is needed to optimize HS techniques to lower false positive screens.

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HEARING SCREENING ASSESSMENT TOOL

GESTATIONAL AGE

+ Is the infant as least 34 weeks corrected gestational age? YES ☐ NO ☐

It is not recommended to conduct newborn hearing screening before the infant is 34 weeks gestational age related to immaturity of the auditory nervous system. It is appropriate to evaluate hearing screening readiness at 34 weeks gestational age.

THERMOREGULATION

+ Is the infant thermodynamically stable? YES ☐ NO ☐

Newborn hearing screening should not be conducted on infants requiring an incubator or radiant heat to maintain body temperature. Once an infant has proved to be thermodynamically stable in an open crib, it is appropriate to evaluate readiness for newborn hearing screening.

RESPIRATORY SUPPORT

+ Is the infant requiring minimal or no oxygen therapy? YES ☐ NO ☐

Infants placed on ventilators, CPAP, or high-flow nasal cannula are not candidates for hearing screen, these respiratory modalities may interfere with the screening process. It is appropriate to screen infants who are stable on regular nasal cannula, low-flow nasal cannula, or not requiring oxygen therapy.

LINES and/or DRAINS

+ Does the infant have any critical lines and/or drains? YES ☐ NO ☐

Infants requiring critical lines or drains for advanced medical support are not considered stable. Examples of critical lines include: umbilical catheters, chest tubes, gastric decompression tubes, etc. If an infant requires a surgical procedure, screening should be performed post-operatively, once the infant is medically stable. However, it is appropriate to screen infants requiring gastric tube placement prior to surgery.

NUTRITION

+ Has the infant reached full enteral feeds? YES ☐ NO ☐

Infants requiring parenteral nutrition are not candidates for screening in the UUMC NICU. Infants should be receiving full enteral feeds to be considered for a newborn hearing screening. It is acceptable to conduct newborn hearing screening on infants receiving feeds via nasogastric tube. It is also acceptable to conduct screening on infants with stable gastric tubes.



MEDICATIONS

+ Is the infant receiving ototoxic medications? YES ☐ NO ☐

Newborn hearing screening should be deferred for infants requiring aminoglycoside administration for more than 5 days. It is appropriate to evaluate hearing screening readiness after the 5-day course has been completed. If an infant does not require a 5-day course of aminoglycoside administration, hearing screening readiness can be evaluated as infant condition warrants. If questions arise regarding the ototoxic potential of other medications an infant may be receiving, consult pharmacy and audiology.

+ Is the infant being treated for Neonatal Abstinence Syndrome (NAS)? YES ☐ NO ☐

Neonatal abstinence syndrome can cause central nervous system hyperirritability, which may interfere with the hearing screening process. Infants being treated for NAS should not be considered candidates for newborn hearing screening during a period of severe withdrawal. It is appropriate to screen NAS infants once Neonatal Withdrawal Index (NWI) scores are trending down, and the infant is consolable.

PHYSIOLOGICAL STABILITY

+ Does the infant tolerate assessment/cares? YES ☐ NO ☐

Any baby who decompensates with care should not be considered stable. Decompensation can be defined as, but not limited to the following: apnea, bradycardia, oxygen desaturations, tachypnea, and tachycardia. It is appropriate to evaluate hearing screening readiness on infants who tolerate assessment without experiencing physiological instability.

AS SOON AS YOU FEEL AN INFANT MEETS THE ABOVE REQUIREMENTS, PLEASE PLACE A PATIENT LABEL ON THE FRONT OF THIS SHEET AND RETURN THIS SCREENING TOOL TO AUDIOLOGY VIA THE DESIGNATED BIN LOCATED IN THE NNP OFFICE. AUDIOLOGY WILL PERIODICALLY COLLECT THESE ASSESSMENTS AND BEGIN EVALUATIONS.

If you have any questions or concerns regarding this tool please contact McKenzie Blatt, NNP or Adrienne Johnson, AuD.

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Adverse Childhood Experiences in Mothers and Their Children with Hearing Loss

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Abstract

This study examined the relationship between maternal and child adverse childhood experiences (ACEs) in children with hearing loss ages 3–12 years old. Mother and child ACE assessments were completed by 124 mother-child dyads. ACEs were measured using the Center for Youth Wellness Adverse Childhood Experiences Questionnaire (CYW ACE-Q; Burke Harris & Renschler, 2015).

Both maternal and child participants in this study reported higher levels of ACE exposure than previously reported in studies of the general population. Maternal and child ACEs were significantly correlated. White/Caucasian mothers experienced significantly fewer ACEs than mothers of other races/ethnicities. Children living in adoptive, foster, or guardianship placements experienced significantly more ACEs than children living with their biological mothers. The results of this study suggest that maternal and child ACEs are significantly correlated in children with hearing loss and their mothers, as has been found in literature on hearing mother-child dyads.

Keywords: children who are deaf and hard of hearing, hearing loss, trauma-informed care, adverse childhood experiences

Acronyms: ACE = adverse childhood experience; CYW ACE-Q = Center for Youth Wellness Adverse Childhood Experiences Questionnaire; PTA = pure tone average

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Parent-child relationships are the foundation from which all learning begins (Ainsworth, 1979; Bowlby, 1988). When infants' early communicative attempts are met with attentive, consistent, emotionally responsive caregiving, their brains have the opportunity to establish the foundations of pre-linguistic development. Higher levels of maternal responsiveness have been associated with improved language outcomes for both children with typical hearing (Spencer & Meadow-Orlans, 1996; Yoder & Warren, 1999) and children with hearing loss (Quittner et al., 2013). Maternal responsiveness and parent-child relationships, in general, are supportive factors for the development of language skills (Bee et al., 1982; Mistry et al., 2010). In contrast, exposure to adverse childhood experiences (ACEs), such as child abuse, neglect, maltreatment, inconsistent caregiving, poverty and so on is a risk factor for child development (Felitti et al., 1998).

This study used self- and parent-report measures of maternal and child ACE exposure to investigate the relationships between maternal and child adverse experiences in mother-child dyads of children with hearing loss. Children ages three to twelve years old, with any degree of permanent, bilateral hearing loss, were studied in mother-child dyads.

Background

Children with Hearing Loss

For the purposes of this study, *children with hearing loss* are considered to be any children with permanent, bilateral hearing loss of any type (sensorineural, conductive, or

mixed) or degree (mild, moderate, severe, or profound) as determined by the child's most recent audiological diagnosis. In the literature, this population is referred to with various terms: deaf, Deaf, hard of hearing, hearing loss, and so on. The current study investigated hearing parents and their deaf children who have elected to use listening and spoken language for communication. The children studied have varying degrees of hearing loss, and are as of yet too young (ages 3 to 12 years old) to articulate a preference for a cultural versus medical model of identification for their deafness. Thus, the term *children with hearing loss* will be used throughout to discuss this group as a whole.

Adverse Childhood Experiences (ACEs)

Adverse childhood experiences, or ACEs, refer to a diverse set of potentially traumatic events in childhood that may be unfavorable to health and development across the lifespan (Shonkoff & Garner, 2012). Although various ACE scales exist, most include questions regarding the child's/family's economic security (ability to consistently secure adequate food and shelter), physical abuse (directed toward and/or witnessed by the child), sexual abuse, mental health of the child's primary caregiver(s), and presence/absence of positive, supportive adult figures. Expanded ACE questionnaires, such as the Center for Youth Wellness Adverse Childhood Experiences Questionnaire - Child (CYW ACE-Q Child) used in this study, also include questions about neighborhood violence, immigration, and school bullying/harassment (Burke Harris & Renschler, 2015).

Literature Review

Theoretical Framework

Ecological Systems Theory

This study investigates language development in children with hearing loss from the perspective of the *ecological systems theory* (also called human ecology or development in context; Bronfenbrenner, 1979). Ecological systems theory views human development as the product of interaction between the individual and her environment, both micro (family systems) and macro (broad societal and cultural factors). As such, language development for a child with hearing loss cannot be seen as a function of the child's audiological status, age of identification, etiology, or other personal factors alone, but must instead be viewed in the context of that child's interaction with her family, culture, and society. This theory posits that "what matters for behavior and development is the environment as it is *perceived* rather than as it may exist in an 'objective' reality" (Bronfenbrenner, 1979, p. 4), lending credence to the use of participant self-report measures (i.e., the *Parent Child Relationship Inventory* and *Adverse Childhood Experiences Questionnaire* used in this study). Likewise, while parent-child dyads were assessed in this study, items on the assessment material probed the dyad's larger ecological context, as Bronfenbrenner (1979) noted that

The capacity of a dyad to serve as an effective context for human development is crucially dependent on the presence and participation of third parties, such as spouses, relatives, friends, and neighbors. If such third parties are absent, or if they play a disruptive rather than a supportive role, the developmental process, considered as a system, breaks down; like a three-legged stool, it is more easily broken if one leg is broken, or shorter than the others. (p. 5)

Bonfenbrenner is credited with shifting the field of child development from a focus on assessing the construct of *attachment* in single-instance *strange situation tasks* in which a child's reaction to separation from a caregiver in a new environment is evaluated (Ainsworth & Bell, 1970) to an appreciation of "contextual variation in human development" (Darling, 2007, p. 203).

Seligman and Benjamin Darling (2007) expand on this framework in the specific context of families with children with disabilities, noting the interconnected nature of all aspects of the family and social system, stating, "each variable in any system interacts with the other variables so thoroughly that cause and effect cannot be separated" (p. 17). Algood et al. (2011) further explored childhood disability through the perspective of ecological systems theory by noting that the presence of protective factors in the child's social circles can shield children with disabilities from maltreatment. For example, early intervention focused on promoting responsive caregiver-child interaction may serve as a barrier against maltreatment by improving the functioning of the family system. For children with disabilities, and all children, development is most holistically viewed in the context of their immediate family relationships as well as broader environmental and sociological phenomena.

Adverse Childhood Experiences

Parental ACEs

Research has demonstrated that parents who exhibit higher levels of adverse experiences during their own childhoods are more likely to exhibit difficulties in relationships with their own children, which often

has a cascading effect on child mental health and childhood behavior problems, both internalizing and externalizing (Steepleton et al., 2018). A study of Head Start participants and their mothers (Randall et al., 2015) found that parental ACE exposure was highly correlated with children's experiences of adversity, noting that, "there was a strong positive association between parental ACE and childhood adversity" and that "this association was strongest among parents with an ACE score of 4 or more, indicating a dose-response relationship" (p. 786).

In addition to the correlation between parent and child ACE scores, parental ACEs have also been associated with deleterious effects on child development, starting even before birth. Mothers with higher ACE scores were more likely to consume alcohol during pregnancy (Frankenberger et al., 2015). Parental mental health has been implicated in negative social and academic outcomes for children (e.g., grade retention, internalizing and externalizing behavior disorders), independent from the children's own ACE scores (Porche et al., 2016). Folger et al. (2018) found that "for each additional maternal ACE, there was an 18% increase in the risk for a suspected developmental delay" (p. 4) among a sample of two-year-old children (311 mother-child dyads and 122 father-child dyads). In addition to the risk of developmental delay, children of parents with higher ACE scores are at increased risk for receiving a diagnosis of Attention Deficit Hyperactivity Disorder (ADHD) and/or behavioral disorders (Schickedanz et al., 2018).

Mothers' ACE scores affect their own parenting practices. Mothers who reported experiencing physical abuse in their own childhoods were significantly more likely to use corporal punishment and spanking as means of discipline with their own children (Chung et al., 2009). In their study of mothers without psychological diagnoses, mothers with depression, and mothers with depression and/or Post-Traumatic Stress Disorder (PTSD), Chemtob et al. (2013) found that mothers with a PTSD diagnosis demonstrated greater physical aggression toward their children and had children who were more likely to be exposed to traumatic experiences themselves.

Factors related to parental/maternal ACE scores have been implicated in child language development. Mothers with depression have been shown to provide poorer-quality caregiving to their offspring, which affects child language when measured at 36 months of age in children with typical hearing (Stein et al., 2008). Likewise, Paulson et al. (2009) found that parental mental health influences the amount of parent-child reading time, another key component in language development. In a factor analysis of mother-child relationships to describe factors linked to child language delay, Sylvestre and Mérette (2010) identified child cognitive development, the mother's history of her own physical and emotional childhood abuse, and mothers' responsiveness to their children as the strongest predictors of language disorders in children who have been neglected. Torrisi et al. (2018) linked maternal exposure to interpersonal violence and maternal post-traumatic stress disorder to maternal caregiving behavior and found a relationship between this caregiving behavior and child language outcomes. Thus, maternal ACE scores affect not only the mother, but also her children's health, growth, and development.

Childhood ACEs

Although ACE questionnaires may be used retrospectively for adults reflecting on events that occurred prior to their eighteenth birthdays, evaluations of pediatric ACEs assess any potentially traumatic life events children have experienced or are currently experiencing. In their

meta-analysis of 241 publications and 551 prevalence rates for child abuse worldwide, Stoltenborgh et al. (2015) found that, although reports of child sexual abuse was most prevalent in the literature, overall rates of child abuse of all types, including neglect and emotional abuse, were quite high worldwide, and concluded, “child maltreatment is a widespread, global phenomenon affecting the lives of millions of children all over the world” (p. 37). In the United States specifically, 7.4 million children were reported to Child Protective Services nationally and 1,750 abuse-related child fatalities were recorded in the year 2016, the last year for which data is available (U.S. Department of Health & Human Services, 2018). Additional children experienced non-abuse related ACEs, such as the death of a caregiver, a serious or chronic illness, or neighborhood violence or discrimination (van der Kolk, 2005).

Another factor assessed in ACE questionnaires is whether or not the child/family have experienced markers of poverty, such as homelessness, food insecurity, or use of other social benefits (e.g., welfare, food stamps). Material deprivation in childhood is tied to a variety of deleterious effects. Particularly germane to this study are the effects of childhood poverty on cognition and language for children with and without hearing loss. Numerous researchers have found that socioeconomic status predicted both the size and rate of growth of two year olds’ vocabularies, with children of high socioeconomic status achieving higher scores, likely due to both quality and quantity of child-directed speech from their mothers (Hart & Risley, 1992; Hoff-Ginsberg, 1998; Pungello et al., 2009; Raviv et al., 2004). Similar findings have been reported linking socioeconomic status with language outcomes in children with hearing loss (Niparko et al., 2010).

Exposure to adverse childhood experiences also predisposes children to respond disproportionately to later stressful but non-traumatic events of daily life, leading to higher incidences of both internalizing and externalizing behavior disorder symptomatology (Grasso et al., 2013). Perry et al. (1995) describe this as the transition from “states” to “traits,” that is, while hyperarousal and dissociation are natural responses to a stressor, children living in environments that are chronically stressful and traumatic may remain in such states to the point that it is maladaptive for learning and retaining new information as well as forming healthy relationships. Sheridan and McLaughlin (2016) argue that these adverse experiences (e.g., decreased cognitive enrichment, increased exposure to violence) affect children’s brains through the neurobiological process of neuroplasticity, influencing the development of both emotional control and cognitive control in ways that prove disadvantageous for later academic success.

These difficulties in developing relationships, executive function skills, and emotional reciprocity affect child language development, a principal concern in this study. Children with a history of abuse and neglect score significantly worse than their non-affected peers on measures of language and social development (Cobos-Cali et al., 2018; Lum et al., 2018). Even when controlling for other aspects that are known to affect cognitive development (i.e., demographic and socioeconomic factors, birth weight, maternal IQ, and the amount of stimulation in the home), exposure to interpersonal trauma has a significant, independent effect on children’s IQ scores. Children exposed to trauma in the first two years of life, on average, score half a standard deviation lower than their non-trauma-exposed peers (Enlow et al., 2012). For children who experience childhood maltreatment and do not receive therapeutic intervention, studies note a significant delay in language scores when compared to

both peers who have not experienced ACEs and peers who have experienced ACEs but received intervention (Culp et al., 1987). Allen and Wasserman (1985) hypothesized that the delays in language observed among children who have been abused may be tied to mothers’ ignoring behaviors and lack of cognitive stimulation.

Although the literature has established that exposure to adverse childhood experiences has undeniably negative effects on child development across domains (and throughout the lifespan), any discussion of trauma would be incomplete without a recognition of the amazing capacity of humans to develop resilience in the face of difficulty, and the factors that contribute to children’s resilience in the face of trauma. In their discussion of five modifiable factors to promote resilience in the face of childhood adversity, Traub and Boynton-Jarrett (2017) identify parenting and the treatment of maternal mental health issues as keys to improving child outcomes, supporting the present investigation’s inclusion of maternal ACE scores in our analysis. Masten et al. (1990) identified several characteristics that predispose a child to recover more successfully from adverse childhood experiences, including, most notably for the purposes of this study, relationships with competent adults and the ability to engage with other people (strongly tied to language skills).

ACEs in Populations of Individuals with Hearing Loss

Minimal research has been conducted investigating the effects of adverse childhood experiences for the population of people with hearing loss. Some research (e.g., Kushalnagar et al., 2020) probes the self-reports of adults who are deaf or hard of hearing of “adverse childhood communication experiences” and links this to poorer adult health outcomes but does not strictly measure ACE exposure as broadly defined in the psychomedical literature. Kvam and Loeb (2010) reported an association between self-reports of childhood adverse experiences and current mental health problems for Norwegian adults who were deaf. Although there are no prior investigations of ACEs among children with hearing loss, it is hypothesized that higher rates of adverse childhood experiences or disruptions in attachment have cascading effects on higher-order neurodevelopment. For the purposes of this study, language development is investigated as a proxy marker of higher-order neurodevelopment, as it is a historical area of weakness of children with hearing loss when compared with their hearing peers, even when matched by age and socioeconomic status (Tomblin et al., 2015).

Purpose of the Study

Although advances in hearing technology, early intervention, and special education law have greatly improved language, academic, and social outcomes for children with hearing loss, there remains a subset of children who are deaf or hard of hearing whose listening, speech, and language outcomes lag behind their peers—both those with hearing loss and without (Ching, 2015; Moeller, 2000; Moeller & Tomblin, 2015; Yoshinaga-Itano et al., 1998). In addition to access to assistive technology and intervention, best practices in family-centered early intervention for children with hearing loss and their families recognize the importance of comprehensive social-emotional support and attention to environmental and relational factors as critical ingredients in achieving desired language outcomes (Moeller et al., 2013). Thus, although there are many plausible explanations for this phenomenon that relate directly to hearing loss (e.g., late identification, abnormal cochlear etiology, lack of highly qualified service providers), another source of this variation in outcomes may be found in more general factors, such as ACE exposure and parental self-efficacy.

Significant bodies of research exist on both the effects of parenting, parent-child interactions, and language outcomes for children both with and without hearing loss (National Institute of Child Health, 2000; Quittner et al., 2010; Quittner et al., 2013) and the negative developmental effects of ACEs in general (Jimenez et al., 2016) yet there is relatively little research examining the relationship between maternal and child ACEs for children with hearing loss. This population is at an increased risk for childhood maltreatment (Schenkel et al., 2014). De Bellis (2001) proposed that, “the potential psychobiological sequelae of child maltreatment may be regarded as an *environmentally induced complex developmental disorder*” (p. 540). What, then, are the compounding effects of this “environmentally induced complex developmental disorders” on children who already have a diagnosis of another communication disorder—hearing loss—and how might maternal trauma history affect children’s experiences?

Although previous studies have documented abuse and maltreatment of children and young adults who are deaf or hard of hearing (e.g., Titus, 2010), there are no prior investigations of ACEs in young children with hearing loss. The current study aims to integrate current knowledge on maternal-child relationships and language outcomes with the field’s emerging focus on ACEs to investigate the relationship of both of these potential risk factors for the population of children with hearing loss who use spoken language. Voss and Lenihan (2016) note that although professionals serving children with hearing loss often serve families in poverty or other adverse circumstances, personnel preparation programs rarely include adequate (if any) instruction in working with families who have experienced trauma. These programs focus more on the technical and educational aspects of language development for children with hearing loss. Instruction regarding the role of ACEs and parent-child attachment is lacking in both preprofessional preparation for students of speech-language pathology, audiology, and deaf education, and professional development for practitioners and educators in the field. Finally, the link between social-emotional risk and protective factors and language outcomes is under-investigated (Voss & Lenihan, 2016). Ko et al. (2008) concur, noting that professionals in the healthcare and education systems receive little to no training on how to create trauma-informed practices to best serve children and families who have experienced ACEs, stress, or disruptions in the parent-child relationship. The results of this study have the potential to inform clinical practice by encouraging professionals to expand their clinical focus to include children’s and families’ interaction patterns and overall psychosocial wellbeing in addition to speech, language, and listening targets by demonstrating the interrelated nature of these constructs.

This study sought to investigate the correlation between maternal and child ACE exposure in children with hearing loss and the demographic factors that influenced ACE exposure in this population. Both general variables (e.g., age, race, education, income) found to affect ACE exposure in the literature more broadly as well as deafness-specific factors (e.g., degree of hearing loss, communication mode, and language) were selected to probe the ways in which this population is similar to, or different from, previous data on maternal-child ACE correlations in dyads with typical hearing.

Method

Participants

In this investigation, children ages three to twelve years of age with hearing loss (defined as children with any degree or type of permanent, bilateral hearing loss as

determined by their most recent audiological diagnosis) and their mothers were studied to assess the relationship between maternal and child ACE scores. Participants were biological, adoptive, or foster/guardian mother-child pairs. Although some participants reported using languages other than English at home, all mothers in the study were able to complete study forms and assessments in English. Participants were recruited via professional listservs, conferences, and social media from cochlear implant centers, audiological clinics, educational programs for children with hearing loss, and private therapy centers across North America. Participants hailed from a wide range of rural, suburban, and urban locales across North America. Following approval by the Institutional Review Board of Teachers College, Columbia University; 124 mother-child dyads consented to participate in the study. The sample size was selected in line with best practices as established by the Council on Exceptional Children for experimental group design studies in special education (Gersten et al., 2005). Maternal and child participant demographics are presented in Table 1 and Table 2, respectively.

Children’s Hearing Loss and Hearing Technology

Mothers reported their child’s degree of hearing loss based on the child’s Pure Tone Average (PTA; calculated as the average of the child’s unaided hearing thresholds at 500, 1000, and 2000Hz, respectively). Slightly under half of the children in this study (57 [46.0%]) had profound hearing loss (PTA of 90 dB or greater). The most common age at identification of hearing loss was at birth (60 [48.4%]) for children in this study. Children’s hearing technology use was reported for their left and right ears, respectively. Children in the study reported use of cochlear implants, bone conduction devices, and hearing aids, and some children reported non-use of hearing technology for one or both affected ears.

Children’s Intervention and Communication Characteristics

The majority of the children used listening and spoken English as their primary communication mode (89 [71.8%]). Twenty-three (18.5%) children were reported to use total communication, seven (5.6%) used American Sign Language or another visual language, and five children (4.0%) were reported to use a spoken language or languages other than English as their primary mode of communication. Other spoken languages used by the children in this sample included Portuguese, Danish, French, German, Hebrew, Hungarian, Italian, Romanian, Russian, and Spanish. Children in this study were enrolled in a range of intervention methods and settings, including individual, family-centered, outpatient Auditory Verbal Therapy, speech-language therapy services, group/center-based educational programs for children with hearing loss, home-based early intervention, and/or instruction in visual communication (e.g., sign language(s) or cued speech).

Assessments

Demographic Questionnaire

To collect information on the demographic variables analyzed in this study, mothers were asked to complete a demographic questionnaire about themselves and their children.

Center for Youth Wellness Adverse Childhood Experiences Questionnaire (CYW ACE-Q)

ACE scores were collected, with parents serving as the reporters, for both the parents’ own childhood experiences and their children’s, using the Center for Youth Wellness Adverse Childhood Experiences Questionnaire (CYW

Table 1
Demographic Characteristics of Maternal Participants

<i>Characteristic</i>	<i>Mothers</i>	
	<i>n</i>	<i>%</i>
Hearing status		
Hearing	119	96
Deaf/hard of hearing	5	4
Native language		
English	112	90.3
Portuguese	2	1.6
Romanian	2	1.6
Italian	2	1.6
Kannada	1	.8
Spanish	1	.8
Hungarian	1	.8
Filipino	1	.8
Hungarian	1	.8
Iceland	1	.8
Age		
Under 29 years old	4	3.2
30-39 years old	72	58.1
Over 40 years old	48	38.7
Race/ethnicity		
White/Caucasian	105	84.7
Asian	6	4.8
Black/African American	5	4.0
Hispanic/Latino/a	5	4.0
Multiracial	2	1.6
Other	1	.8
Highest level of education		
High school diploma	13	10.2
Bachelor's degree	56	45.2
Master's, professional, or doctoral degree	55	44.6
Family income (yearly, in USD)		
Under \$40,000	6	4.8
\$40,000-49,000	6	4.8
\$50,000-59,000	9	7.3
\$60,000-69,000	6	4.8
\$70,000-79,000	10	8.1
\$80,000-89,000	8	6.5
\$90,000-99,000	14	11.3
Over \$100,000	65	52.4

Table 2
Demographic Characteristics of Child Participants

<i>Characteristic</i>	<i>Children</i>	
	<i>n</i>	<i>%</i>
Gender		
Female	71	57.3
Male	53	42.7
Race/ethnicity		
White/Caucasian	92	74.2
Asian	10	8.1
Black/African American	5	4.0
Hispanic/Latino/a	6	4.8
Multiracial	8	6.5
Other	3	2.4
Family status		
Biological	111	89.5
Adoptive/foster	13	10.5
Additional disabilities*		
No	84	67.7
Yes	40	32.3
Pure Tone Average (PTA)		
Mild	7	5.6
Moderate	15	12.1
Severe	10	8.1
Profound	57	46.0
Unreported	19	15.3
Primary mode of communication		
Spoken English	89	71.8
Total communication	23	18.5
Visual communication (American Sign Language or other signed language)	7	5.6
Spoken language other than English	5	4.0

*Additional disabilities reported included: Ehlers Danlos Syndrome, failure to thrive, SLC6A1 genetic mutation, epilepsy, global developmental delay, Post-Traumatic Stress Disorder, apraxia of speech, kidney malformations, vertebral malformation, hyperparathyroidism, congenital cytomegalovirus, Attention Deficit Hyperactivity Disorder, Usher Syndrome, Binder Syndrome, Disruptive Mood Dysregulation Disorder, skeletal disorders, 16p13.3 deletion, Generalized Anxiety Disorder, Sensory Processing Disorder, speech and language impairment, dyslexia, microtia/atresia, Duane Syndrome, cardiac malformations, prematurity, low muscle tone, craniofacial disorders, anxiety, mitochondrial DNA mutation, Pendred Syndrome, neurogenic bladder, asthma, Auditory Neuropathy Spectrum Disorder, vision impairment, cleft palate, feeding and digestive issues, Von Willebrand's Disease, Ring 13 chromosomal disorder, microcephaly, and Dandy Walker Syndrome.

two groups of items for research purposes. For scoring purposes, the total score is used (Bucci et al., 2015). A total score of greater than or equal to four ACEs endorsed indicates clinically significant exposure to adverse childhood experiences, as does a score of 1 to 3 with symptomatology (Bucci et al., 2015). For the purposes of this study, ACE scores of 0 to 3 were classed as *low ACEs* and ACE scores greater than or equal to four were put in the *high ACEs* category. No formal assessments of psychopathology were conducted as part of this study, and symptomatology was not considered as a factor in classifying participants into the low or high ACE groups.

In this study, we investigated both maternal and child ACE scores. Mothers were asked to complete the ACE-Q twice,

ACE-Q; Burke Harris & Renschler, 2015). The ACE-Q has versions for children, teens, and adults/parents. Each questionnaire asks respondents to indicate the number of adverse childhood events they have experienced, though respondents are not asked to reveal the exact ACEs they have experienced. ACEs in the questionnaire include things such as, "At any point since your child was born...your child's parents or guardians were separated or divorced," "...your child lived with someone who had a problem with drinking or using drugs," and "...your child often saw or heard violence in the neighborhood or in her/his school neighborhood." Items are categorized into two groups. The first covers the original 10 ACEs identified by the seminal Adverse Childhood Experiences study from the Kaiser Permanente health system (Felitti et al., 1998) and the second includes additional "early life stressors" (Bucci et al., 2015, p. 10). The instrument takes respondents between two and five minutes to complete and yields a numerical score for each of the

once for themselves and once on behalf of their child. When inquiring about mothers' own adverse experiences, the original ACE-Q Child assessment was modified by the researcher, changing language from "At any point since your child was born..." to "At any point before your eighteenth birthday..." for mothers to report on their own experiences. Both mothers' and children's total ACE scores were grouped into low ACEs (0-3 total score) and high ACEs (total score greater than or equal to 4) for the purposes of categorical analysis.

A copy of the CYW ACE-Q is included as Appendix A. This study's adaptation of the CYW ACE-Q for maternal participants is included as Appendix B.

Results

Preliminary Data Analysis

Primary data analysis was conducted using IBM SPSS version 26 for Mac, with Alpha for all significance tests set at $p < .05$ (two-tailed). All analyses were conducted by the primary investigator and reviewed by two additional colleagues in the field of Education for the Deaf or Hard of Hearing with PhDs and at least three years' experience with advanced statistics courses.

Center for Youth Wellness Adverse Childhood Experiences Questionnaire (CYW ACE-Q)

All mothers who participated in this study completed the CYW ACE-Q, reporting on the number of adverse childhood experiences (ACEs) that they had experienced before the age of eighteen and the number of ACEs their child had experienced in his/her lifetime, respectively. Both mothers and children reported ACE scores ranging from 0 to 13, though mothers had a higher average ACE score ($M = 2.63$, $SD = 2.89$) than children ($M = 1.66$, $SD = 2.27$). The Center for Youth Wellness classifies ACE scores of greater than or equal to four as High ACEs. In this sample, 28.2% of mothers ($n = 35$) and 15.3% of children ($n = 19$) had ACE scores of four or greater.

A one-way ANOVA was conducted to investigate the relationship between demographic characteristics and maternal and child ACE scores. Results are presented in Table 3 and Table 4, respectively.

With regard to ACE exposure, maternal ACE scores differed significantly by participant race ($F[5, 118] = 2.300$, $p = .049$), though post-hoc analyses to determine specific differences between categories could not be conducted because the categories of *Multiracial* and *Other* had two or fewer participants. Maternal education level was significantly associated with maternal ACE exposure ($F[2, 121] = 3.523$, $p = .032$). A post-hoc Tukey test indicated that there was an increase in ACE exposure among mothers reporting that they had attained a high school diploma ($M = 4.54$, $SD = 4.719$) when

Table 3
Relationship Between Demographic Characteristics and Maternal Adverse Childhood Experience (ACE) Scores

Demographic variable	<i>df</i>	<i>F</i>	<i>p</i>
Household income	116	1.799	.098
Race	118	2.300	.049*
Education level	121	3.532	.032*

* $p < .001$

Table 4

Relationship Between Demographic Characteristics and Child Adverse Childhood Experience (ACE) Scores

Demographic variable	<i>df</i>	<i>F</i>	<i>p</i>
Adoptive/foster placement	122	-.506	.001*
Maternal education level	121	1.775	.174
Household income	116	1.688	.119
Maternal race/ethnicity	118	1.635	.156
Child race/ethnicity	118	.919	.471
Maternal age	121	.422	.657

* $p < .001$

compared to mothers reporting that they had attained a graduate or professional degree ($M = 2.25$, $SD = 2.374$), a mean increase of 2.284, 95% CI [.23, 4.33], which was statistically significant ($p = .025$). Maternal age was significantly associated with maternal ACE exposure in this sample ($F[2, 121] = 3.881$, $p = .023$), with mothers over the age of 40 ($M = 1.77$, $SD = 2.065$) differing from mothers between the ages of 30 to 39 years ($M = 3.13$, $SD = 3.117$) reporting a mean increase of 1.354 ACEs, 95% CI [.12, 2.59], $p = .028$. Household income did not significantly predict maternal ACE exposure in this sample ($F[7, 116] = 1.779$, $p = .098$).

Children's ACE scores did not differ significantly by child race/ethnicity ($F[5, 118] = .919$, $p = .471$), mother's age ($F[2, 121] = .422$, $p = .657$), maternal education level ($F[2, 121] = 1.775$, $p = 1.74$), or household income ($F[7, 116] = 1.688$, $p = .119$). A Pearson correlation between child age and ACE exposure yielded nonsignificant results $r(122) = .132$, $p = .143$. Children who had been adopted or were living in foster/guardianship placements had significantly higher ACE exposure ($M = 5.00$, $SD = 3.536$) than children living with their biological parents ($M = 1.27$, $SD = 1.705$), $t(122) = -6.483$, $p < .001$. No confounding variables were identified in the analyses of either maternal or child ACEs in this study.

Discussion

In this study, maternal and child ACE scores were found to be significantly associated. This is consistent with data on maternal and child ACE associations in mother-child dyads of children with typical hearing (Randall et al., 2015) but had not yet been substantiated in the literature on mothers and their children with hearing loss. The fact that mothers' ACE exposure significantly predicts ACE exposure in children with hearing loss is a novel finding of this investigation.

Adverse Childhood Experiences (ACEs)

Overall, the participants in this study reported levels of ACE exposure greater than previous research has indicated are prevalent in the general population. In their groundbreaking and wide-ranging ACE investigation with nearly 10,000 participants, Felitti et al. (1998) found that 52% of the adults surveyed reported having experienced at least one ACE, and 6.2% reported four or more. Later studies, such as Merrick et al. (2019) found that 15.6% of adults surveyed

reported four or more ACEs. Among children, Ager (2013) reported that 48% of children in the United States report at least one ACE, and Bethell et al. (2014) found that 22.6% of children in the United States have been exposed to two ACEs or more. In the present investigation, 28.2% of mothers (and 15.3% of children) fell into the high ACEs (ACE exposure ≥ 4) category. The unusually elevated ACE exposure in this sample may be due to several factors. The ACE questionnaire used in Felitti et al.'s (1998) original study included fewer ACEs than the CYW ACE-Q instrument used in the present investigation. Participant self-selection cannot be discounted. Perhaps mothers who felt strongly about the study topic of adverse childhood experiences were more likely to enroll and participate in this investigation. Likewise, an unusually high proportion of children in this study were reported to have been adopted or be living in foster/guardianship placements, placing them in a higher risk category for ACEs than children living with their biological mothers.

ACE exposure among mothers and children in this sample was significantly correlated, a finding in line with previous investigations. Randall et al. (2015) found a significant correlation between maternal and child ACE scores in dyads with hearing mothers and hearing children. This study substantiated that result among children with hearing loss and their mothers, the majority of whom reported typical hearing.

Mothers' race/ethnicity and level of formal education were found to be significantly associated with ACE exposure. In this sample, mothers who reported their race/ethnicity as White/Caucasian and mothers reporting higher levels of formal education had significantly lower levels of ACE exposure. Felitti et al. (1998) found that Asian participants were less likely to have high levels of ACE exposure, differing from the findings in the present investigation. However, other studies (Maguire-Jack et al., 2020; Vásquez et al., 2019) have had White/Caucasian participants report lower levels of ACE exposure, similar to this study. Felitti et al. (1998) found that participants who reported having attained a college degree were significantly less likely ($p < .001$) to report high levels of ACE exposure. Household income was not found to be predictive of maternal or child ACE exposure, a finding somewhat in line with the research of Halfon et al. (2017), which concluded that, although the proportion of children experiencing high ACEs increased as income decreased, "higher income was not necessarily found to be a protective factor against ACEs" (p. S70).

The only significant demographic factor measured in this study affecting child ACE exposure was the child's adoption status. Children living with adoptive or foster/guardian mothers reported significantly higher numbers of ACEs than peers living with their biological mothers. Other studies (Anthony et al., 2019; Turney & Wildeman, 2017) of children living in adoptive and foster/guardianship placements have similar findings.

Limitations

The demographics of the participants in this study differed from the population at large in several notable ways. Overall, the sample had a higher percentage of people who were white/Caucasian, had higher income levels, and had attained higher levels of formal education than the United States population as a whole. It is widely accepted that approximately 40% of children with hearing loss have additional disabilities (Holden-Pitt & Diaz, 1998; Picard, 2004), however the percentage of children with additional disabilities in this study (32%) is slightly lower. Likewise, both mothers and children in this study reported rates of ACE exposure greater than those found in previous

studies of the general population (Felitti et al., 1998). Self-selection effects among the participant pool cannot be discounted as a potentially significant source in the lack of association between independent variables and language outcomes in this study.

Family income, absence of additional disabilities, and higher levels of maternal education have been established in the literature to be strongly correlated with improved language outcomes for children with hearing loss (Calderon, 2000; Sarant et al., 2009; Yoshinaga-Itano et al., 2017). Other studies have demonstrated robust associations between early identification, early hearing technology use, and early intervention and child language outcomes (Fulcher et al., 2012; Holzinger et al., 2011; Tomblin et al., 2015). Results from this sample did not concur, likely due to the size of this sample and self-selection effects among participants, not the credibility of earlier studies on the topic.

Although self-report measures of past experiences, particularly those from an adult's recollections of childhood, may be subject to scrutiny, Hardt and Rutter's (2004) meta-analysis of adult reports of childhood trauma found that adults' retrospective recollections of childhood trauma were much more predisposed to false negatives than false positives (if anything, people tend to underreport childhood trauma), and concluded that, although there was inherent bias in self-reporting measures, "such bias is not sufficiently great to invalidate retrospective case-control studies of major adversities of an easily-defined kind" (p. 260). Similarly, in their assessment of the correlation between clinical interview and self-report of childhood traumatic experiences among adults, Bifulco et al. (2005) reported satisfactory reliability and validity when comparing the parallel interview and self-report instruments. Research has also confirmed the reliability of parent reports on children's behavior, mental health, and other ACE-related factors (Bishop et al., 2003; Nauta et al., 2004; Oh et al., 2018; Theunissen et al., 1998). An additional limitation is that the children in this study were not asked to complete their own ACE questionnaires, even though some of them would likely have possessed the language and literacy skills to do so, particularly if aided by an impartial third party. Thus, although parent reports are generally considered valid and reliable measures of children's health and behavior, an investigation of the potential differences between parent-reported and self-reported ACE scores in this population would be of interest for future investigations.

Future Directions

Given the dearth of research on ACE exposure in the population of children with hearing loss, this study produced a notable addition to the literature by establishing, in this sample, a significant correlation for maternal and child ACEs for children with hearing loss, which is in line with findings on hearing mother-child dyads (Randall et al., 2015). The connection between maternal and child ACE exposure in this population suggests that ACE exposure should be viewed as a relevant aspect of children's and families' case histories upon enrollment in intervention services for childhood hearing loss. Given that maternal ACEs have the potential to negatively affect children's physical and psychosocial development (Racine et al., 2018), and children's ACE exposure has been linked with negative academic and developmental outcomes (Blodgett & Lanigan, 2018), professionals in the field of hearing loss would be wise to consider ACEs as another risk factor among those more commonly assessed during intake (e.g., premature birth, exposure to ototoxic drugs,

hyperbilirubinemia, etc.). Attention to trauma-informed care may have the potential to improve outcomes for children with hearing loss.

It is the hope of this researcher that the present investigation will raise awareness of the link between maternal and child ACE exposure among all stakeholders (e.g., teachers of the deaf, audiologists, speech-language pathologists, pediatricians, social workers, DHH adult mentors, early interventionists, policy makers, and others) serving children with hearing loss and their families. Incorporating training on trauma-informed care into family education and professional preparation programs and ongoing professional development initiatives may represent a positive first step in this direction.

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Appendix A

Center for Youth Wellness Adverse Childhood Experiences Questionnaire (CYW ACE-Q; Burke Harris & Renschler, 2015)

Count the number of statements that are things your child has experienced in his/her lifetime and write the TOTAL NUMBER below.

You DO NOT need to indicate which events your child has experienced.

- Your child's parents or guardians were separated or divorced
- Your child lived with a household member who served time in jail or prison
- Your child lived with a household member who was depressed, mentally ill, or attempted suicide
- Your child saw or heard household members hurt or threaten to hurt each other
- A household member swore at, insulted, humiliated, or put your child down in a way that scared your child OR a household member acted in a way that made your child afraid he/she might be physically hurt
- Someone touched your child's private parts (genitals) or asked your child to touch their private parts (genitals) in a sexual way
- More than once, your child went without food, clothing, a place to live, or had no one to protect him/her
- Someone pushed, grabbed, slapped, or threw something at your child OR your child was hit so hard that he/she was injured or had marks
- Your child lived with someone who had a problem with drinking or using drugs
- Your child often felt unsupported, unloved, and/or unprotected
- Your child was in foster care
- Your child experienced harassment or bullying at school
- Your child lived with a parent or guardian who died
- Your child was separated from his/her primary caregiver through deportation or immigration
- Your child had a serious medical procedure or life-threatening illness
- Your child often saw or heard violence in his/her neighborhood or his/her school neighborhood
- Your child was often treated badly because of his/her race, sexual orientation, place of birth, disability, or religion

Appendix B

Center for Youth Wellness Adverse Childhood Experiences Questionnaire (CYW ACE-Q; Burke Harris & Rent-schler, 2015), Maternal Adaptation by Elizabeth A. Rosenzweig

How many of the following did you experience before the age of 18?

- Your parents or guardians were separated or divorced
- You lived with a household member who served time in jail or prison
- You lived with a household member who was depressed, mentally ill, or attempted suicide
- You saw or heard household members hurt or threaten to hurt each other
- A household member swore at, insulted, humiliated, or put you down in a way that scared you OR a household member acted in a way that made you afraid you might be physically hurt
- Someone touched your private parts (genitals) or asked you to touch their private parts (genitals) in a sexual way
- More than once, you went without food, clothing, a place to live, or had no one to protect you
- Someone pushed, grabbed, slapped, or threw something at you OR you were hit so hard that you were injured or had marks
- You lived with someone who had a problem with drinking or using drugs
- You often felt unsupported, unloved, and/or unprotected
- You were in foster care
- You experienced harassment or bullying at school
- You lived with a parent or guardian who died
- You were separated from your primary caregiver through deportation or immigration
- You had a serious medical procedure or life-threatening illness
- You often saw or heard violence in your neighborhood or your school neighborhood
- You were often treated badly because of your race, sexual orientation, place of birth, disability, or religion

EHDInfo



Abstract Submission for the 2024 EHD Annual Conference will open July, 2023

Parent and Provider Perspectives on Early Intervention in Ohio: A Community Collaborative Approach

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Abstract

The network of early intervention (EI) for families with children who are deaf or hard of hearing (DHH) consists of multiple professionals that partner with parents. As part of a community collaborative initiative, diverse perspectives were gathered via online surveys in a state-wide needs assessment to evaluate the landscape of EI in Ohio. The qualitative and quantitative feedback were analyzed in light of the Joint Committee on Infant Hearing's (2007) goals, the 2013 early intervention supplement to those goals, and 2019 update. Care coordination and equitable access emerged as top priorities across all three stakeholder groups queried (audiologists, EI providers, and parents). Through a theory of change framework, these results offer a pathway to strengthening EI to promote the well-being of families and children who are DHH.

Keywords: early intervention, theory of change

Acronyms: ASL = American Sign Language; AVT = Auditory-Verbal Therapy; DHH = deaf or hard of hearing; EHDl = Early Hearing Detection and Intervention; EI = early intervention; PSP = primary service provider

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Early intervention (EI) promotes positive outcomes for children who are deaf or hard of hearing (DHH). Language development flourishes when parents provide contingent, responsive input (Dave et al., 2018; Madigan et al., 2019; Masek et al., 2021; Olson & Masur, 2015). Because approximately 95% of children who are DHH are born to hearing parents, there is a critical need for parents to receive support in fostering their child's language development (Mitchell & Karchmer, 2004). EI providers who work with families and children who are DHH should be equipped to provide caregivers with specialized knowledge about the language and communication needs of children who are DHH so that caregivers may engage with their child in developmentally-appropriate ways (JCIH,

2019). Child outcomes benefit in both the short-term with improved vocabulary in toddler years (Yoshinaga-Itano et al., 2017) and in the long-term, reflected in stronger verbal comprehension and expressive language scores, as well as kindergarten readiness skills as children approach 5 to 6 years in age (Meinzen-Derr et al., 2020; Vohr et al., 2012). This happens most effectively when families receive evidence-based EI services from qualified providers (Kasprzak et al., 2020). Across developmental domains, EI supports the bidirectional nature of language development by empowering parents to scaffold children's emerging communication skills through responsive, age-appropriate social interactions.

Critically, the timing and quality of early intervention services modulates the effectiveness. The Joint Commission on Infant Hearing recommends a 1-3-6 timeline (JCIH, 2019). When screened by 1 month, diagnosed by 3 months, and enrolled into EI services by 6 months, children's vocabulary scores are higher relative to peers who did not meet all three recommendations (Yoshinaga-Itano et al., 2017). Likewise, children's pragmatic language skills are impacted by the timeliness of meeting all three JCIH benchmarks (Yoshinaga-Itano et al., 2020). Additionally, converging evidence from developmental psychology, speech and hearing sciences, and public health has guided the development of best practices for EI: Family-centered, coordinated and collaborative care that is evidence based provided by specialized experts. A framework that invites professionals to view parents as partners in supporting their child's language development is holistic, flexible, and based on families' goals for their child (Moeller et al., 2013; Paul & Roth, 2011; Woods et al., 2011).

The application of these principles into practice can be a challenge, however. Research has identified potential barriers and challenges that impede families during the 1-3-6 timeline (Bush et al., 2017; Jimenez et al., 2012; Shulman et al., 2010). These pinch points in the process reflect the reality that EI is nested within local communities and public health policies. Consequently, applied research addressing barriers to EI are increasingly collaborative in nature, bringing together diverse stakeholders to investigate the lived experiences of providers, families, and children within a particular context (Blaiser & Bargen, 2020; Russ et al., 2010). One way to examine the extent to which one state's Early Hearing Detection and Intervention (EHDI) program (e.g., Ohio) reflects the JCIH (2013) principles specific to early intervention in practice is to seek and understand the perspectives of parents, EI providers, and audiologists through the efforts of a community collaborative.

Community collaboratives can be effective agents of change (Burdine et al., 2010; Clancy et al., 2013). Public health initiatives such as EHDI programs consist of a vast network of providers and supports, including audiologists, speech-language pathologists, teachers of the deaf, pediatric otolaryngologists, Deaf mentors, service coordinators, pediatricians, and other parents. For a family facing a new diagnosis, navigating this network can be a challenge (Holte et al., 2012; Jackson et al., 2008; Scarinci et al., 2018). Recognizing the perspectives and priorities of various stakeholders, a needs assessment offers a starting point for change by identifying barriers and obstacles within a system, given a shared commitment to a common goal (Hickey et al., 2018). Hands & Voices, a parent-led parent-to-parent non-profit organization dedicated to supporting families of children who are DHH regardless of communication modality, in collaboration with the National Center for Hearing Assessment and Management (NCHAM; Ward et al., 2019), conducted a national needs assessment to identify both current strengths and opportunities for growth within EHDI programs. A mixed methods research

design collecting both quantitative and qualitative data for analysis was used to survey parents and EHDI coordinators in addition to focus groups about the resources received/provided and desired. Parents requested access to family-to-family support and comprehensive information for families about their child's diagnosis and communication options. EHDI coordinators identified increasing family engagement in the EHDI system as a top priority moving forward. These two perspectives converge upon a shared recognition that empowering parents with the knowledge and resources to navigate the early intervention system is both necessary and desired.

The purpose of the present study was to elicit perspectives from multiple stakeholder groups about the Ohio EHDI system through a collaborative, interdisciplinary approach. A theory of change framework explicitly identifies potential causal mechanisms that might be responsible for an intervention's success (De Silva et al., 2014; Maini et al., 2018). We adopted the methodology of Ward and colleagues (2019) to conduct a needs assessment specific to the state of Ohio. This step is the first when adopting a Theory of Change framework to strengthen public health initiatives. This approach allows for a localized and nuanced lens through which potential mechanisms for change can be identified to strengthen the EHDI program moving forward, guided by the direct input of those who participate in the system in hopes of engaging more families in the periphery. These data can then inform other states' EHDI programs who are engaging in quality improvement.

Method

Overview

This project was deemed exempt from review by The Ohio State University's Institutional Review Board because no protected health information was collected. A community collaborative approach was used to guide the design and implementation of this needs assessment about Part C early intervention services in Ohio. A network of professionals and parents was formed with funding from the Oberkotter Foundation to engage in a year-long discovery period to conduct a needs assessment of the early intervention landscape across the state of Ohio. The aim was to formulate a theory of change to strengthen the early intervention system for families with children who are DHH. A core group of 22 stakeholders from diverse backgrounds were led in this endeavor by a steering committee between Fall 2017 and Summer 2018. Members included parents, pediatric otolaryngologists, speech and language pathologists, audiologists, Part C state representatives, and DHH adults. A professional facilitator ensured that all participants had equitable opportunities to share their perspectives during in-person and virtual meetings as well as through electronic surveys. To maximize engagement of stakeholders, tiered levels of participation also included regional groups who hosted the focus groups as well as a review group that provided written feedback on documents developed by the community collaborative.

Qualitative and quantitative data were gathered from three key stakeholders: parents, EI providers, and audiologists.

Surveys were conducted online using REDcap. Focus group sessions were held in person in four regions of the state. The current article summarizes the survey data.

Survey Participants

Over the course of survey data collection spanning April through August 2018, a total of 158 respondents initiated the survey. Ultimately, 82 respondents completed the survey through the last question, 22 partially completed the survey, and 54 respondents initiated the survey but abandoned it prior to answering any questions. Data from the 104 complete and partially complete survey responses were included in this analysis. Complete surveys represented answers from 14 audiologists, 33 EI providers, and 35 parents while partially complete surveys represented answers from 5 additional EI providers and 17 additional parents.

Audiologists

Audiologists who responded to the survey (*n* = 14) primarily came from the hospital setting (pediatric, non-profit, or general hospital; *n* = 10, 71.4%) and had a clinical case load between 6 and 25% (*n* = 5, 35.7%) of exclusively pediatric patients. Only one audiologist reported that their clinical caseload was 0–5% and one other reported that their caseload was 76–100% pediatrics. The top three clinical services provided by audiologist respondents included hearing evaluations/assessments (*n* = 12, 85.7%), hearing aid fitting and management (*n* = 10, 71.4%) and hearing aid technology counseling (*n* = 12, 85.7%). Additional demographic information for audiologists appears in Table 1.

Table 1
Audiologist Demographic Information

Employment Setting	<i>n</i>	%
Pediatric Hospital/Hospital	10	71.4
School/EI Program	2	14.3
Private Clinic/Speech & Hearing Center	2	14.3
Pediatric Case Load Percent	<i>n</i>	%
0–5	1	7.1
6–25	5	35.7
26–50	3	21.4
51–75	4	28.6
76–100	1	7.1
Services Provided	<i>n</i>	%
Hearing Evaluations/Assessments	12	85.7
Hearing Aid Technology Counseling	12	85.7
Hearing Aid Fitting/Management	10	71.4
Communication Mode Counseling	9	64.3
Cochlear Implant Management	7	50.0
Aural Rehabilitation/Therapy	1	7.1
Other (Screening and family coaching)	1	7.1

Note. Includes work setting, pediatric caseload, and clinical services provided. For clinical services, respondents indicated all services provided. EI = early intervention.

Early Intervention Providers

Table 2 provides an overview of EI providers' background and education. Early intervention providers who responded to the survey (*n* = 38) reported a variety of educational backgrounds, with the most frequently reported including speech language pathology (*n* = 10, 26.5%), elementary education (*n* = 4, 10.5%), and special education (*n* = 3, 7.9%). Respondents also noted a variety of specialty certifications, most often including: speech language pathology (*n* = 10, 26.3%), deaf and hard of hearing early intervention specialist (*n* = 9, 23.9%). Lastly, respondents reported a variety of years of experience in the EI field, with most reporting 16+ years (*n* = 15, 39.5%). EI providers reported a mean of 10.9 children with hearing loss on their current clinical caseload, with a range of 1 to 100.

Table 2
Early Intervention Provider Demographics

Educational Background	<i>n</i>	%
Speech-Language Pathology	10	26.3
Elementary Education	4	10.5
Special Education	3	7.9
Audiology	3	7.9
Other*	11	28.9
Not disclosed	7	18.4
Specialty Certificates	<i>n</i>	%
Speech-Language Pathology	10	26.3
DHH Early Intervention Specialist	9	23.9
Audiologist	5	13.2
Teacher of the Deaf	3	7.9
Educational Audiologist	2	5.3
LSLS/Auditory Verbal Therapist	2	5.3
Other**	7	18.4
Years of Experience	<i>n</i>	%
0–2	6	15.8
3–5	4	10.5
6–10	5	13.2
11–15	7	18.4
16+	15	39.5
Not disclosed	1	2.6

Note. Includes educational background, specialty certification, and years of experience providing early intervention services. *Other educational backgrounds included nursing, social work (*n* = 4), child & family community services, physical therapy (*n* = 2), human development and family studies, school psychology, and public administration. **Other specialty certifications included Department of Developmental Disabilities (DS), early interventionist (*n* = 2), service coordination (*n* = 3), and early childhood special education administration. DHH = Deaf or Hard of Hearing; LSLS = Listening and Spoken Language Specialist

Parents

Fifty-two parents provided answers to survey questions either in part or in whole. Parents represented families with children of varying ages with a range of birth years from 2009 through 2017 and diagnosis years ranging from 2009 through 2018. Parents who provided data related to specific hearing diagnosis related date for their children ($n = 45$) reported a mean age of diagnosis of 6.02 months with a range from 1 to 75 months and a median of 1 month. Ninety percent of the children ($n = 47$) reportedly had bilateral hearing loss (8 asymmetric) while four (7.6%) had unilateral hearing loss and one parent did not report the laterality of hearing loss. Table 3 represents the degree and configuration of hearing loss reported by each family as well as the etiology (if known), devices, and communication mode used by the children most of the time. One quarter ($n = 13$) of the children presented with profound hearing loss and 55.8% reported idiopathic/unknown etiology ($n = 29$). Binaural hearing aids were worn by 55.8% ($n = 29$) and 21.2% reported using bilateral cochlear implants ($n = 11$). Total communication was reported as the primary communication mode for 46.2% ($n = 24$) and listening/spoken language was reported for 34.6% ($n = 18$). Additionally, 69.2% ($n = 36$) of parents reported their children presented with speech/language delay, but a variety of comorbid diagnoses were also reported, including: motor delay ($n = 16$, 30.8%), social/emotional delay ($n = 16$, 30.8%), reduced vision ($n = 10$, 19.2%), global developmental delay ($n = 8$, 15.4%), and autism ($n = 3$, 5.8%). Sensory processing disorder ($n = 2$), attention deficit hyperactivity disorder ($n = 2$), and dyslexia were noted for “other” developmental delays reported for children.

Lastly, families represented various races and socioeconomic status levels based on reported household income, insurance coverage, and primary caregiver education level. For race, 34 families (65.4%) identified themselves as white/Caucasian, one family (1.9%) identified as Asian, and one family (1.9%) identified as Pacific Islander. Sixteen families (30.8%) declined to answer this question. Thirty-five families reported household income with the most frequently reported income between \$75,000 and \$99,999 ($n = 9$, 25.7%). Health insurance coverage of the children was reported by 36 families and respondents could indicate if they had more than one source for health insurance. The vast majority of respondents reported having private insurance ($n = 30$, 83.3%), and 10 (27.8%) reported that they had Medicaid coverage. Additionally, 16 families reported that they benefited from Ohio-based supplemental insurance programs including the Children with Medical Handicaps (CMH) supplement ($n = 14$, 38.9%) and the Ohio Hearing Aid Assistance Program (OHAAP) for the purchase of hearing aids ($n = 2$, 5.5%). For primary caregiver educational level, most respondents reported having a Bachelor's Degree ($n = 11$, 31.4%).

Materials

Three surveys were developed to query audiologists, parents, and providers, respectively (available upon request). These surveys were adapted with permission from NCHAM's EI SNAPSHOT project (Ward et al., 2019) by the community collaborative so that all questions were specific to Ohio's EHDI program and resources. Although the focus of survey questions was Part C Early Intervention services, a spectrum of questions was asked to gauge the entire family journey. Therefore responses likely reflect input regarding a combination of screening, diagnosis, Part C EI and general early intervention processes in Ohio.

Table 3

Hearing-Related Demographic Data for Children of Parents who Responded to this Survey

Degree/Laterality	<i>n</i>	%
Profound	13	25
Moderately-Severe	9	17.3
Mild	4	15.4
Severe	5	15.4
Asymmetric	8	9.6
Moderate	8	7.7
Unilateral	4	7.7
Did not respond	1	1.9
Etiology	<i>n</i>	%
Unknown/Idiopathic	29	55.8
Genetic	16	30.8
cCMV	5	9.6
Other*	2	3.8
Device(s)	<i>n</i>	%
Binaural HA	29	55.8
Binaural CI	11	21.2
None	5	9.6
Bone Conduction Device	2	3.8
Bimodal	2	3.8
Unilateral CI	1	1.9
Unilateral HA	1	1.9
No response	1	1.9
Communication Mode (most of the time)	<i>n</i>	%
Total Communication	24	46.2
Listening/Spoken Language	18	34.6
No Response	3	5.8
Pointing/Grunting	3	5.8
American Sign Language	2	3.8
Cued Speech	2	3.8

Note. Includes degree/laterality, cause, device use, and communication mode. *Other etiologies reported were Cochlear Dysplasia/Mondini Malformation and prematurity. cCMV = congenital cytomegalovirus, CI = cochlear implant; HA = hearing aid(s).

Study data were collected and managed using Research Electronic Data Capture (REDCap) tools hosted at Ohio State University (Harris et al., 2009, 2019). REDCap is a secure, web-based software platform designed to support data capture for research studies, providing (a) an intuitive interface for validated data capture; (b) audit trails for tracking data manipulation and export procedures; (c) automated export procedures for seamless data downloads to common statistical packages; and (d) procedures for data integration and interoperability with external sources.

Recruitment

Survey links were disseminated via email to participating members of the community collaborative, who were invited to share the link with their colleagues and families. The link also was shared through Ohio's Early Hearing Detection and Intervention (EHDI) listserv to providers and affiliated professionals. Focus group information was shared this way as well.

Data Analysis Plan

Quantitative questions from the survey were categorized according to which JCIH goal they addressed. The community collaborative made *a priori* decisions regarding which goals to focus on when developing the survey. Specifically, the collaborative concentrated its efforts on the goals concerning access to timely services (Goal 1), resources (Goal 3), and parent-to-parent support (Goal 9). Survey questions were categorized by topic according to JCIH (2013) EI supplement goals. Whenever possible, data from multiple perspectives (i.e., parents, providers, and/or audiologists) for a particular topic are reported.

Respondents of the online survey were often asked to rate aspects of their clinical practice, process, or knowledge based on a four-point Likert scale, 1 meaning *excellent* and 4 meaning *poor*. Quantitative data were analyzed both in terms of the percentage of respondents indicating a particular rate and via mean ratings (M_{rating}) which were calculated and reported according to theme. Mean ratings closer to 1 suggest positive reports while mean ratings closer to 4 reflect negative reports.

Qualitative data from open-ended prompts on each survey (Appendix) were analyzed using content analysis to derive themes (Hsieh & Shannon, 2005; Krippendorff, 1989). First, comments were parsed to identify specific utterances that could be considered as regarding the same topic so that long-form answers from single respondents that addressed multiple topic areas could be coded individually. Each of the respondent groups were then coded for theme independently by two of three available reviewers (JR, CD, UF). A consensus process was then facilitated by the third reviewer to identify the ultimate theme(s) conveyed in the utterance. A natural filtering effect occurs in the process of sorting and identifying themes which allows for specific themes to be highlighted for each of the respondent groups, some of which ultimately overlapped across groups.

Results

Quantitative results and rankings will be discussed within the context of JCIH (2013) EI supplement goals below. For qualitative responses, after parsing for topic there were 26 utterances from audiologists, 53 utterances from providers, and 46 utterances from parents after consensus was obtained from all three reviewers (JR, CD, UF). Overarching themes present in the utterances are presented in Tables 4 and 5. Qualitative quotations will be incorporated in the discussion about quantitative data below to illustrate commonalities or disparate themes indicated across the data.

Goal 1: Timely Access to EI Services

Families must navigate the process of initial screening after birth to diagnostic testing, confirmation of hearing loss, navigating management options, and enrollment into early intervention services. The JCIH recommends that families have access to timely and coordinated entry into EI.

Parents

Eighty-five percent of families agreed with the statement that it was "easy to get information about how to enroll in EI." However, 31.1% of families felt that the process to get services for their child was confusing.

Audiologists

Audiologists reported several challenges related to timely and coordinated entry into EI. More than one-third (35.7%) reported difficulty when trying to contact Part C EI. Furthermore, 78.6% of audiologists reported that they heard from families directly about difficulties they experienced with the EI system. Almost all audiologists expressed concern that the children on their caseload were receiving inappropriate EI services (92.9%), and over half of parents (71.4%) requested information about private services from the audiologists.

EI Providers

EI providers were invited to rate their ability to assist families with various aspects of early intervention using a Likert rating from 1 (*excellent*) to 4 (*poor*). More than half of respondents (69.7%) self-reported their ability to help families learn about all communication modalities as excellent or good; the same percentage reported that they were able to adequately (i.e., rating of excellent or good) connect families with services specific to their family's communication choice. An overwhelming majority of EI providers (97.0%) felt confident in their ability to coordinate with other EI providers, with ratings of either excellent or good.

Additionally, the survey probed for EI providers' perceptions of barriers faced by families. Respondents indicated if various potential obstacles were not a barrier, a small barrier, or a large barrier. Finding out about the free, state-provided EI services was considered a small barrier by 54.4% of EI providers; 18.2% considered it to be a large barrier. Thirty-nine percent of providers did not believe that finding out about EI was a barrier at all. Enrolling into free, state-provided EI services was not considered a barrier to

Table 4*Themes and Examples*

Theme	Included within theme	Examples
Care coordination	Tracking/monitoring referrals, communication among professionals, progress monitoring	<p>“No thorough system in place to track and monitor children referred for hearing loss specifically.” (Audiologist)</p> <p>“It would be amazing if there were a way to communicate between pediatricians and EI.” (Parent)</p> <p>“The EI program is continually attempting to educate and encourage counties to reach out as soon as they have a child with a hearing loss, but it is not always happening. There are too many people involved with a family before they actually get involved with early intervention hearing services, leaving many opportunities to fall through the cracks. It would be great to have a system that directly notifies hearing specialists of a child with a newly diagnosed hearing loss.” (Provider)</p>
EI quality concerns	Unbiased support for all languages and communication modes, changes to state EHDI program	<p>“Also, home based services once/month is not the same as weekly private speech therapy, and I feel like families are not counseled properly by the EI therapists on these differences.” (Audiologist)</p> <p>“The only thing I regret is that I wish there would have been more support for us to learn ASL for our moderate to severe bilateral hearing loss child. We wanted to use sign for night times at home, bath time and summers in the water. But we were told since our child was listening and our ultimate goal was for her to speak and listen and use English, there were not ASL supporters for us.” (Parent)</p> <p>“I would like to see a family choose a communication modality and then have the opportunity to be paired with an EI provider that supports their choice. If LSL is their choice, then working with a LSL provider through EI should be an option. If working with a family who chooses ASL, they should be paired with a provider who can teach and support the family with learning ASL.” (Provider)</p>
Equitable access	Limited access to services in regions of the state, lack of access to specialized providers, lack of training for EI providers, lack of EI curriculum, access to telehealth	<p>“Hearing specific services are few and far between in the state of Ohio. Families that do not live in an urban area are often underserved.” (Audiologist)</p> <p>“Could professionals with an expertise in hearing loss develop a program and curriculum that is offered to all families in Ohio?” (Provider)</p>
Parent knowledge and education	What services are available, importance of EI, counseling on communication, resources for families	<p>“Counseling regarding the importance of early intervention, communication mode, resources, etc.” (Audiologist)</p>
Positive experience	Quality resources available, referrals made in rural areas, in-home services available, unbiased support	<p>“I do think families appreciate the in-home service model and those that do get timely EI services do have good things to say about the providers and the process of working with Early Intervention in general.” (Audiologist)</p> <p>“It is without a doubt their unbiased support that also helped me think about communication methods.” (Parent)</p>

Note. ASL = American Sign Language; EI = early intervention.

families of children birth to three by 39.4% of providers. Forty-two percent felt that enrolling was a small barrier; the remaining 18.2% felt it to be a large barrier. In contrast, a majority of providers felt that “getting providers to talk to one another” was either a small (45.5%) or large (42.4%) barrier. Only 12.1% of respondents felt that coordination among providers was not a barrier at all.

Open-Ended Responses

Themes derived from the content analysis of open-ended responses are presented in Table 5. Comments related to care coordination were the most frequent, and all three groups of respondents provided feedback related to this topic. Comments related to care coordination spanned the entire EHDI process and included comments regarding provider-parent coordination as well as provider-provider coordination. Twenty-two percent of all open-ended responses addressed some aspect of care coordination. Specifically, 20.3% of feedback from providers, 10.9% of parents’ feedback, and 46.2% of input from audiologists addressed topics and concerns about the process of identifying, enrolling, and navigating the EI system. Coordinated communication among professionals emerged as an area of concern, as evidenced by responses such as this quote from an audiologist, “There is also a lack of communication/training between providers on all fronts (audiologist, speech pathologist, EI provider, etc.) that leads to miscommunication or conflicting recommendations with families.”

Table 5
Summary of Derived Themes from Qualitative Comments Compared across Providers, Parents, and Audiologists

Theme	Provider	Parent	Audiologist	Tally
Care Coordination	12	5	12	29
Positive Experience	0	20	5	25
Equitable Access/Equity of Services	21	1	2	24
Quality of EI Services (negative)	2	15	5	22
Policy/System Changes/Funding	14	2	0	16
Resources	6	0	2	8
Family Choice & Family Journey	2	3	0	5

A second theme identified from the open-ended responses that aligned with the JCIH’s recommendation that state EHDI programs “develop a mechanism that ensures family access to all available resources and information that is accurate, well-balanced, comprehensive, and conveyed in an unbiased manner” (JCIH, 2013; pg. e1327). This *resources* theme accounted for approximately six percent of the comments ($n = 8$). Providers mentioned this theme more frequently than audiologists, and interestingly, no parents made note of any topic related to high-quality, unbiased information. One provider expressed the desire to “Get all county programs to the same level with the same resources and knowledge” and for “a compilation of affordable resources for additional training for providers and a list of resources to refer families.” Another provider posed the question of “Could professionals with an expertise in hearing loss develop a program and curriculum that is offered to all families in Ohio?”

When looking at the nature of comments across respondent groups overall, parents more often offered comments about the experience of EI while audiologists were more concerned with effective care coordination. Lastly, EI providers offered more comments about access to services and policy-related factors. Below we explore how the quantitative data and qualitative open-ended comments address the JCIH (2013) goals of interest in this study.

Goal 3: Specialized Providers Equipped with Expertise

Supporting the language development of children who are DHH has cascading effects on their later communication and literacy skills. Consequently, JCIH (2007; 2013) recommends that families have access to specialized providers with the professional qualifications to provide evidence-based intervention to children and their families following diagnosis. Specifically, JCIH recommends that families have access to “qualified providers” regardless of their communication modality (i.e., American Sign Language or spoken language).

Parents

Eight-two percent of families felt that they were provided choices related to the supports and services available to them. However, 50% of the parents who desired sign language instruction reported problems accessing such services through Ohio’s EI system. In contrast, only one-quarter of families who needed Auditory-Verbal Therapy (AVT) experienced difficulties accessing AVT through Ohio’s EI system.

Audiologists

Audiologists were queried about receiving information or training about Ohio’s EI system. Less than one third of audiologists (28.6%) reported receiving information or training about Ohio’s early intervention system.

EI Providers

The survey queried EI providers to rate their knowledge about various topics related to providing early intervention

services to children who are DHH using a Likert scale from 1 to 4 (1 = *excellent*, 2 = *good*, 3 = *fair*, 4 = *poor*). Providers felt knowledgeable about the administrative aspects of their role, such as service coordination ($M_{\text{rating}} = 1.82$) and IDEA Part C regulations ($M_{\text{rating}} = 1.94$). In regard to their clinical care responsibilities, EI providers reported feeling knowledgeable about supporting families ($M_{\text{rating}} = 1.85$) and providing telehealth/teleintervention ($M_{\text{rating}} = 1.54$). In contrast, they reported feeling much less knowledgeable about topics related specifically to deafness and hearing loss, particularly assessing children who are DHH ($M_{\text{rating}} = 3.03$), language development ($M_{\text{rating}} = 2.77$), and teaching children who use either sign ($M_{\text{rating}} = 2.56$) or listening and spoken language ($M_{\text{rating}} = 2.8$).

The survey also inquired about EI providers' training through formal education, on-the-job training, and in-service/continuing education. A majority of EI providers reported receiving formal education in early intervention (75.7%), family support (91.9%), service coordination (78.3%), and IDEA Part C regulations (78.4%). However, only half of respondents indicated that they received any formal training on the assessment of children who are DHH (52.8%), teaching children using sign language (52.8%), teaching children using listening and spoken language (59.5), and teaching children who use total communication (48.6%).

For a majority of EI providers, on-the-job training experiences centered on early intervention (59.5%). Almost half of respondents reported that they received on-the-job training related to assessment and teaching children who use Listening and Spoken Language as their primary communication modality (47.2% and 48.6%, respectively). Very few EI providers (< 1%) reported learning about any topic during in-service or continuing education opportunities, with the exception of pre-literacy instruction, for which 41.7% of respondents endorsed participating in continuing education or in-service training.

When asked to rate the adequacy of education and background in relation to preparedness for providing services for families and children who are DHH, 42.1% reported their background was *adequate* ($n = 16$), although the same percentage/number reported it was *inadequate*. Only two respondents (5.3%) reported their educational background was *very adequate* and one (2.6%) reported *very inadequate*, while three respondents (7.9%) chose not to answer.

Open-Ended Responses

Equitable access/equity of services emerged from the open-ended responses as a key theme that directly addresses Goal 3. This theme was present in comments from all three groups of respondents; 7.7% of audiologists' comments, 2.2% of parents' feedback, and 36.8% of providers' input related to equitable access/equity of services, particularly as it related to the more rural regions of Ohio. Overall, 18.6% of all comments addressed equitable access/equity of services.

The theme of equitable access/equity of services primarily consisted of two subcategories of comments: those related

to equitable access regardless of geographic location and those related to a need for specialized service providers. The interrelated nature of these two themes is exemplified via provider feedback such as a desire for "more access to a variety of specialized supports for small rural counties in the state." Mirroring our survey data in which providers reported a relative lack of formal training on a variety of EI-related topics, providers inquired about a variety of possibilities to address a need for equitable access to specialized services. Ideas ranged from "a compilation of affordable resources for additional training for providers" to a more unified approach: "Could professionals with an expertise in hearing loss develop a program and curriculum that is offered to all families in Ohio?"

The qualitative analysis of comments revealed a second theme of family choice/the family journey. This theme accounted for 3.9% of the comments overall. For example, one parent remarked, "What I'd like to say to any professional listening: stop telling us you and your 'side' or 'camp' know best how to educate our child. You have valuable information and we want to hear it. We want to help our child with any strategies you know about."

Goal 9: Parent-to-Parent Support

Connecting with other families who also have a child who is DHH is both a powerful resource network and a support system for a family whose young child was recently diagnosed with a hearing loss. Shared experiences create common ground. As such, JCIH (2013) recommends the development and implementation of formally-trained parent-to-parent support systems within EHDl systems.

Parents

Access to parent-to-parent support emerged as a significant area in need of improvement. Only 47.06% of parents who answered the question agreed with the statement that "My service coordinator helped me get in touch with other parents for help and support." Furthermore, only 53.1% of families felt that the information they received about connecting with families who also had children who are DHH was excellent or good. Almost half rated that information as fair or poor, with poor meaning not mentioned at all. Less than half felt that the information that they received about AG Bell and Hands & Voices (both at the national and state level) was excellent or good, while approximately one-third of the respondents reported they received no information on these resources.

Audiologists

The survey for audiologists did not ask any questions specific to parent-to-parent support.

EI Providers

EI providers were asked to rate their knowledge of various organizations that foster parent-to-parent connections, such as Hands & Voices. Over half of respondents (52.9%) rated their knowledge of the national Hands & Voices organization as poor; 45.5% indicated that their knowledge

of local Ohio chapters of Hands & Voices was poor. A similar percentage of providers (48.5%) reported that they never referred families to local chapters of Hands & Voices. However, 81.8% of EI providers rated their ability to connect families with other families as excellent or good, although they do so with varied frequency: one-third indicated that they do so sometimes, 27.3% reported doing so often. One third of providers endorsed that they always connected families with other families. Lack of family-to-family support was considered a small barrier for families by almost three-quarters of EI providers (72.7%); 18.2% considered it to be a large barrier. The remaining 9.1% of providers felt that a lack of access to parent-to-parent support was not a barrier at all.

Open-Ended Responses

No provider, audiologist, or parent response addressed parent-to-parent support.

Additional Themes

The nature of the open-ended survey questions allowed parents, providers, and audiologists to share ideas or opinions on *any* topic. Consequently, several themes emerged from the open-ended data that did not fall under a particular JCIH goal. More than a third of all responses (36.4%) expressed an explicitly positive or negative opinion. Audiologists expressed an equal number of positive and negative comments ($n = 5$ for each). Parents, on the other hand, expressed more positive ($n = 20$) than negative ($n = 15$) comments. Only 3.5% of provider comments were evaluative in nature, with two comments coded as expressing a negative experience.

Comments related to policy, including funding and systemic changes, accounted for 12.4% of all comments. EI providers were responsible for 87.5% of these comments; the remaining two comments were from parents. Providers noted that navigating the various policy changes within the Ohio EHDI system has been a challenge; specifically, the 2016 shift to a Primary Service Model was referenced.

Discussion

The goal of this community collaborative was to study the perspective of three key stakeholder groups in Ohio's EHDI program: parents, audiologists, and EI providers. Using the JCIH goals as a framework, these stakeholders were afforded an opportunity to reflect upon their lived experiences within the EI system through both qualitative (i.e., focus groups) and quantitative (i.e., surveys) methods. This design allowed for an exploration of a single topic (e.g., access to parent-to-parent support) from various viewpoints. The data revealed several points of consensus as well as divergence among and within the three stakeholder groups.

Loss-to-follow-up emerged as a critical concern expressed by providers and audiologists when considering the first JCIH goal of timely access to EI services. Given the nature of the recruitment methods employed in this study, only families who were connected in some way to EI services

were queried. Nevertheless, among parent respondents, almost one-third reported that the process of enrolling into EI was "confusing." These results are consistent with previous survey data from parents who reported navigating the EHDI system as overwhelming with limited information shared for next steps to happen in a timely manner (Larsen et al., 2012). Despite significant progress in EHDI systems as a whole, many of the same concerns regarding loss-to-follow-up remain today, decades after the first universal programs were instituted.

EI providers expressed seemingly conflicting viewpoints: they felt equipped to support families. However, they were much less confident in their ability to teach children who use either American Sign Language (ASL) or spoken language. Such a discrepancy in confidence may be rooted in the changing role of the EI provider. To minimize the number of professionals working with a family in the home, a primary service provider (PSP) model has been implemented, whereby families receive EI services that are developmental, rather than deafness-specific, and delivered using a transdisciplinary approach. Theoretically, in a PSP model for families of children who are DHH, the PSP implements strategies informed by specialists in deafness and early language development. However, due to the complexity of state EHDI systems and potential for lack of qualified providers in a specific geographical location, families are not guaranteed services by a deafness-specific provider. The EI providers queried for the current study did not report backgrounds specifically related to facilitating language development in young children who are DHH. Further, a lack of confidence may also stem from a lack of training programs or variability in providers' backgrounds. Speech-language pathology training programs do not routinely include coursework and training related to the unique language and communication needs of this population. Parents reported a similar paradox: although most reported that they were given choices about their family's preferred communication modality, half of families who decided to pursue ASL had difficulty accessing high-quality services provided by the state. Collectively, these barriers to high-quality EI services due to limited qualified providers or limited service options could be addressed by improving university training through specialized DHH coursework, offering ongoing professional development to strengthen knowledge of current EI providers about the impact of deafness on development, and providing care coordination for families to navigate the often complicated EHDI system so that they can be connected with appropriate services and resources.

Although EI providers endorsed an excellent grasp of the logistics of providing EI, they concurrently endorsed feeling only good to fair in topics regarding assessment of children who are DHH and language development. One possible explanation for these findings might be due to the fact that, for the most part, the logistics of EI systems cut across disability categories; therefore, in-service training opportunities related to the EI system might be more readily available to providers than training related

to assessment. The shortage of deaf education training programs (Johnson, 2004) has also created the challenge of employing EI providers with expertise in assessing young children who are DHH. Universities, policy makers, and advocates might consider opportunities for addressing this shortage given that JCIH recommends families receive services from providers with specialized knowledge. Furthermore, the refinement of research related to early language development in young children who are DHH and the effects of EI stands in contrast to the emerging nature of the nuances of high-quality EI services in practice.

Across all three stakeholder groups, parent-to-parent support emerged as a need when explicitly asked, reflecting previous research demonstrating the high value that parents of children who are DHH place on peer engagement (Haddad et al., 2019; Hintermair, 2000; Zaidman-Zait et al., 2016). In fact, parents cite parent-to-parent support as critical to their ability to navigate their child's care and an important avenue for acquiring knowledge about raising a child who is DHH (Haddad et al., 2019). Among EI providers, insufficient knowledge of parent-to-parent support organizations was reported despite providers' assertion that they consistently connected families. However, this topic did not arise in any open-response comments, suggesting that other areas may be prioritized, such as equitable access to specialized providers.

Across the three goals of timely entry, access to experts, and parent-to-parent support, a common theme emerges: There is a great onus placed on families to coordinate their care, from navigating the EI enrollment process to securing services that match their family's goals. Many families, however, do not have the time, resources, or knowledge to navigate this process with ease. All too often, the family is the hub in a multi-spoke wheel. The current Ohio EHDI system overall is not achieving what the national EHDI goals strive for based on perspectives from multiple stakeholder groups.

Through the lens of a Theory of Change framework, this state-wide needs assessment identifies several potential mechanisms to strengthen the efficacy of early intervention. For instance, converging data suggest that parent-to-parent support that equips and connects families may mediate the extent to which families advocate for and engage in the early intervention system. The identification of this (and other) potential causal mechanisms offers a pathway for future research. The needs assessment conducted by De Silva and colleagues (2014) during their implementation of a Theory of Change framework to develop a mental health intervention revealed several potential markers to measure when evaluating a pilot program. Likewise, stakeholders in Ohio's EDHI system may consider the role of parent-to-parent support when evaluating future EHDI programs.

Limitations

Several limitations need to be considered before extrapolating these results to other states' stakeholders within their own EHDI programs. First, the respondents who completed the online survey were predominantly

white/Caucasian and self-reported a high level of SES. The extent to which these perspectives would apply to families from diverse race/ethnic, socioeconomic, or linguistic backgrounds is worthy of future investigation. Additionally, for many respondents, data was retrospective in nature as families reported on children who spanned 0 to 7 years old. Although the focus of this project was on Part C EI services, it cannot be ruled out that providers and parents also included other types of early intervention (e.g., speech therapy, private group or family therapies, etc.) when responding to survey questions. Finally, this project only addressed three JCIH (2013) EI supplement goals, given the particular focus of the community collaborative. Additional research that addresses the extent to which parents and professionals endorse that their lived experiences align with the recommended best practices is warranted.

Conclusion

EHDI systems are notoriously complex and often difficult for families, and sometimes providers, to navigate. How state EHDI systems function can fluctuate depending on funding, consolidation of resources (PSP model versus specialist-oriented model), and other unforeseen circumstances. However, gathering stakeholder input is one path to illuminating the difficulties and identifying potential solutions unique to families of children who are DHH and the providers who serve them. The community collaborative in Ohio was able to identify barriers to EI, including limited equitable access to specialized providers, limited information sharing and access to parent-to-parent support, and the need for care coordination to facilitate enrollment into EI. Our experience in identifying these key attributes can serve as a model for other states to evaluate their own programs to identify their unique needs.

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Appendix

Open-ended Questions Posed to Respondents

Questions for Parents:

- Knowing what you know now, what would you have done differently in obtaining Early Intervention Hearing services for your child?
- What is the most important piece of advice you would give a parent whose child has recently been diagnosed with hearing loss regarding early intervention and family supports?

Questions for Providers:

- In your opinion, what changes could be made to help early intervention work better in your state?

Questions for Audiologists:

- What challenges do families face who have an infant or toddler (0–3) who is DHH living in Ohio?
- What do you think is working well in your state for families who have an infant or toddler (0–3) who is DHH?

EHDInfo



Improving South Dakota Parents' Knowledge of Congenital Cytomegalovirus

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Abstract

Introduction: Congenital cytomegalovirus (cCMV) is the most common infectious condition present at birth and the leading non-genetic cause of sensorineural hearing loss in children. Despite cCMV being common and preventable, knowledge of cCMV remains low among individuals in the United States (Doutre et al., 2016). Sudden infant death syndrome (SIDS) is also common among infants in the United States, but unlike cCMV, several studies have researched which educational methods have been most effective for improving parents' knowledge of safe sleep practices. The purpose of this study was to investigate whether educational methods shown to be most effective for improving parents' knowledge of SIDS could also be used to improve parents' knowledge of cCMV.

Method: Forty-five participants completed an online study consisting of a pre-education survey on cCMV, written and verbal education on cCMV, and a post-education survey. Methods were based off a SIDS education study conducted by Dufer et al. (2017).

Results: Results of this study revealed that the provision of verbal and written education resulted in a significant increase in South Dakota parents' knowledge of cCMV ($p < 0.05$).

Conclusion: Educational methods used to improve parents' knowledge of SIDS can also be used to improve South Dakota parents' knowledge of cCMV.

Keywords: Congenital cytomegalovirus, sudden infant death syndrome, parent education, parent knowledge

Acronyms: cCMV = congenital cytomegalovirus; SIDS = Sudden Infant Death Syndrome

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Cytomegalovirus (CMV) is a common viral condition. By age 40, more than 50% of adults have been infected with CMV; by age 80, that percentage rises to 90% of adults (CDC, 2020; Staras et al., 2006). If the virus passes from a pregnant woman to her child in utero, the infant is born with congenital cytomegalovirus (cCMV). Approximately one out of three pregnant women infected with CMV will pass the virus to their child in utero, leading to an estimated 30,000 annual cases of cCMV in the United States (National CMV Foundation, "What is CMV?", n.d.; National CMV Foundation, "Newborn Screening", n.d.).

In most cases, infected adults present with symptoms that mimic the common cold, or they will show no symptoms of infection (National CMV Foundation, "Signs and Symptoms of CMV Infection", n.d.). Unlike typical CMV infection in adults, cCMV infection in infants can cause severe symptoms. Of the 30,000 infants infected with cCMV each

year, about 6,000 infants (or one in five) will develop long-term health effects (CDC, 2020).

Some of the long-term health effects associated with cCMV include microcephaly, seizures, vision loss, cognitive impairment, and problems of the liver, spleen, and lungs (National CMV Foundation, "Signs and Symptoms of CMV Infection," n.d.). Another common symptom of cCMV infection is hearing loss. In fact, cCMV is the primary cause of non-genetic, sensorineural hearing loss in infants. The hearing loss may be progressive and can have a delayed onset (Goderis et al., 2014). Around 10 to 15% of asymptomatic infants (i.e., no visible symptoms of CMV infection) and up to 75% of symptomatic infants (i.e., visible symptoms of CMV infection) may develop hearing loss (National CMV Foundation, "Possible Outcomes of Congenital CMV," n.d.).

CMV can be spread through bodily fluids, such as saliva, urine, and blood (CDC, 2020). Women who are infected with CMV during pregnancy most commonly contract the virus from children younger than three years of age (Adler & Nigro, 2013). Therefore, women who are frequently in contact with young children (e.g., daycare providers, women with young children of their own) may be at a greater risk for CMV infection. Although CMV infection is common, it is also preventable. Behavioral hygienic practices can reduce one's risk of being infected with CMV. Such practices include the following: not sharing food, utensils, or a toothbrush with young children; not putting a used pacifier in one's mouth; avoiding contact with saliva when kissing a child; and washing one's hands after changing a diaper or touching children's toys (National CMV Foundation, "CMV Prevention and Healthy Pregnancy Tips," n.d.).

Although CMV infection is both serious and preventable, few individuals in the United States are aware of the virus. A 2005 *HealthStyles* survey revealed that only 14% of female respondents had heard of CMV (Ross et al., 2008). Five years later, the 2010 *HealthStyles* survey showed that 13% of women and 7% of men (whose responses to CMV questions in 2005 were not reported) were aware of CMV (Cannon et al., 2012). The most recent 2016 *HealthStyles* survey showed that 9% of women and 5% of men were aware of CMV. The decrease in women's awareness of CMV from 2005 to 2016 is statistically significant (OR = 0.94, 95% CI = [0.93, 0.95], $p < .0001$; Douthett et al., 2016). Respondents' awareness of CMV was lower than all other conditions surveyed, such as spina bifida, Down syndrome, and fetal alcohol syndrome.

A previous study by Reimann et al. (2020) looked at South Dakota parents' knowledge of cCMV and found similar trends. South Dakota has no current legislation for cCMV screening; however, the two main hospital systems in the state (Sanford Health and Avera Health) have implemented hearing-targeted cCMV screening protocols (i.e., cCMV is screened for if the infant does not pass the newborn hearing screening). Regardless of whether the infant was born at a hospital with a hearing-targeted screening protocol, parents lacked confidence in their knowledge of cCMV, the problems associated with the virus, and ways to minimize exposure to the virus. This study revealed the need for parent education of cCMV throughout the state of South Dakota.

Although education and behavioral change can prevent the transmission of maternal CMV infection, few studies have researched the most effective means through which to convey this message (Hughes et al., 2017; Revello et al., 2015; Vauloup-Fellous et al., 2009). Like cCMV, sudden infant death syndrome (SIDS), or the unexplained death of an infant younger than one year of age, is common among infants in the United States. In fact, SIDS is the third leading cause of infant mortality (Carrier, 2009). Another similarity between the two conditions is the role of cultural norms in slowing the change of certain behaviors. For example, kissing a child on the lips (a risk factor for CMV

infection) and placing a child to sleep on their stomach (a risk factor for SIDS) are common behavioral practices in the United States, and parents may be more resistant to change these behaviors (Thackeray, 2017).

Unlike cCMV, several studies have identified educational methods and mediums most effective for improving parents' knowledge of SIDS and safe sleep practices. For example, providing parents with verbal and written education on SIDS has been shown to increase parental knowledge of SIDS and compliance to safe sleep practices; modeling safe sleep practices in the hospital has also been shown to help (Burd et al., 2007; Dufer & Godfrey, 2017; Gelfer et al., 2013; Grael et al., 2010). Using written material alone as an educational method may be unsuccessful when attempting to change safe sleep behaviors (Moon & Omron, 2002).

The effectiveness of educational materials is also dependent on individuals' health literacy levels. Health literacy can be defined as "the degree to which individuals have the capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions" (Selden et al., 2000, p. vi). Studies have shown that health literacy levels are strongly related to economic status among various other demographic factors (Kirsch et al., 2002). As such, the readability of educational materials should be assessed when designing handouts and programs to accommodate various health literacy levels.

The purpose of this study was to investigate whether educational methods shown to be most effective for improving parents' knowledge of SIDS and safe sleep practices can also be used to improve South Dakota parents' knowledge of cCMV and preventative hygiene behaviors. The study was designed to be highly replicable by other researchers interested in improving parental knowledge of cCMV with written and verbal educational materials.

Method

Design

The Institutional Review Board (IRB) at the University of South Dakota approved this research project. Implied consent from the participants was inferred by voluntary completion of the study. G*Power software (version 3.1.9.4) was used for sample size calculations. A two-tailed *t*-test for dependent means with a moderate (0.5) effect size, 0.05 significance level, and 0.8 power determined the minimum sample size to be 34 participants.

Setting and Participants

The study was first piloted with parents whose children are enrolled in a local Head Start program to evaluate the study's design, to assess the efficacy of research materials, and to correct potential errors. Following the pilot study, researchers moved forward with the official study. The official study took place online and included a single group of research participants. Participants were recruited through a social media post on various public

and private Facebook groups, including those groups specifically designed for parents. Interested participants were required to be age 18 or older, a parent of a child younger than age 5, and a resident of the state of South Dakota. The study materials were designed to screen for these criteria prior to allowing participants full access to the study (see Project Development and Procedures). Data collection began on November 30, 2021 and was completed on December 14, 2021.

Project Development and Procedures

A cCMV educational program with both written and verbal components was created for this study. The written component consisted of a one-page handout with information on cCMV (see Appendix A), while the verbal component consisted of a 3-minute YouTube video with the researcher's voice-over narration. The video can be accessed at <https://youtu.be/UKsFGauCbvl>. These educational materials were approved by the program director of the National CMV Foundation.

Pre- and post-education surveys were disseminated via *Qualtrics* software (see Appendix B). The surveys consisted of the same ten questions and were used to assess parents' knowledge of cCMV both before and after viewing the educational materials. Parents were asked about their understanding of (c)CMV, modes of CMV transmission, and ways to prevent transmission. Readability statistics on Microsoft Word revealed both surveys to have a 10th grade Flesch-Kincaid reading level; when the words *congenital cytomegalovirus* were removed, this statistic changed to a 6th grade reading level. Since these words were necessary to include in the survey, the reading statistics were acceptable to researchers. In addition, demographic questions were included at the beginning of the pre-education survey. These questions required respondents to provide their age, gender, and educational level. Respondents were also asked if they were previously or currently employed in a healthcare profession; if they have other children at home; and if they have a child with cCMV.

The researchers designed a social media post and provided a single link for all study materials. Three questions were asked at the beginning of the study to ensure participants met the aforementioned inclusion criteria (i.e., age 18 or older, parent of a child younger than age 5, and resident of South Dakota). If participants answered "no" to any of these questions, the study ended. If participants answered "yes" to all three questions, they were given access to the pre-education survey. They were then automatically redirected to the YouTube video (verbal education component) and educational handout (written education component). Following the education session, participants were automatically redirected to the post-education survey. Finally, participants who completed the entire survey were asked if they would like their name entered into a drawing for a \$20 Walmart gift card. These gift cards were mailed to the participants after the study was completed.

This research design was modeled after a one-group, pre-post study completed by Dufer et al. (2017). Their study revealed that parents' knowledge of sudden infant death syndrome (SIDS) and safe sleep practices was significantly higher following a verbal and written educational session. Researchers hypothesized that verbal and written education of cCMV would lead to a change in parents' knowledge of the virus.

Statistical Analysis

Descriptive statistics (i.e., percentages) were used to compare the change in performance on each individual question between the group's pre- and post-knowledge surveys. In addition, a two-tailed, dependent *t* test ($p < 0.05$) was used to compare the overall change in the group's pre- and post-education survey performance. IP addresses were used to pair the participants' pre-education scores with their post-education scores. If an IP address was found in the pre-education results but not the post-education results, that data was eliminated from the analysis, as the participant did not complete the entire study.

Results

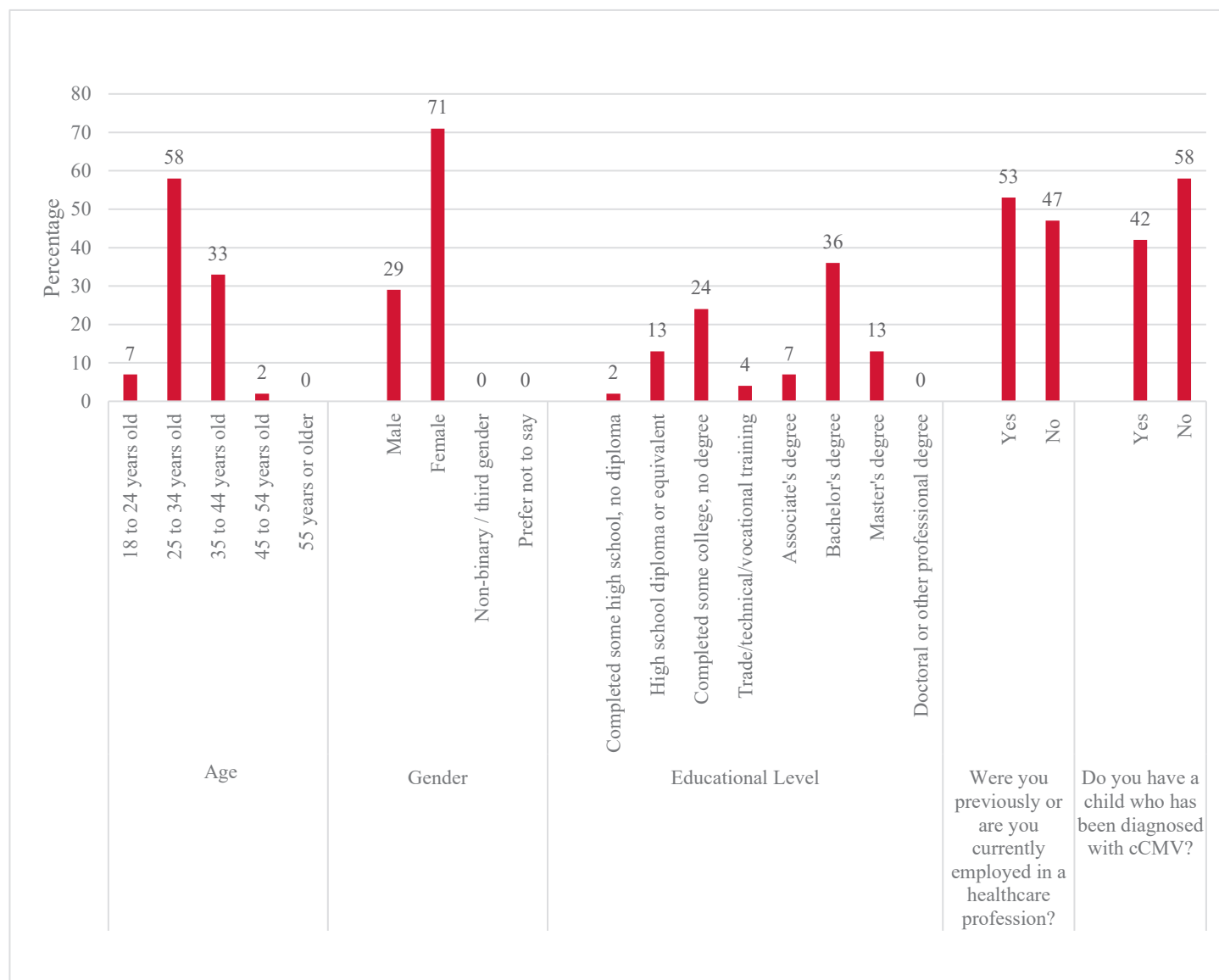
Forty-five participants completed the official study in its entirety. Partially complete responses were eliminated from the data analysis.

Demographic Factors

Answers to the demographic questions on the pre-education survey were analyzed (see Figure 1). The most commonly selected age range was 25 to 34 years old ($n = 26$, 58%), and the majority of participants identified as female ($n = 32$, 71%). When asked about their educational level, the most commonly selected option was bachelor's degree ($n = 16$, 36%), and the second most commonly selected option was "completed some college, no degree" ($n = 11$, 24%). When asked if they were previously or currently employed in a healthcare profession, 53% of participants responded with yes and 47% responded with no. Finally, when asked if they have a child who has been diagnosed with cCMV, 42% of participants selected yes and 58% selected no. The high percentage of participants who reported that their child has been diagnosed with cCMV may be due to the specific Facebook groups that shared the social media post to their pages.

Change in Performance on Individual Questions

For all ten questions included on the pre- and post-education surveys, the researcher used descriptive statistics to compare the change in performance on each individual question (see Table 1). Eight of the ten questions showed an increase in correct responses on the post-education survey as compared to the pre-education survey. Two questions (questions 8 and 9) showed a decrease in correct responses on the post-education survey as compared to the pre-education survey. Question 8 stated, "Cytomegalovirus (CMV) can spread from one person to another through..." and participants were required to pick from the following options: (a) saliva, (b) urine, (c) blood, or (d) all of the above. The correct answer

Figure 1*Analysis of Participants' Demographic Factors (n = 45)*

was all of the above, which 25 participants answered correctly on the pre-education survey and 20 participants answered correctly on the post-education survey. Question 9 asked, "When is a child with congenital cytomegalovirus (cCMV) at risk for developing hearing loss?" Participants were required to pick from the following options: (a) at birth, (b) sometime after birth, (c) both at birth and sometime after birth, or (d) none of the above. The correct answer was both at birth and sometime after birth, which 28 participants answered correctly on the pre-education survey and 27 participants answered correctly on the post-education survey.

The decrease in performance on these two questions may be explained by several factors. Participants may have fatigued toward the end of the study and spent less time reading the questions. It is also possible that some participants may not have completed the educational session in its entirety, which is a limitation of conducting an online study.

Overall Change in Performance

After reviewing the change in performance on each individual question, the researcher analyzed the overall change in performance on the post-education survey as compared to the pre-education survey. Data analysis revealed a statistically significant increase in scores on the post-education survey as compared to the pre-education survey ($p < 0.05$).

Discussion

The statistically significant increase in post-education scores as compared to pre-education scores supports the researchers' hypothesis that verbal and written education of cCMV would lead to a change in South Dakota parents' knowledge of the virus. Just as providing parents with verbal and written education on SIDS has been shown to increase parental knowledge of SIDS and compliance to safe sleep practices (Burd et al., 2007; Dufer & Godfrey, 2017; Gelfer et al., 2013), the same educational methods

Table 1
Change in Performance on Individual Quiz Questions

Question	# Correct Responses / Total Responses		Direction of Change
	Pre-Education Survey	Post-Education Survey	
1) Based on the number of children born with each condition per year, congenital cytomegalovirus (cCMV) is more common than most other conditions, except for...	$\frac{17}{45}$	$\frac{20}{45}$	↑
Correct answer: cCMV is more common than A, B, and C			
2) How many children are born with congenital cytomegalovirus (cCMV) each year?	$\frac{17}{45}$	$\frac{32}{45}$	↑
Correct answer: 1 in 200 infants			
3) True or False: An adult infected with cytomegalovirus (CMV) usually experiences severe symptoms.	$\frac{20}{45}$	$\frac{27}{45}$	↑
Correct answer: False.			
4) True or False: All symptoms of congenital cytomegalovirus (cCMV) infection can be seen at birth.	$\frac{23}{45}$	$\frac{26}{45}$	↑
Correct answer: False			
5) Which of the following options is the most common long-term health problem caused by a congenital cytomegalovirus (cCMV) infection?	$\frac{20}{45}$	$\frac{29}{45}$	↑
Correct answer: Hearing loss			
6) Women who are infected with cytomegalovirus (CMV) during pregnancy most commonly acquire the virus from...	$\frac{24}{45}$	$\frac{31}{45}$	↑
Correct answer: Children under the age of 3			
7) All the following activities place a pregnant woman at risk for cytomegalovirus (CMV) infection except for...	$\frac{26}{45}$	$\frac{32}{45}$	↑
Correct answer: Scooping a cat's litter box			
8) Cytomegalovirus (CMV) can spread from one person to another through...	$\frac{25}{45}$	$\frac{20}{45}$	↓
Correct answer: All of the above (i.e., urine, saliva, and blood)			
9) When is a child with congenital cytomegalovirus (cCMV) at risk for developing hearing loss?	$\frac{28}{45}$	$\frac{27}{45}$	↓
Correct answer: Both at birth and sometime after birth			
10) True or False: Practicing simple, healthy habits, such as hand washing, cannot reduce a pregnant woman's risk of being infected with cytomegalovirus (CMV).	$\frac{16}{45}$	$\frac{20}{45}$	↑
Correct answer: False			

may also be used to improve South Dakota parents' knowledge of cCMV. Future research should evaluate whether cCMV education may also be used to increase parental compliance to preventative hygiene behaviors.

Limitations must be considered when reviewing the results of this study. First, an online study requires a certain amount of trust in research participants. Researchers cannot guarantee that participants watched the educational video in its entirety or read through the educational handout. Creating more interactive handouts and/or videos may increase participation in the educational component. In addition, results may not be representative of the entire state of South Dakota. As the study was disseminated on a social media platform, the geographic location of participants could not be controlled. Some regions of the state may be overrepresented in the data while other regions may be underrepresented, and participants' demographic factors (e.g., 42% report having a child diagnosed with cCMV and 53% report working in the healthcare field) could bias test results. Selecting a participant recruitment strategy more inclusive of geography and demographic factors may change future test results.

Conclusion

Educational methods shown to be most effective for improving parents' knowledge of SIDS (i.e., written and verbal education) can also be used to improve South Dakota parents' knowledge of cCMV. This finding has implications for future education of parents on cCMV, problems associated with the virus, and ways to minimize exposure to the virus. Further research will be needed to assess the efficacy of these educational materials on parents residing in other geographic regions. In addition, future research should assess the change, if any, in parental compliance to preventative hygiene measures both before and after receiving education on cCMV.

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cCMV Handout

FACT SHEET

cCMV

Congenital Cytomegalovirus

What is cCMV?

Congenital cytomegalovirus (cCMV) is one of the most common conditions present at birth, having a higher incidence rate than Down syndrome, spina bifida, and fetal alcohol spectrum disorder. It is also the leading non-genetic cause of hearing loss in infants.

Most adults have been infected with CMV, and symptoms often mimic the common cold.

However, if CMV passes from a pregnant mother to her child in utero, the infant is born with congenital cytomegalovirus (cCMV). Unlike CMV in adults, cCMV in infants may cause

long-term health problems:

- Hearing loss (at birth or delayed onset)
- Vision loss
- Microcephaly
- Seizure disorders
- Cognitive impairment

**Not all symptoms are visible at birth.*

How is CMV spread?

CMV is spread through bodily fluids, such as saliva, blood, and urine. Women who acquire CMV during pregnancy most commonly acquire the virus from a child younger than three years of age. Therefore, women who are frequently in contact with young children (e.g., nurses, daycare providers, teachers, women with young children of their own) and who are or plan to become pregnant should adhere to **hygienic precautions**.



Do not share food, utensils, drinks, or straws.



Do not put a pacifier in your mouth.



Avoid kissing a child directly on the lips.



Do not share a toothbrush.



Wash your hands after changing diapers or handling kids' toys.

Information from the
National CMV Foundation

nationalcmv.org



1 in 200

infants in the U.S. is born with
congenital CMV infection.

1 in 5

infants with congenital CMV
infection will have long-
term health problems.

Hearing loss is the most
common symptom.

Only **9% of women** and **5% of men** in the
United States know about congenital CMV!



Appendix B

Surveys

Demographic questions (only included on pre-education quiz):

- ☐ Age
 - ☐ Under 17 years old
 - ☐ 18 to 24 years old
 - ☐ 25 to 34 years old
 - ☐ 35 to 44 years old
 - ☐ 45 to 54 years old
 - ☐ 55 years or older
- ☐ Gender: How do you identify?
 - ☐ Female
 - ☐ Male
 - ☐ Non-binary/third gender
 - ☐ Prefer not to say
- ☐ Educational Level
 - ☐ Completed some high school, no diploma
 - ☐ High school diploma or equivalent
 - ☐ Completed some college, no degree
 - ☐ Trade/technical/vocational training
 - ☐ Associate's degree
 - ☐ Bachelor's degree
 - ☐ Master's degree
 - ☐ Doctoral or other professional degree
- ☐ Were you previously or are you currently employed in a healthcare profession?
 - ☐ Yes
 - ☐ No
- ☐ Do you have other children at home younger than age 18?
 - ☐ Yes
 - ☐ No
- ☐ Do you have a child who has been diagnosed with congenital CMV?
 - ☐ Yes
 - ☐ No

Appendix B (cont.)

Quiz Questions (included on both pre- and post-education surveys):




Answers in bold text

1. Based on the number of children born with each condition per year, congenital cytomegalovirus (cCMV) is more common than most other conditions, EXCEPT FOR:
 - a. Down syndrome
 - b. Spina bifida
 - c. Sudden Infant Death Syndrome (SIDS)
 - d. **cCMV is more common than A, B, and C.**
2. How many children are born with congenital cytomegalovirus (cCMV) each year?
 - a. 1 in 10 infants
 - b. 1 in 1000 infants
 - c. **1 in 200 infants**
 - d. 1 in 500 infants
3. An adult infected with cytomegalovirus (CMV) usually experiences severe symptoms.
 - a. True
 - b. **False**
4. All symptoms of congenital cytomegalovirus (cCMV) infection can be seen at birth.
 - a. True
 - b. **False**
5. Which of the following options is the most common long-term health problem caused by a congenital cytomegalovirus (cCMV) infection?
 - a. Jaundice
 - b. Kidney disease
 - c. Vision loss
 - d. **Hearing loss**
6. Women who are infected with cytomegalovirus (CMV) during pregnancy most commonly acquire the virus from:
 - a. Teenagers
 - b. Children ages 5 to 7
 - c. **Children under the age of 3**
 - d. College students
7. All the following activities place a pregnant woman at risk for cytomegalovirus (CMV) infection EXCEPT FOR:
 - a. Not washing your hands after picking up children's toys
 - b. **Scooping a cat's litter box**
 - c. Putting a used pacifier in your mouth
 - d. Kissing a child on the lips

Appendix B (cont.)

8. Cytomegalovirus (CMV) can spread from one person to another through:
- Saliva
 - Urine
 - Blood
 - All of the above**
9. When is a child with congenital cytomegalovirus (cCMV) at risk for developing hearing loss?
- At birth
 - Sometime after birth
 - Both at birth AND sometime after birth**
 - None of the above
10. Practicing simple, healthy habits, such as hand washing, cannot reduce a pregnant woman's risk of being infected with cytomegalovirus (CMV).
- True
 - False**

EHDInfo



The goal of the congenital CMV Public Health and Policy Conference (CMV-PHP) is to present the latest research on diagnosis and treatment, raise awareness, delineate prevention efforts, provide information about early intervention options, and disseminate family support resources in an effort to reduce the number of babies born with CMV and connect families affected by CMV with the resources they need to improve their quality of life.

With your help, we can reduce the number of babies born with CMV!

REGISTER NOW

The Signed Linguistic Input of Mothers with Typical Hearing to Children who are Deaf or Hard of Hearing

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Abstract

This study investigated the distribution of ten facilitative language techniques (FLT) in the linguistic input of mothers with typical hearing who use sign language with their children who are deaf or hard of hearing (DHH). Four mothers with typical hearing and their children who are DHH under the age of three participated in six, ten-minute caregiver-child interaction sessions via Zoom. The recorded sessions were coded for mother FLT and child utterances. Results indicated that the mothers tended to use more initiative than responsive types of FLT, consistent with findings of previous studies that examined the input of mothers who were using spoken-only language with their children who are DHH. Additionally, the mothers tended to use combined signed and spoken input more frequently than signed language alone. These findings point to the need for focused intervention to increase the use of responsive and linguistically stimulating FLT by caregivers with typical hearing who use sign language. Findings also suggest that caregivers with typical hearing may need more ongoing support to learn to use American Sign Language (ASL) effectively with their children.

Keywords: deaf or hard of hearing, early intervention, caregivers, facilitative language techniques

Acronyms: ASL = American Sign Language; DHH = deaf or hard of hearing; FLT = facilitative language techniques

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Caregivers are a child's earliest and most important communicative partners. Language is a social transaction, and language develops via social interactions (Tomasello, 1992; Vygotsky, 1978). Therefore, a child's caregivers have a large impact on the child's acquisition of language. Both the quantity and quality of adult language exposure and interaction impacts the child's language outcomes (Gilkerson et al., 2018; Hart & Risley, 1995; Hoff-Ginsberg, 1986; Rowe, 2008). Although children who are deaf or hard of hearing (DHH) with caregivers who are deaf usually acquire language similarly to their peers with typical hearing (Freel et al., 2011; Newport & Meier, 1985), children who are DHH with parents who are hearing are at risk for language delay due to a lack of exposure to accessible language in their environment. Caregiver/child interaction has been found to be impacted by hearing status mismatch. Caregivers with typical hearing with children who are DHH have been found to be less responsive and more directive, produce shorter utterances, and engage in less coordinated joint attention compared to dyads who share a hearing status, even in children with moderate hearing levels and amplification (Dirks & Rieffe, 2019; Fagan et al., 2014; Meadow-Orlans & Spencer,

1996; Spencer, 2000). This disruption in caregiver-child interaction could contribute to difficulties in language acquisition for children who are DHH.

Several studies have investigated the impact of caregiver hearing status and/or communication mode on caregiver-child interaction. For example, Meadow et al. (1981) reported differences in interaction styles among hearing mother/hearing child, deaf mother/deaf child, hearing mother/deaf child using spoken-only communication, and hearing mother/deaf child using combined signed and spoken communication. The dyads with matched hearing status (hearing/hearing and deaf/deaf) did not differ significantly in their interaction or conversational styles. Mothers with typical hearing who used spoken only communication with their children who were DHH interacted less and exhibited the least mature interaction styles compared with the three other groups. However, both hearing/deaf groups exhibited significantly more mother-initiated communication episodes compared to the groups with a hearing status match. It is important to note, however, that this study was conducted before universal newborn hearing screening was in place; consequently, children in this study were not identified with hearing loss

until the second year of life and may not have received the same type of family-centered early intervention that families benefit from in the present day.

Caregivers who are deaf and use sign language have been found to engage in more episodes of intersubjectivity, defined by Loots et al. (2003) as “an interaction state of shared involvement in a reciprocal exchange” (p. 405), compared to caregivers who are hearing with children who are DHH. However, caregivers with typical hearing who used combined signed and spoken language with their children who were DHH engaged in more episodes of intersubjectivity than caregivers with typical hearing using auditory/oral only communication (Loots et al., 2005). Further, parents who are deaf have been found to employ certain visual-tactile communication strategies—such as tapping the child to gain attention, signing on the child’s body, and using the child’s hands to form a sign—more often than parents who are hearing do with their children who are DHH. Not surprisingly, caregivers with typical hearing who use combined speech and sign practice more visual-tactile communication with their children who are DHH than caregivers with typical hearing who use auditory-oral only (Loots & Devisé, 2003). Results of these studies indicate that disruption in caregiver/child interaction caused by hearing status mismatch may be mitigated to some degree by the caregiver’s use of visual language.

Other research has examined the spoken language input of caregivers with typical hearing to their children who are DHH. DesJardin (2006) described the distribution of ten facilitative language techniques (FLT) used by hearing mothers of young children (ages 25–72 months) who were DHH. This study found that maternal use of techniques such as recast and open-ended questions, which facilitate conversational interaction, were positively associated with child language. On the other hand, maternal use of label and directive, techniques which do not encourage conversation, were negatively associated with child language. Other studies found similar results, with FLT termed as *higher-level* (those which are useful to facilitate language growth in children who are producing single word utterances) demonstrating positive associations with child language, and FLT termed as *lower-level* (those which are employed to facilitate language growth of children at the pre-linguistic level) demonstrating negative associations with child language (DesJardin et al., 2009; DesJardin & Eisenberg, 2007). Further, parents with typical hearing use more lower-level FLT with their children who are DHH compared to children who have typical hearing, and they may not intuitively adapt their input to their child’s needs in the same way as caregivers of children with typical hearing (DesJardin et al., 2014, 2017). Additional studies have demonstrated that hearing caregivers use more directive spoken language and provide less high-quality responsive input to their children who are DHH (Ambrose et al., 2015; Dirks et al., 2020; Su & Roberts, 2019).

Fewer studies have investigated the linguistic content of the signed input used by mothers to their children who are DHH. Fieldsteel et al. (2020) analyzed the signed linguistic input of mothers who are deaf to their children

who are deaf ranging in age from 21–39 months. The researchers found that verbs were used significantly more often than nouns, and that mothers were noted to use pointing for both linguistic (as pronouns) and attention getting purposes. Additionally, the mothers exhibited an average mean length of utterance (MLU) of 2.4, with most utterances consisting of 2–3 signs. This study was the first to report on the content and form of language used by signing mothers and provides valuable information about American Sign Language (ASL) input from native signers to their young children.

Recent research points to the benefit of caregivers with typical hearing using sign language with their children who are DHH. A study of young children who are DHH and have caregivers with typical hearing found that children who were exposed to ASL early (before the age of 6 months) had vocabulary comparable to children who are DHH with caregivers who are deaf and communicate with sign language. These findings indicate that caregivers with typical hearing have the potential to be good sign language models for their children, even if they are not yet fluent themselves (Caselli et al., 2021).

Another recent study found a positive association between American Sign Language (ASL) vocabulary and spoken English vocabulary in children ages five and younger who are DHH, even in families with caregivers who have typical hearing (Pontecorvo et al., 2023). These studies indicate that it could be advantageous for children who are DHH to have sign language exposure from their caregivers with typical hearing; however, there is a paucity of research exploring the signed linguistic input of caregivers with typical hearing. To date, studies investigating the content and quality of language input of mothers who have typical hearing to children who are DHH has focused almost exclusively on spoken language. The present study seeks to address this gap by exploring the distribution of FLT in the linguistic input of four mothers with typical hearing who sign. Further, the study explores the relationship of caregiver language modality (spoken vs. signed) with FLT use.

Methods

Participants

Following ethical approval from the supporting institution’s institutional review board, participants were recruited from two southeastern states’ statewide early intervention programs and via the social media platforms of the supporting institution. Families were eligible to participate in the study if they had a child between the ages of 9 and 36 months with an identified permanent childhood hearing loss and if they were using sign language with the child. The use of spoken English in addition to sign was acceptable, as long as the family reported active sign language use with the child as well. Families who completed the study in its entirety received \$20 in compensation for their time.

The study enrolled four participant dyads, all of which included mothers with typical hearing who used combined signed and spoken language with their children. All

mothers self-reported as hearing, rated themselves at “beginner” level ASL fluency, and were current or former participants in ASL lessons with a trainer who was deaf. Each child participant is identified by a pseudonym. Candice was a white female who was 21 months old at the time of enrollment in the study. Her mother reported that Candice has a moderately severe hearing loss in the right ear and a severe hearing loss in the left ear. She was fit with amplification at 6 months of age and currently uses a hearing aid in the right ear and a cochlear implant in the left ear. Both of her parents hold four-year college degrees. David was a Hispanic male who was 19 months old at the time of enrollment in the study. His mother reported that David has a severe hearing loss in the left ear and a profound hearing loss in the right ear. He was fit with amplification at 11 months and currently uses bilateral cochlear implants. Both of his parents hold two-year college degrees. Eve was a white female who was 24 months old at the time of enrollment in the study. Her mother reported that Eve has a mild hearing loss in the right ear and does not use amplification. Eve’s mother also reported that she has additional developmental delays. Her mother holds a master’s degree, and her father holds a four-year college degree. Kevin was a white male who was 35 months old at the time of enrollment in the study. His mother reported that he has a mild to moderate hearing loss in both ears and does not currently use amplification. Educational level was not reported for Kevin’s caregivers. Mothers of Candice, David, and Eve indicated that their child stayed at home full-time with them or their partner, while Kevin attended preschool daily.

Data Collection

Dyads participated in six 10-minute caregiver/child interaction sessions using Zoom. The mothers were instructed to gather a selection of their child’s preferred toys, books, and activities ahead of the session. During the session, they were instructed to play with their child as they normally would. The researcher’s camera and microphone were turned off during recording to minimize distractions to the dyad. Video data were coded using the Behavioral Observation Research Interactive Software (BORIS; Friard & Gamba, 2016), an open-source video coding software that allows for point-by-point behavioral coding of video data.

Coding

The videos were coded for mothers’ use of ten facilitative language techniques (FLT) adapted from those defined by Cruz et al. (2013) and DesJardin (2006). Cruz et al. (2013) and DesJardin (2006) stratified the FLT) as lower-level FLT) (linguistic mapping, comment, imitation, label, close-ended question, and directive) and higher-level FLT) (narration, open-ended question, expansion, and recast). In addition to the higher-level/lower-level categories, some FLT) may be viewed as *initiative*, meaning that they are employed by the caregiver to direct or initiate conversation in

the absence of a child utterance. Other FLT) may be viewed as *responsive*, meaning that they are employed in response to a child utterance. See Table 1 for descriptions of the FLT). Each mother FLT) was also coded as one of three modalities: spoken only, simultaneously spoken and signed, or signed only.

Table 1
Facilitative Language Techniques

Facilitative Language Technique (FLT)	Definition
Responsive	
Linguistic mapping (LM)	Coded when the caregiver interprets into words a child vocalization that is not recognizable as a word
Comment (CM)	Coded when the caregiver responds verbally to acknowledge a child utterance
Imitation (IM)	Coded when the caregiver repeats a child’s vocalization verbatim
Expansion (EP)	Coded when the caregiver repeats the child’s vocalization, adding one or more morphemes or words to provide a more grammatically complete model and/or add new information to the child’s vocalization
Recast (RC)	Coded when the caregiver repeats the child’s vocalization, adding one or more morphemes or words, and rephrasing it into a question
Initiative	
Label (LB)	Coded when the caregiver produces a verbal statement to name something
Directive (DIR)	Coded when the caregiver tells child to do or not to do something
Close-ended question (CEQ)	Coded when the caregiver asks the child a question that has a short, specific (usually one-word) response
Narration (NR)	Coded when the caregiver uses either parallel talk or self-talk, to describe what they or the child are doing or thinking
Open-ended question (OEQ)	Coded when the caregiver asks the child a question that does not have a specific, one-word answer

Child utterances were also coded as one of four different types of productions. *Word* was coded when the child produced an utterance that was recognizable by the coder as a true word; *babble* was coded when the child produced an utterance that was not recognizable as a word; *gesture* was coded when the child produced a non-verbal communicative movement such as a reach or show; and *reflexive* was coded when the child produced a non-linguistic vocalization such as a grunt, sigh, cry, or laugh. Each child utterance coded as word or babble was also coded as one of three modalities: spoken only, simultaneous spoken and signed, or signed only.

Utterances coded as gesture or reflexive were not coded for modality, because by definition, gestures could only be manual, and reflexives could only be spoken.

Results

Figure 1 represents the mothers' and children's utterances by modality (spoken, signed, or both simultaneously). All mothers produced more utterances than their children, and all children and three out of four mothers (David, Eve, and Kevin) used spoken language most frequently. Candice's mother used simultaneous signed and spoken language most frequently.

Figure 1
Facilitative Language Techniques

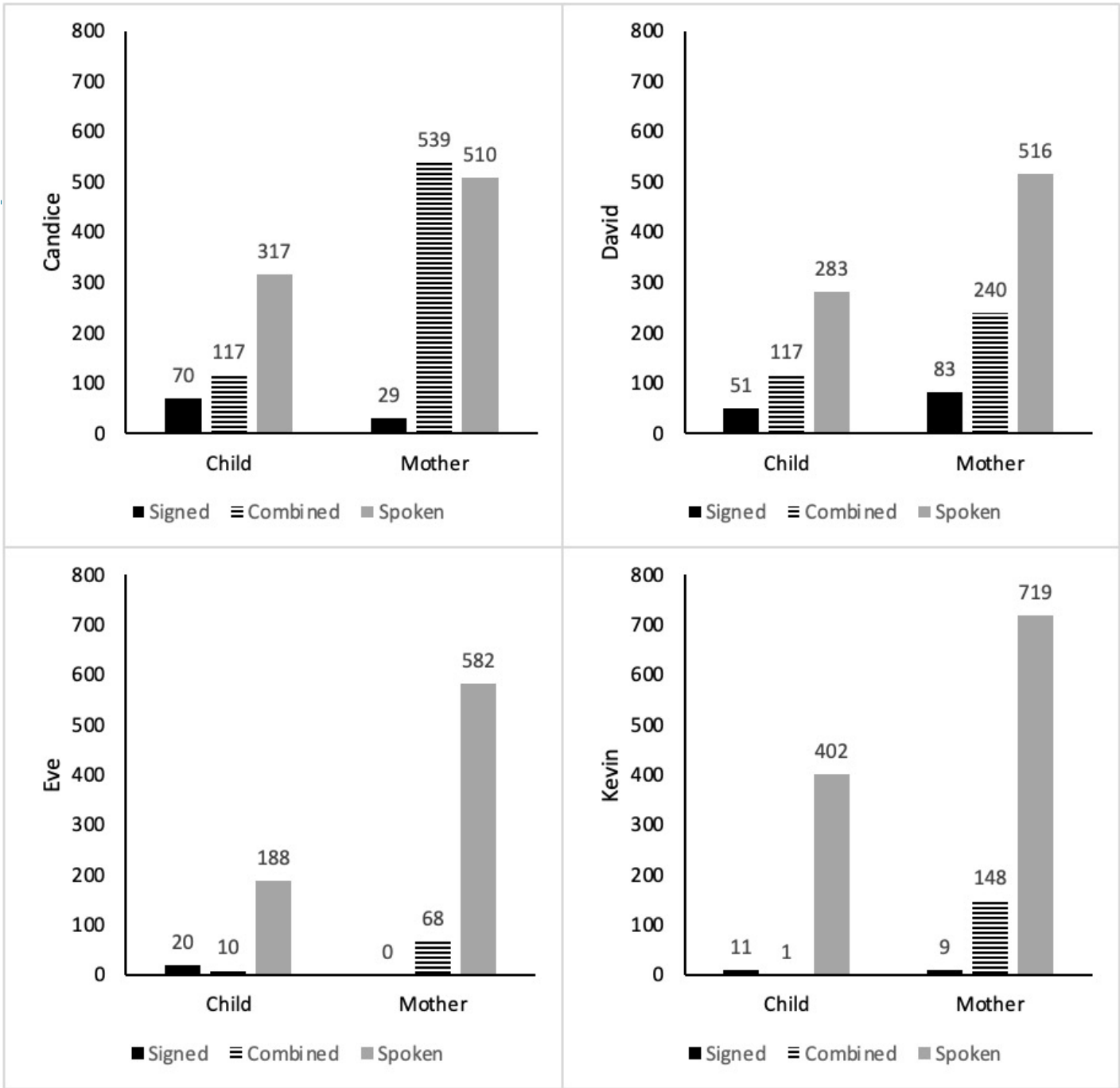
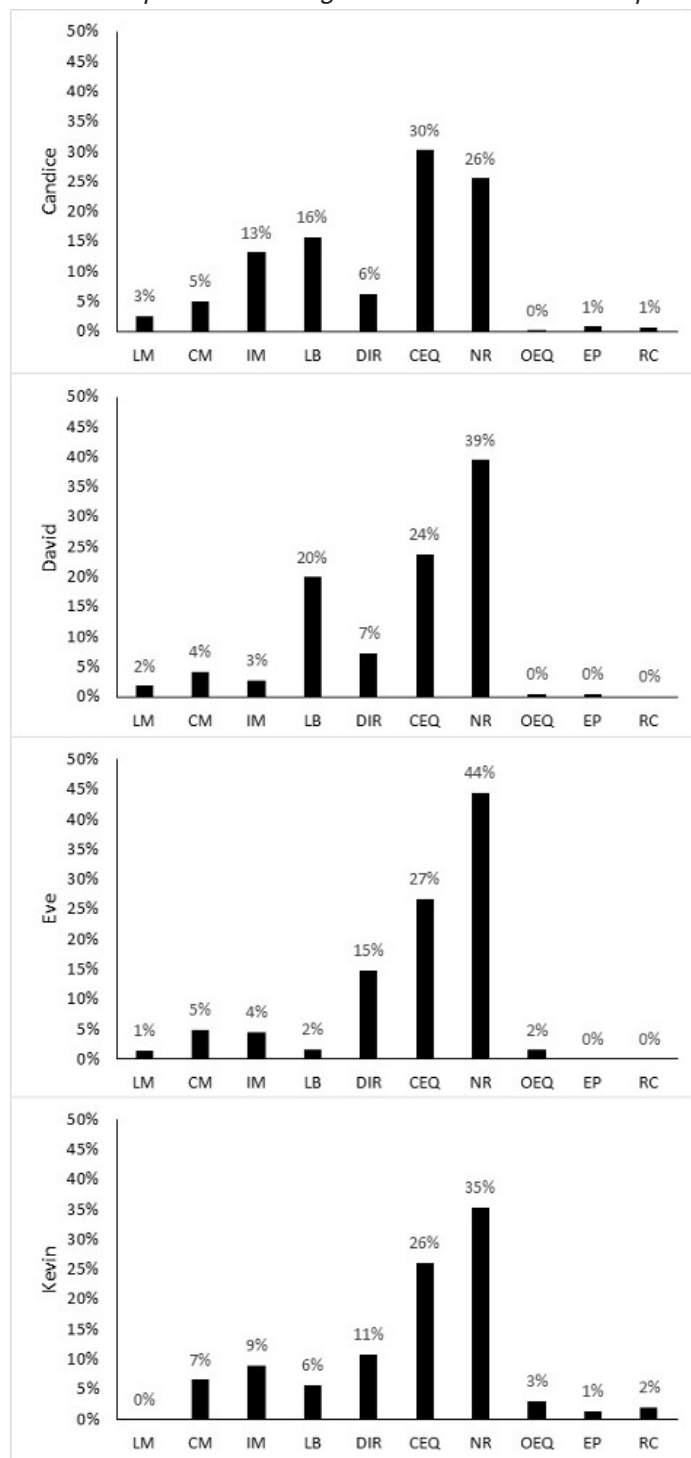


Figure 2 illustrates the overall distribution of FLTs in the mothers' samples. In both Candice's and David's samples, the caregiver initiated FLTs of close-ended question, narration, and label occur most frequently. In Eve's and Kevin's samples, the caregiver initiated FLTs of close-ended question, narration, and directive occurred most frequently.

Figure 2

Overall Proportion of Caregiver FLTs Across the Samples



Note. LM = linguistic mapping, CM = comment, IM = imitation, LB = label, DIR = directive, CEQ = close-ended question, NR = narration, OEQ = open-ended question, EP = expansion, RC = recast

Figure 3 illustrates the caregivers' FLT use by modality (spoken, signed, or both simultaneously). Candice's mother used spoken language only for most occurrences of narration, close-ended question, comment, and directive, and she used simultaneous spoken and signed language for most occurrences of label, imitation, linguistic mapping, expansion, and recast. David's mother used spoken language only for most occurrences of narration, directive, linguistic mapping, and imitation, and she used simultaneous sign and spoken language for most occurrences of close-ended question, label, comment, and expansion. Eve's caregiver used spoken language only for most occurrences of all FLTs. Kevin's caregiver used spoken language only for most occurrences of all FLTs except label, for which most occurrences were simultaneous sign and spoken language.

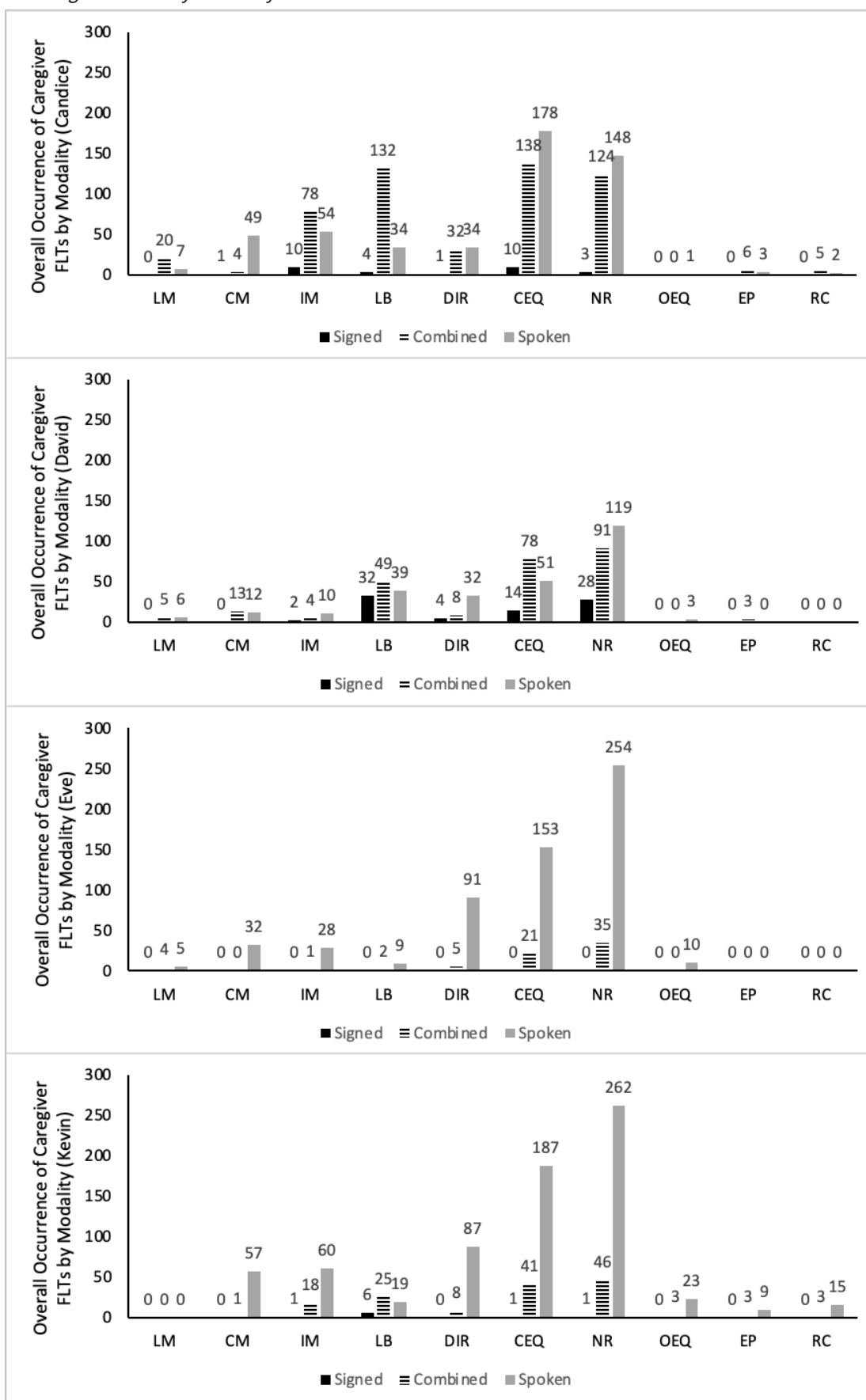
Figure 4 represents the mothers' responses and initiations to their children by modality (spoken only or signed). In this figure, signed only and simultaneous spoken/signed utterances were collapsed into one group representing signed utterance. This was due to the relatively low occurrence of signed only utterances (ranging from 0–10% of linguistic input) across all mothers' samples. All mothers produced initiative FLTs much more frequently than responsive FLTs, with initiative FLTs accounting for 78–91% of the caregivers' samples. Candice's and David's mothers used signed and spoken-only language input at similar rates for both responses and initiations. In Candice's sample, 51% of her mother's responses were signed and 49% were spoken-only, while 53% of her mother's initiations were signed and 47% were spoken-only. In David's sample, 49% of his mother's responses were signed and 51% were spoken-only, while 52% of his mother's initiations were signed and 48% were spoken-only. Eve's and Kevin's caregivers used spoken-only input much more frequently than signed input for both responses and initiations. In Eve's sample, 7% of her mother's responses were signed and 93% were spoken-only, while 11% of her mother's initiations were signed and 89% were spoken-only. In Kevin's sample, 16% of his mother's responses were signed and 84% were spoken-only, while 18% of his mother's responses were signed and 82% were spoken-only.

Discussion

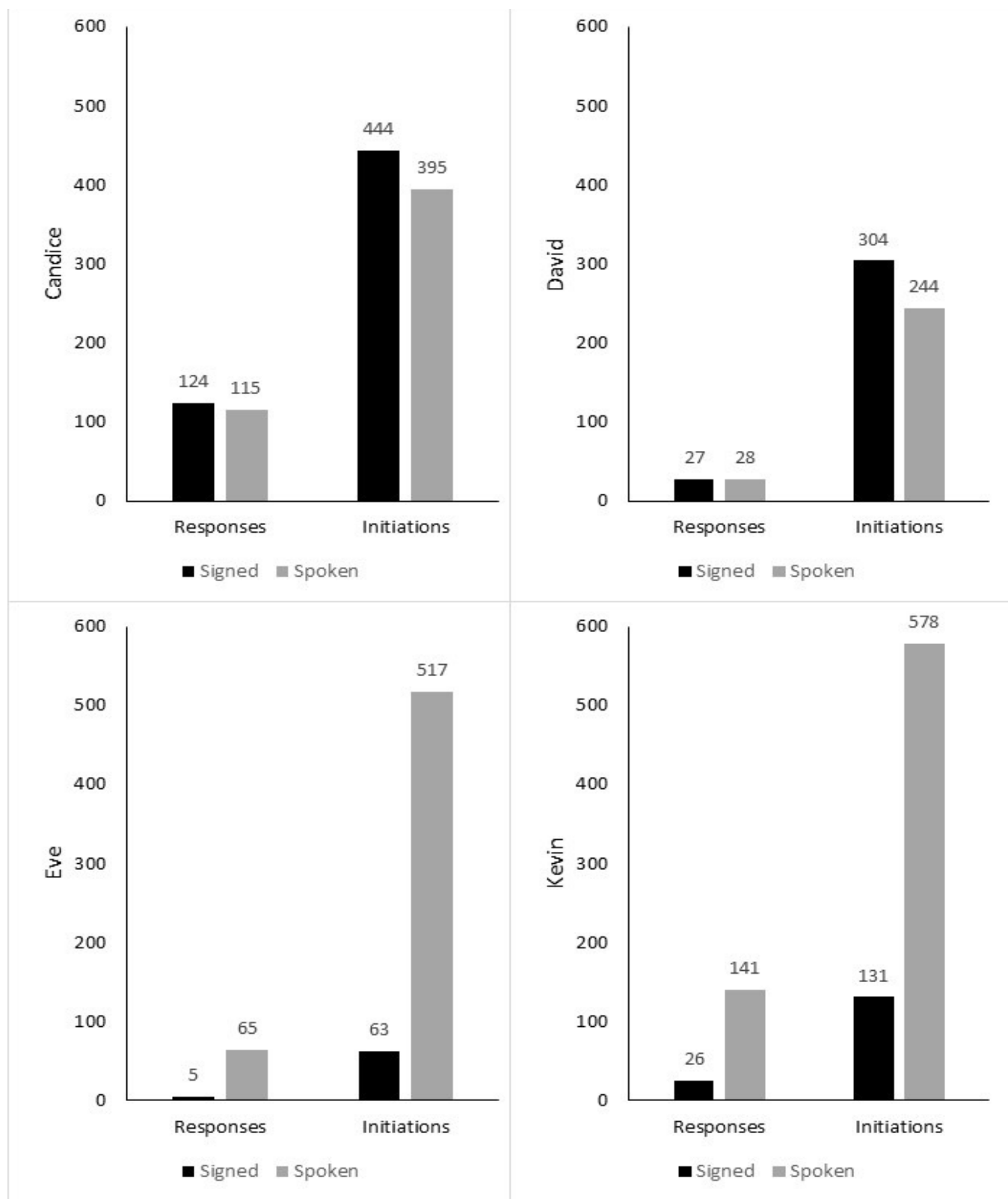
Most (three out of four) mothers used spoken-only language most frequently (Figure 1). All mothers used signs in conjunction with their spoken input more frequently than signed-only language. Candice and David also used combined sign/speech more often than signs alone. Eve and Kevin used predominantly spoken language, but produced signs alone more frequently than signs combined with spoken utterances. All mothers in this study were hearing, non-native signers who rated themselves as beginner level fluency. Each had received ASL support from a Deaf adult mentor to assist their ASL development. Although some instances of basic ASL sentences were noted, most of the mothers' signed utterances consisted of key words used for sign-supported speech. All children also used spoken-only language

Figure 3

Proportion of Caregiver FLTs by Modality



Note. LM = linguistic mapping, CM = comment, IM = imitation, LB = label, DIR = directive, CEQ = close-ended question, NR = narration, OEQ = open-ended question, EP = expansion, RC = recast

Figure 4*Caregiver Responses and Initiations by Modality*

most frequently, with spoken-only utterances comprising 63–97% of children's samples. Eve and Kevin used sign infrequently, but when they did, tended to produce signed rather than combined sign/speech utterances. However, signed utterances made up only about 14% of Eve's and about 3% of Kevin's total utterances. Signed

utterances made up about 37% of both Candice's and David's samples, and both children produced combination signed/spoken utterances more frequently than sign-only utterances. It is unclear if the predominance of spoken only productions in both mothers' and children's samples is due to mothers mirroring the child's most frequently used

modality, or vice versa; however, it is likely that there is a bidirectional influence of mother and child on each other's productions.

The overall distribution (including all productions, both signed and spoken) of FLT in the mothers' samples (Figure 2) indicates that the mothers tended to use initiative-type FLT such as narration, close-ended question, label, and directive the most frequently. This is consistent with findings from previous studies which show that parallel talk (narration), directive, and close-ended question were among the most frequently used FLT of hearing caregivers using spoken language with their children who are DHH (Cruz et al., 2013; DesJardin & Eisenberg, 2007). This indicates that these mothers with typical hearing who use sign-supported speech demonstrate patterns of linguistic input similar to mothers with typical hearing who use spoken language only with children who are DHH.

All mothers used spoken language only when employing narration and directive (Figure 3). Directive is an utterance telling the child to do or not to do something and narration is a one-sided production in which the parent describes what she or the child is doing without expectation for response. These two FLT are arguably two of the most controlling types, as they neither respond to nor invite child utterances. Even Candice's and David's mothers, who used signed input at a high frequency, were more likely to use spoken language for these more controlling utterance types. Techniques such as self- and parallel-talk are often taught as ways to increase children's exposure to language (Hearing First, n.d.). However, research indicates that children who are DHH benefit from caregiver linguistic input that is responsive and balanced in turns (Glanemann et al., 2013; Nicastrì et al., 2021; Roberts, 2019), and may be hindered in language development by caregiver input that is overly directive or intrusive (Ambrose et al., 2015; Vohr et al., 2010). Therefore, although initiative techniques may have value for children's language development, overuse of these strategies may be detrimental if they prevent caregivers from balancing turns and responding to their child. Three out of four mothers (Candice, David, and Kevin) used sign or sign supported speech most frequently when employing label, which is a single word or short utterance to provide a name or description for an item, person, or action. (Eve's mother used label at a level low frequency altogether.) This may be a function of the mothers' ASL fluency levels, as it is possible that mothers felt more comfortable signing this FLT, which is simpler in nature compared to a more complex utterance like narration.

Finally, all mothers used initiations much more frequently than responses in both spoken only and sign supported productions. These findings agree with findings from previous studies which indicate that mothers with typical hearing tend to initiate often, be more directive, and be less responsive in their interactions with their children who are DHH (Ambrose et al., 2015; Fagan et al., 2014; Meadow et al., 1981; Meadow-Orlans & Spencer, 1996; Su & Roberts, 2019). This extends extant literature by

demonstrating that mothers with typical hearing who use sign are similarly directive in their linguistic input, whether that input is signed, spoken, or both.

Limitations and Future Directions

The findings presented here represent an investigation of the distribution of FLT within samples of mothers with typical hearing who use sign with their children who are DHH and is the first to the author's knowledge to investigate these FLT in sign supported language samples. There are several limitations to be considered. First, the small sample size of this preliminary study precludes tests of statistical significance and limits generalizable conclusions from being drawn from the data. However, the data illustrate the feasibility of coding these FLT in signed samples and represent elements of mother-child signed interaction that may be expounded upon with a larger sample in future studies. Second, this study focused solely on FLT used by mothers to the exclusion of other elements of interaction previously discussed in the literature between signing mothers and children, such as signing on the child's body and visual attention getting strategies (e.g., Loots & Devisé, 2003; Waxman & Spencer, 1997). The omission of analysis of these behaviors in the present study may have overlooked some responsive actions which were not the focus of this study. Additionally, there are limitations related to the characteristics of the participants themselves. For example, it is unknown how long Kevin has been attending preschool or how he communicates while at school, and Eve was reported to have developmental disabilities which might impact her communication as well as her mother's responses to her. Each child's daily usage of their hearing technology and their audibility and access to spoken language is also unknown. It is unknown what other intervention services the families and children may have been receiving, aside from support from their Deaf Mentor, such as spoken language or sign language early intervention services. Further, caregivers did not indicate their desired long-term communication outcomes for their children and did not report their ratio of spoken to sign language use in the home.

Finally, all mothers who enrolled in this study were hearing, with limited ASL experience and fluency. The linguistic input of novice signing mothers is naturally different from that of mothers who are deaf or even mothers with typical hearing who are fluent signers. Therefore, this study can only speak to the linguistic input of this limited sample. Further, the mothers in this study exhibited limited ASL usage, tending instead toward signing key words and short phrases concurrently with their spoken utterances. As such, the study cannot make assumptions about the interactions of mothers who are deaf or have typical hearing who are providing more consistent ASL input to their children. Future studies should investigate the distribution of FLT in the linguistic input of mothers who are fluent in signing to their children who are DHH. Additionally, further investigations are planned to explore the effects of an intervention to increase the use of responsive FLT used by mothers who sign.

Clinical Implications

The findings of the present study have implications for clinical practice in early intervention for families of children who are DHH. The results indicate that, like hearing caregivers with typical hearing who use spoken language, caregivers with typical hearing who use signed language tend to be more controlling than responsive in their interactions with their children who are DHH. Intervention providers could provide education on language development and implement direct training of parents to use more responsive, linguistically stimulating techniques including linguistic mapping and expansion/recast, techniques which occurred at low frequency in each of the coded samples. Additionally, although each of these mothers reported participation in one-on-one ASL learning experiences with Deaf mentors, they all rated themselves as beginner level fluency. Most of the mothers' signed input was produced in the form of key words produced simultaneously with spoken language, rather than in ASL productions. This indicates a need for more support for caregivers with typical hearing to implement ASL with their children who are DHH.

Conclusions

To the author's knowledge, this study represents the first investigation of FLT types within a sample of mothers with typical hearing who sign to their children who are DHH. The findings indicate that the mothers, who rated themselves at a beginner level of ASL fluency and tended to use simultaneous sign and spoken productions, exhibited many similarities compared to previous studies of caregivers with typical hearing using spoken-only input. For example, the mothers in this study used predominantly initiative FLT types and most frequently employed narration, close-ended question, and directives in their input to their children. Results of this analysis suggest the need for focused intervention to increase use of responsive and linguistically stimulating FLT types within this population.

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Early Experiences of Parents of Children who are Deaf or Hard of Hearing: Navigating through Identification, Intervention, and Beyond

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Abstract

Guidelines created by the Joint Committee on Infant Hearing (JCIH, 2019) were designed to aid in the early identification of infant hearing loss. Despite these guidelines, a quarter of children who do not pass their initial screening are lost to the follow-up process and many more do not receive care in line with the 1-3-6 guidelines (Centers for Disease Control and Prevention [CDC], 2018; JCIH, 2019). To acquire more information about the experiences of families and identify specific barriers to timely diagnosis and intervention, interviews were conducted with 13 parents of children who are deaf or hard of hearing whose children were enrolled in a larger longitudinal study. These interviews revealed common themes regarding delayed identification, frustrations about timely intervention, and confusion when choosing communication modalities. Common themes amongst families who felt well-supported were also identified.

Keywords: Parent perspectives, hearing loss, qualitative design, hearing screening

Acronyms: ASL = American Sign Language; CI = cochlear implant; EHDI = Early Hearing Detection and Intervention; HA = hearing aid; JCIH = Joint Committee on Infant Hearing

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Early identification is key to minimizing potential language delays in children who are deaf or hard of hearing. Children who are deaf or hard of hearing exhibit deficits in oral language compared to their typical hearing peers (Tomblin et al., 2015), especially in the preschool years (Lund, 2016; Werfel et al., 2022). Delays in early oral language skills (i.e., vocabulary) can improve with amplification and therapy (Moeller et al., 2010; Yoshinaga-Itano et al., 2017). Even as children with hearing loss eventually develop age-appropriate language skills (Ching & Leigh, 2020), deficits in literacy skills remain (Camarata et al., 2018; Nittrouer et al., 2018). Early identification of hearing loss has been associated with improved language (Ching et al., 2017; Fulcher et al., 2012; Grey et al., 2021) and literacy skills (Pimperton et al., 2016). Despite this support for early hearing loss identification and intervention, many children are still not receiving amplification or being enrolled in intervention by the recommended six months of age (Grey et al., 2021; Muñoz et al., 2011).

Establishment of Guidelines

To obtain optimal language and literacy outcomes for children who are deaf or hard of hearing, the Joint Committee on Infant Hearing (JCIH, 2019) created a series of guidelines for universal newborn hearing screenings and follow-up through Early Hearing Detection and Intervention (EHDI). The guidelines recommend that hearing screenings be completed by one month of age, hearing loss diagnosis obtained by three months of age, and enrollment in early intervention by six months of age. Hearing technology should be introduced within one month of diagnosis for those pursuing spoken language outcomes. New guidelines by the JCIH (2019) suggest moving the timelines of diagnosis and early intervention even earlier—diagnosis by two months of age and enrollment in early intervention by three months of age. Universal hearing screenings before hospital discharge were first recommended by the JCIH in 2000 and programs were subsequently established in all 50 states

(JCIH, 2000). The implementation of newborn hearing screenings has resulted in earlier diagnosis of congenital hearing loss (Gaffney et al., 2010; Muñoz et al., 2013; Yoshinaga-Itano et al., 2017) and proves to be critical for improved language outcomes and vocabulary scores (Grey et al., 2021; Yoshinaga-Itano, 2003; Yoshinaga-Itano et al., 2017).

Barriers to Attainment of Guidelines

Despite these guidelines, many infants who do not pass newborn hearing screenings do not meet this recommended timeline and even become *lost* in the follow-up process. The most recent EHDI data reports that 25.9% of infants in the United States who did not pass a hearing screening are lost to the follow-up process (CDC, 2018). This number varies greatly by state; nearly 90% of the infants who failed a hearing screening in Washington DC were lost to follow-up. In South Carolina, 50% of infants were lost to follow-up. Conversely, Vermont and Wyoming reportedly have no infants who were lost to follow-up post hearing screening. Factors responsible for a delay in obtaining timely services include improper documentation or lack of transportation, funding, or resources (Russ et al., 2010; Sass-Lehrer, 2004). Difficulty obtaining funding and long wait times for appointments were also cited as contributing delays for parents whose children were born as late as 2010 (Muñoz et al., 2013).

It is critical to have an objective measure of the progress toward meeting the 1-3-6 guidelines for all children across the United States. However, these numbers cannot provide a description of the lived experiences of the families who had a confirmed hearing loss or were lost to follow up. Qualitative inquiries into the experiences of parents and families can provide us with information about the diagnostic process that may help us improve outcomes and experiences for future families and children who are deaf or hard of hearing. Additionally, qualitative inquiries may aid in the identification of specific barriers to completing additional audiological assessments and eventually decrease the number of families who do not receive timely and adequate services.

Parent Experiences with Hearing Diagnosis and Follow Up

Previous qualitative investigations have examined parent experiences with pediatric hearing loss identification across Canada, the United Kingdom, Australia, South Africa, and rural areas of the United States (Davids & de Jager, 2018; Elpers et al. 2016; Fitzpatrick et al., 2008; Minchom et al., 2003; Nickbakht et al., 2019; Porter & Edirippulige, 2007). Several studies have closely examined the hearing screening and diagnostic process from the family perspective (DesGeorges et al., 2003; Fitzpatrick et al., 2008; Gilbey, 2010; Scarinci et al., 2018). Among these studies, parents have reported confusion about next steps after a hearing loss diagnosis (Fitzpatrick et al., 2008; Fitzpatrick et al., 2016; Gilbey, 2010). Parents have also wondered about their child's prognosis in developing spoken communication (Fitzpatrick et al., 2008), as well as the impact on academic performance and overall concerns

for their child's development (Fitzpatrick et al., 2016). Regarding hearing healthcare professionals, parents reported that overuse of medical terminology negatively impacted the communication of hearing loss results (Gilbey, 2010) and delayed communication of results to families prolonged the follow-up process (Elpers et al., 2016). Furthermore, professionals may neglect to share follow-up information such as early intervention services or medical referrals (Larsen et al., 2012). Previous qualitative investigations have reported that parents desire connections with other families and peers with hearing loss to develop an understanding of the long-term impact of a hearing loss diagnosis (Nickbakht et al., 2019).

The reported needs and desires of families receiving a hearing loss diagnosis for their child have largely remained unchanged in the past few decades. The two primary needs of parents after a hearing loss identification identified by Luterman and Kurtzer-White (1999) were (a) connections to families and children who are deaf or hard of hearing and (b) unbiased information from professionals regarding communication modality or educational method. For many years, we have known about the needs of parents in the process of a hearing loss diagnosis and have been working to meet the guidelines established by the JCIH. However, some of the same barriers to expedient care present twenty years ago continue to prevent timely acquisition of diagnosis and care today.

Current Study

There has not been a qualitative study exploring parent's experiences with the diagnostic process for children who are deaf or hard of hearing since the implementation of the updated EHDI guidelines in 2019. Furthermore, previous studies that have explored parent's experiences with the early diagnostic and intervention system have been limited primarily to other countries (i.e., Canada, Fitzpatrick et al., 2008; Fitzpatrick et al., 2016, Israel, Gilbey 2010; and the United Kingdom, Minchom et al., 2003) or subsets of the United States (i.e., an Appalachian region of Kentucky, Elpers et al., 2016). The purpose of this study was to obtain information from families of children who are deaf or hard of hearing about their identification, amplification, and therapeutic journey. Parent experiences were also evaluated in relation to child scores on language measures collected as part of a longitudinal study. This study is one essential step to gaining information of the lived experiences of families of children who are deaf or hard of hearing following the implementation of more rigorous newborn hearing screening guidelines. Interviews were conducted with parents of children who are deaf or hard of hearing to answer the following questions:

1. How do parents describe their experience of the journey of hearing loss identification, amplification, and intervention for their child?
2. How do parents of children who are deaf or hard of hearing describe the services they obtained related to their child's hearing loss and communication between the providers of these services?

Method

All study procedures were approved by the University of South Carolina Institutional Review Board. Consent for participation in recorded interviews was obtained prior to each interview.

Participants

Participants included 13 caregivers of children who are deaf or hard of hearing who use amplification and spoken English. The caregivers consisted of nine mothers, three fathers, and one grandmother who participated in 12 interviews. Between the caregivers, there were 14 children who are deaf or hard of hearing who were participating in a longitudinal study. One father and one grandmother each had two children in the study, and one interview involved both the mother and

father of one child with hearing loss. One mother had two children who are deaf or hard of hearing, but only one was a participant in the longitudinal study. The caregivers were recruited from the ongoing longitudinal Early Language and Literacy Acquisition in Children with Hearing Loss Study (ELLA; Werfel, 2017; Werfel et al., 2022; Werfel et al., 2023). The established qualitative inquiry convention of recruiting participants until saturation is reached was used; saturation occurs when no new themes emerge, evidenced by an absence of new codes (Higginbottom, 2004; Saunders et al., 2018). Table 1 presents demographic information of the caregivers and children. Demographic information was gathered from a survey distributed to all guardians of children in the longitudinal study.

Table 1

Caregiver and Child Demographics at Time of Interview

Caregiver	Caregiver Hearing Status	Caregiver Education	Child Age (years; months)	Child Gender	Child Amplification	Language and Communication Mode*	Urban Status
Mother	Hearing	Bachelor's Degree	8;1	Female	CI	100% English	Not Urban
Father	Hearing	Associate's Degree	4;7	Female	HA	75% English; 25% ASL	Urban
Mother	Hearing	Some College	7;4	Male	HA	75% English 25% Spanish	Urban
Mother	Hearing	Graduate/ Professional Degree	7;0	Female	CI	100% English	Not Urban
Mother	Hearing	Bachelor's Degree	7;4	Female	CI	100% English	Urban
Mother	Hearing	Graduate/ Professional Degree	5;2	Female	HA	90% English; 90% Signed Exact English**; 5% Chinese; 5% Spanish	Not Urban
Grandmother	Hearing		8;9	Female	HA	95% English; 5% ASL	Urban Urban
			4;8	Male	HA	95% English; 5% ASL	
Mother	Hearing loss	Bachelor's Degree	7;8	Male	CI	50% ASL; 50% English	Urban
Father	Hearing	Associate's Degree	10;4 6;0	Female Female	CI CI	100% English 100% English	Urban Urban
Mother	Hearing	Bachelor's Degree	4;9	Female	CI	93% English; 7% ASL	Not Urban
Mother	Hearing	Graduate/ Professional Degree	7;6	Female	HA	85% English 15% Russian	Urban
Father	Hearing loss	Bachelor's Degree					
Mother	Hearing	Graduate/ Professional Degree	4;10	Female	HA	99% English; 1% ASL	Urban

Note. ASL = American Sign Language; CI = cochlear implant; HA = hearing aid

*Parent reported language and communication mode use at home

** Signed Exact English used in conjunction with ASL

Procedures

Child Testing

In the national longitudinal ELLA study, from which participants were recruited, children who are deaf or hard of hearing completed a comprehensive battery of early language and literacy measures at set intervals. Study personnel conducted the testing in the child's home or a local library. Study personnel conducted the testing for preschool-aged children at the child's home or local library every six months. School-age children completed a comprehensive battery of language and literacy measures annually after each school year, beginning after first grade at a two-day summer camp.

For the purpose of this study, we used the children's scores on language measures, described below, from the testing session at which their caregiver participated in the semi-structured interview or the following testing session if the interview was via Zoom. Preschool-aged children completed the Peabody Picture Vocabulary Test—Fifth Edition (PPVT; Dunn, 2019), a measure of receptive single-word vocabulary skills. School-aged children completed either the Comprehensive Assessment of Spoken Language—Second Edition (CASL; Carrow-Woolfolk, 2017) or the Clinical Evaluation of Language Fundamentals—Fifth Edition (CELF; Wiig et al., 2013) depending on their age. The CASL and CELF are both omnibus measures of language, assessing language across multiple domains.

Semi-structured Interviews

Caregivers participated in semi-structured interviews in person while their child was participating in the longitudinal study testing or via an online Zoom video call. Twelve interviews in total were completed (one interview involved both the father and the mother so there were 13 participants in total). The majority of interviews took place with caregivers who accompanied their child to the summer camp affiliated with the larger study. Two interviews took place via Zoom, and two took place in person during study visits when study personnel had traveled to the families' location. The interview questions focused on the time around diagnosis of their child's hearing loss, as well as questions they had, support they received, professionals they worked with during this period, what information they were given about childhood hearing loss, subsequent services (e.g., early intervention, speech-language, academic) their child received, and their satisfaction with those services. All interviews were conducted by the first and second author or a trained lab member who had previously observed at least one interview. The interviews lasted an average of 38 minutes.

Coding Process

The interviews were video recorded and transcribed verbatim using NVivo Transcription. The first author reviewed and approved the final transcripts before coding began. Interview data were analyzed using two types of coding: process coding and in vivo coding. We chose

to use these two coding approaches to triangulate the themes identified in the interviews.

Process coding. The procedure for process coding was as follows: first, the second author read a printed copy of each interview and made handwritten notes that consisted of initial ideas. Next, she read each interview again and constructed process codes that emerged from the initial ideas. A data analytic memo for each interview and a codebook consisting of all codes used across interviews were created during this phase. The second author then created a process model that was reviewed by and discussed with the research team. Finally, she made a final pass through each interview that involved streamlining the codes within the codebook and incorporating feedback from the research team, resulting in a final codebook and process model. The entire research team reviewed and agreed upon these final products from the process coding analysis.

In vivo coding. The procedure for in vivo coding was as follows: first, the first author read a copy of each interview. Next, she read each interview again and coded the text following in vivo coding methods, in which the codes use the exact wording of the participants. The first author wrote analytic memos to document her thought process during the coding process. After a first pass was completed for each interview, the first author read through the interviews, codes, and analytic memos. She then completed a second pass of in vivo coding focusing on codes that emerged across interviews. Finally, she compiled all of the codes in a codebook that contained the codes, the corresponding quote from the interview, the participant code, and additional notes for context as needed. The entire research team reviewed and agreed on the final codebook from the in vivo codebook analysis.

Results

In this investigation, we were interested in parents' experiences from the time of suspicion that their child might not be hearing as expected to attainment of a diagnosis and enrollment in intervention services. Two different experience paths emerged based on the reported information from the parents. Children were classified on paths according to parent report. Qualitative coding revealed themes across and between paths.

Path Classification

First, we identified the beginning of the process of interest as either a newborn hearing screening that was not passed or the time at which a parent described first becoming concerned about their child's hearing. For most families, this time was at the point of the newborn hearing screening; however, some families reported passing or not receiving a newborn hearing screening and later becoming concerned about their child's hearing. We then considered the time at which the parents received a definitive diagnosis of hearing loss. Children whose parents felt the diagnosis went quickly were classified in the Fast to Diagnosis group and had a time until diagnosis of 4 months or less ($n = 9$ children). Children whose parents felt the diagnosis did not go smoothly were classified in the Slow to Diagnosis group and had an average time

to diagnosis of 5 months or more ($n = 5$ children). Of note, those whose time was four months or less felt like this process went smoothly even though it is one month beyond the recommended guidelines from EHDI. We used parent experience, and not time to diagnosis, to determine this grouping. Importantly, because this metric is based on the time from suspicion of a hearing problem, it does not necessarily correspond to chronological age. Average age at identification was 5.39 months for the Fast to Diagnosis group ($SD = 11.54$) and 13.25 months ($SD = 13.33$) for the Slow to Diagnosis group.

In addition to this classification, we also considered each child’s language skills. All children in the Fast to Diagnosis group had spoken language skills within the average or above average range. For children who were in the Slow to Diagnosis group, there was a mix of performance, such that one child had average to above average language scores, one child had low average scores, one had a mix of average and below average scores, and two children had below average language scores. Figure 1 shows the boxplots of language scores for children in the two groups. The groups were corroborated via mixed methods of parent-reported time between suspecting hearing loss and receiving a diagnosis (as detailed above), process coding, and current language scores. We refer to the Fast to

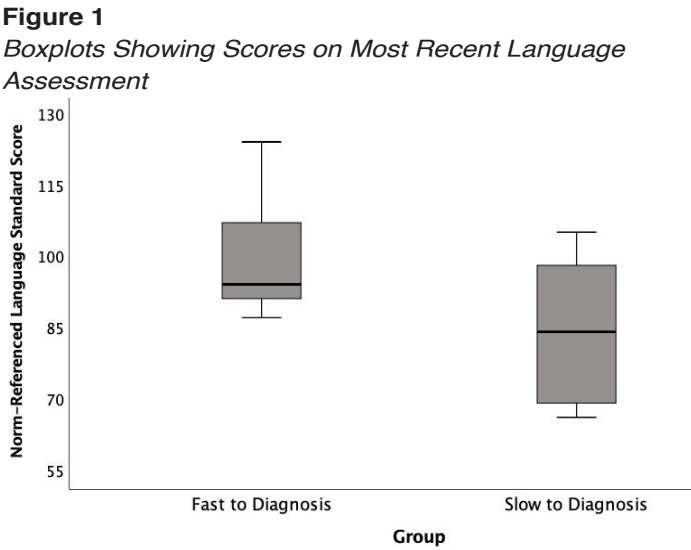
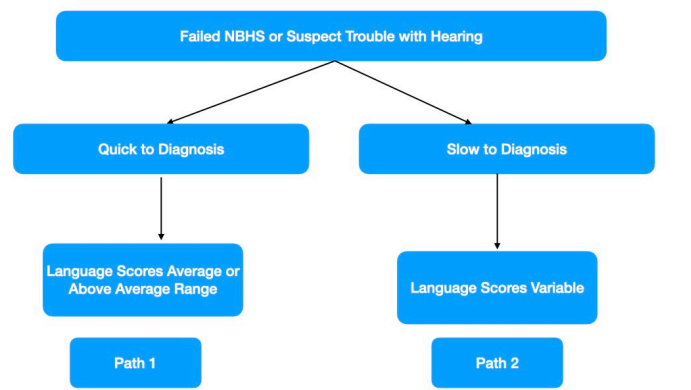


Figure 2
Classification of Families Depending on Perceived Time to Diagnosis and Language Scores



Diagnosis group as Path 1 and the Slow to Diagnosis group as Path 2. See Figure 2 for path classification details.

Process Coding

Path 1

Using process coding, we identified characteristics and experiences of parents on two paths. For families on Path 1, five prominent themes repeatedly were observed in the interviews. First, these parents were connected with hearing healthcare professionals who had specific knowledge of pediatric hearing loss and who the parents trusted. The combination of knowledge and trust was an important defining characteristic for Path 1. On Path 2, parents often reported *either* trusting their professionals or that they were knowledgeable, but no parents on Path 2 reported both. Second, parents on Path 1 often conveyed a *just do it* attitude. These parents expressed that they listened to their providers and did what they were told. This attitude also reflects the importance of having trusted and knowledgeable professionals; some parents on Path 2 also reported following the providers’ recommendations, but those recommendations did not follow best practice for pediatric hearing loss. Some examples of this include: not following up on a newborn hearing screening that was not passed on the recommendation of the child’s pediatrician; returning to the audiologist every six months for three years to try to obtain a valid conditioned play audiometry result; selecting a communication approach recommended for a child by their speech-language pathologist without fully considering the implications for their child and family. Third, parents on Path 1 often had previous experience navigating the hearing healthcare system, either as a result of having hearing loss themselves or having an older child with hearing loss. Those with prior experience reported encountering fewer obstacles, even in cases where the families were navigating different healthcare systems (i.e., they had moved to a new city). Fourth, families on Path 1 often reported that all of their child’s services, including ENT, audiology, speech-language pathology, and in some cases, preschool, were located in the same building. Parents reported that this was beneficial not only for convenience of appointment scheduling but also because of the high occurrence of intercommunication among the members of their child’s hearing healthcare team. Finally, parents on Path 1 reported that when their child reached elementary school, the schools were either willing to provide the services their child needed *or* the parents were well-prepared to advocate for them.

Path 2

The shared experiences of parents on Path 2 included four primary themes. First, parents reported that someone downplayed the need for follow-up; this downplaying occurred for some families after their child did not pass a newborn hearing screening and for others after they reported to their pediatrician a concern about their child’s hearing. Many families reported that after their child did not pass their newborn hearing screening, they were told to not worry. Often the families heard things like, “Failing

the screening doesn't mean your child has hearing loss," or "It's probably just fluid." Parents reported pediatricians often downplayed the need for follow-up to a hearing screening when a child, for example, flinched in response to a loud sound while in their office for a visit. Newborn hearing screeners' counseling was often interpreted by families to mean that the child's hearing was okay, even in the presence of a refer test result. Second, multiple parents on Path 2 reported being told that their child passed their newborn hearing screening. These children often received multiple screenings that they did not pass before the one they passed, even in the case of a family history of pediatric hearing loss. Third, parents on Path 2 often reported getting the run-around from professionals prior to their child's diagnosis. Importantly, this occurred across all types of professionals that a child with hearing loss might encounter before their diagnosis, including pediatricians, audiologists, speech-language pathologists, and early interventionists. Additionally, some parents reported that community audiologists had them come in for many testing visits before they received a diagnosis; in one case, these visits spanned up to three years. Other parents reported that early interventionists and early intervention speech-language pathologists were slow to refer the child for a hearing screening, even in the presence of a family history of hearing loss and speech sound production errors characteristic of children who are deaf or hard of hearing.

Most parents on Path 2 reported that sometime after their child's hearing loss diagnosis, they connected with hearing healthcare professionals who were knowledgeable about pediatric hearing loss and who the parents trusted. Importantly, families generally were not connected to these professionals via the healthcare system. Instead, an intervening figure, such as a family member or a family friend, connected families with these professionals. In fewer, but multiple, cases, the intervening figure came in the form of other families of children who are deaf or hard of hearing that the parent met through a local or online support group. One parent on Path 2, however, reported a continued lack of access to professionals with pediatric hearing loss knowledge as a result of their geographic location, a lack of available services in their area, and a lack of resources to seek services elsewhere.

In Vivo Coding

Triangulating with the results of the process coding analysis, four themes, seven subthemes, and four categories emerged from the in vivo coding of the transcribed interviews (see Table 2 for an overview). First, themes were identified. Within those themes, subthemes were identified. Finally, categories within the subthemes were identified when applicable. Some themes were reported by parents on both paths and some were only experienced by parents on one path.

Table 2
Themes, Subthemes and Categories from In Vivo Coding

Theme	Subtheme	Category
Limited access to hearing loss services	Parents reported issues with professionals during hearing loss diagnosis	Parents reported pediatricians lacked hearing loss knowledge
		Parents reported receiving insufficient information from professionals regarding next steps in process
	Parents reported lack of local hearing loss services	
Parents received insufficient information from hearing loss professionals	Parents reported concerns over communication modality	
	Parents desired connections	Parents desired a connection with a family who had a child diagnosed with hearing loss
		Parents desired a connection with a member of the Deaf community
Parents reported concerns about their child's development	Parents felt something was not right with their child's hearing	
	Parents reported guilt about not obtaining diagnosis sooner	
Parents reported good experiences	Parents reported ease of having multiple services in one location	

Theme 1: Limited Access to Hearing Healthcare Services

One of the overarching themes identified that was present across parents on both paths was *limited access to hearing healthcare services*. This limited access was due to struggles in their interactions with professionals in the process of obtaining a hearing loss diagnosis for their child, represented in the subtheme *parents reported issues with professionals during hearing loss diagnosis*. One category that emerged from this subtheme was *parents reported pediatricians lacked hearing loss knowledge*. Several parents brought their concerns to their pediatrician but were assured it was likely “just fluid” in their child’s ear canal. Based on this feedback from their healthcare provider, many families in our study on Path 2 were satisfied in the short term which resulted in a delayed diagnosis of their child’s hearing loss. One father said of his experience seeking a diagnosis of his daughter’s hearing loss, “Every time we contacted a pediatrician’s office, they all kind of had the same one liner, ‘Oh it’s fine. Don’t worry about it. It’s just fluid.’” Notably, parents on Path 1 reported hearing this information from a healthcare provider but were not satisfied and sought second opinions or continued to request an audiological evaluation. Another category we identified from this subtheme was *parents reported receiving insufficient information from professionals regarding next steps in process*. Several parents also reported that at the time of the hearing loss diagnosis, they felt they received insufficient information from the audiologist. One mother recounted from her daughter’s initial diagnosis:

A very young audiologist that did the test and she came into the recovery room and said “She’s deaf but it’s more than that. She actually has something called auditory neuropathy and we don’t treat that here so you’ll have to go somewhere else” and she walked out and left us with nothing.

Another mother said of the audiologist who made her son’s initial diagnosis,

When he has hearing loss and everything was...we were crying like [shrugs]... she barely had time to talk to us because she was getting ready to leave for Thanksgiving break.

Parents reported feeling overwhelmed emotionally with the information of the diagnosis. Parents also reported feeling unsupported by not being given information on what the next step was for their child. One mom said, “We should have had just more information up front. Someone to give us a clear path, or a clearer path of at least what our next step was.” Another father shared this about his experience, “Nobody ever gave me that big arc like this is how it works.”

In some cases, the limited access to hearing healthcare services was caused by a physical restriction, such as lack of local hearing healthcare services or a limited number of hearing loss professionals sufficiently familiar with pediatric

hearing loss. These physical limitations are reflected in the subtheme *parents reported lack of local hearing healthcare services*. Parents, especially those in rural or less populous areas, reported traveling far distances to find professionals who were sufficiently familiar with pediatric hearing loss. For many of these parents, the professionals in the towns in which they lived were not sufficiently familiar with pediatric hearing loss. One mother said, “I really wish we were in an area where there was more than one pediatric audiologist so I could have gotten a second opinion instead of going to [nearest large city].” Another mother responded, in answer to a question about what she disliked about the services she received, “The fact that we had to go so far to get quality access, access to quality professionals.”

Even for parents on Path 1 who had overall positive experiences, the distance they traveled for their child’s audiology appointments and therapy sessions was notable. One mother said, “It’s about 45 minutes...you know there’s nothing really offered in our town.” Another mother recounted how she and her daughter did not move with her husband when he was stationed across the country in order to remain close to her daughter’s preschool for children who are deaf or hard of hearing. She said, “We were like, we’re not moving [child’s name] from her services at [hearing loss preschool]... It was definitely a deciding factor as to where our family would stay.” The distance families traveled for their children’s services was mentioned in eight of the 12 interviews.

Theme 2: Parents Received Insufficient Information from Hearing Loss Professionals

Several parents on both paths reported limited information received from hearing healthcare professionals and desiring additional information from other routes. This was supported by the subtheme *parents reported concerns about their child’s communication modality*. Specifically, these concerns were often related to the process of selecting a communication modality for their child, spoken English or American Sign Language (ASL). Only one mother was a fluent native signer of ASL. For several parents, they identified this process as a negative experience even if the chosen communication modality has been working for their family. Many parents reported that the decision to use spoken language or ASL was theirs to make; however, they felt ill-equipped to make this decision and wish they were given more information about either ASL or long-term spoken language outcomes. One mother said of her speech-language pathologists’ recommendation to use ASL instead of spoken English, “We would have liked to understand the rationale for the things he was recommending.” Another mother whose daughter attends a school for the Deaf said, “It’s just kind of like here’s your choice, like well how do I choose if I don’t have some additional information, you know.” A mother whose daughter uses spoken English told us of the recommendation to use only spoken language, “I would have liked to have gotten a more balanced opinion and seen quality research on both sides of that opinion.” This mother recounted an instance when she regretted

heeding the advice of professionals who encouraged her to exclusively use spoken language with her daughter:

[Child's] processor failed and she was without hearing for three weeks and we don't know enough sign language to carry on quality conversations. So, it would have been nice to have had... access to more quality balanced research to know where to go.

Parents also reported that at the time of their child's diagnosis they would have benefitted from being connected with an individual with hearing loss, identified in the subtheme *parents desired connections*. Two categories emerged; the first being *parents desired a connection with a family who had a child diagnosed with hearing loss*. Parents reported wanting to know what the future looked like for their child. Several of them either had no or limited familiarity with individuals with hearing loss. During the interview, one dad recounted this question he had at the time of his daughter's diagnosis, "I didn't know if that meant you're going to speak like me, you're going to speak like that kid I remember from school." Parents reported that being put in contact with a family with a child with hearing loss who was a few years older than their child would have helped them see what their choices would look like in the future. One mother told us:

One of the other things I wish had been done was I wish I had been given the opportunity to meet families who are a few years down the path from me because that was really like I remember the first time I met a high schooler who was implanted and was like he can talk. He has been through you know a normal public-school education. He plays football. He kind of seems like a normal kid and that he really was like for the first time for me like a sigh of relief like OK it's going to be fine. [child's name] is going to have a normal life.

Another category that emerged is *parents desired a connection with a member of the Deaf community*. Some parents reported that they desired the opportunity to learn sign language or Deaf culture from a member of the Deaf community. Two parents that we interviewed sought these services to learn sign language on their own as well as to better communicate with their children. One mother desired that opportunity, but did not know how to make that initial connection with a member of the Deaf community. She reported

Anybody that's taught sign language to us is a hearing person....That's been difficult. We've got videos and stuff like that. But there's still that incidental stuff in sign culture that we don't get. There's good access to the base language but conversational language and colloquial stuff, I don't know where it is.

Theme 3: Parents Reported Concerns about Their Child's Development

A subset of parents across both paths reported multiple instances throughout the diagnosis process where they were concerned about their child's development. One subtheme that emerged was *parents felt something was not right with their child's hearing*. Many parents described knowing their child's development was not proceeding normally even before they knew their child's diagnosis. One mother said, "I had this mama gut feeling that there's probably something more going on." Another mother said of their experience, "We saw something like seven audiologists and five ENTs between when she was born and when she was 13 months old because they kept saying, 'she's fine, she can hear.' But we knew that she couldn't."

Another subtheme we identified for parents on Path 2 was *parents reported feeling guilt about not obtaining diagnosis sooner*. One father said "I blew it. I was an idiot," in regard to not seeking hearing aids for his child earlier. One mother said, "I didn't push as hard as I could have because I was upset I was even having to do this" and "I felt like I didn't push hard enough." This mother repeatedly mentioned her suspicions of her daughter's hearing loss to her pediatrician and attempted to have her tested by a friend who was an audiologist before obtaining an appointment for a sedated ABR that would provide the diagnosis. Another mother said, "I wish I had done something sooner" and "I felt cheated that we had not had this diagnosis earlier." This mother's child did not pass the newborn hearing screening and was seen by three different audiologists before being diagnosed at age three.

Theme 4: Parents Reported Good Experiences

Although most parents reported difficulties or frustrations with aspects of the diagnosis and amplification process, parents on Path 1 and some parents from Path 2 also reported positive experiences. One mother shared,

They immediately fitted her with hearing aids and put hearing aids on her even though they didn't work really. And they immediately started, I think by the time she was two months old, they were doing a speech service.

When asked if she had questions at the time of her daughter's initial diagnosis, another mother said, "We didn't have to ask a lot of questions because our audiologist was awesome. Just love her to death. She's just the best."

By seeking out a medical center in a major city, some families were able to get many of the necessary services, such as audiology appointments, speech-language therapy, and parent education, under one roof, reflected in the subtheme *parents reported ease of having multiple services in one location*. One father said, "I liked having all the services in one place...For us living so far away, it was great to have everything under one roof, even the audiologist, because we already spent so much time in the car."

One father said of the preschool his daughter attended for children who were deaf or hard of hearing, “I felt like this was the place to be.” Another mom describes the close relationship she and her daughter have with her daughter’s first teacher of the deaf; she describes texting her after meeting with new professionals and that she continues to check in about her child even though she is no longer in her class.

Discussion

The purpose of this study was to obtain information from families of children who are deaf or hard of hearing about their identification, amplification, and therapeutic journey. The children of the participants in this study were born after the implementation of the EHDI 1-3-6 standards. However, for many families in this study, those guidelines were not met. The results of the coding and analysis revealed common elements that contributed to overall positive or overall negative outcomes for the families in our study.

Two paths emerged for families on their child’s hearing journey in this study. On Path 1, parents felt the period of time between suspected hearing loss and diagnosis was short and that they were quickly connected with knowledgeable and trusted professionals. On Path 2, parents felt there was a long period of time between suspected hearing loss and diagnosis, but upon diagnosis families were *typically* connected with knowledgeable and trusted professionals.

Connections of Themes to Previous Work

An overarching theme that emerged was limited access to hearing healthcare. Some families were not able to access services in a timely manner, preventing them from meeting recommended EHDI guidelines, whereas other families were inconvenienced by the difficulty in obtaining these services but were able to access them. This difficulty in limited access to hearing healthcare services was also reported in investigations exploring barriers to timely healthcare for families in rural America (Elpers et al., 2016) and Canada (Fitzpatrick et al., 2008).

The importance of frequenting hearing healthcare providers knowledgeable about pediatric hearing loss was evident from our participants. Many parents in our study related their experiences with pediatricians lacking hearing loss knowledge who played primary roles in preventing them from obtaining a timely diagnosis. Similar difficulties were encountered by some parents who were seeing audiologists not familiar with *pediatric* hearing loss. These professionals prevented the parents from obtaining a confirmation of the diagnosis of hearing loss. In a survey study in the United States, Larsen et al. (2012) found that more than 50% of participating parents were lacking information regarding medical referrals. Parents in previous studies also reported professionals minimizing hearing loss, resulting in delay of diagnosis and even speech and language delays (Elpers et al., 2016; Fitzpatrick et al., 2016).

Several parents reported some misgivings regarding the process of selection of communication modality, either because they themselves desired more information or they did not feel they were working with a professional knowledgeable about hearing loss. In fact, unbiased information from professionals surrounding communication modality has been identified as one of two primary needs of parents surrounding a hearing loss diagnosis (Luterman & Kurtzer-White, 1999). A nationwide study by the National Center for Hearing Assessment and Management (NCHAM; 2021) reported some families’ frustration regarding locating professionals who used their desired communication modality.

Parents expressed a desire to connect with others throughout their hearing loss journey, in particular families with other children who are deaf or hard of hearing who were a few years older than their children to see what their future might look like. This theme supports previous findings. Following a hearing loss diagnosis, parents desire connections with other members of the hearing loss community, particularly children (Luterman & Kurtzer-White, 1999; Nickbakht et al., 2019). Nickbakht and colleagues (2019) reported that parents of children who are deaf or hard of hearing in Australia wanted to connect with other families “in the same boat” (p. 677) and desired professionals to “walk the journey” (p. 677) with them. The difficulty families encounter connecting with family-to-family support systems has been identified as a major hurdle (NCHAM; 2021); NCHAM suggests these organizations formalize partnerships with state-level EHDI programs.

Future Directions and Limitations

Many parents, in our study and in the existing literature, refer to the “steps in the process” of navigating a hearing loss diagnosis. Some of our parents desired a “blueprint” or a “roadmap.” In many ways, EHDI guidelines provide a blueprint for hearing loss providers. Hearing healthcare professionals should inform families of next and future steps to decrease families’ feelings of frustration and increase adherence to recommended guidelines. Future work should investigate the impact of increased parent education and support on adherence to recommended EHDI guidelines.

It should be noted, all participants in this study had a child who was enrolled in a longitudinal study for children who are deaf or hard of hearing, many of whom found this study through connections with professionals well-versed in hearing loss or through preschools and centers for children with hearing loss. Every child in this study eventually obtained a diagnosis of hearing loss. Even so, one parent on Path 2 never was connected with a knowledgeable professional. This suggests that there may be additional paths or subsets of paths for families who have struggled or are still struggling to obtain a diagnosis and connect with hearing healthcare professionals that were underrepresented in these interviews. These families may have experiences that differ from the caregivers in our study. Additionally, parents in this study were generally

highly educated, their experiences may not be reflective of parents from all socioeconomic and educational backgrounds.

Conclusion

This study investigated the experiences of parents whose child had been diagnosed with hearing loss. Important themes that emerged highlight areas of hearing loss service delivery that can be improved, including limited access to hearing healthcare services and receiving insufficient information from professionals. The results from this study shed light on the navigation of hearing healthcare services from the parents' point of view and may aid in the identification of barriers to timely diagnosis and intervention and children lost to follow-up. Furthermore, collaboration between professionals and a trusted network of referrals may significantly decrease family frustration and increase adherence to EHDI 1-3-6 guidelines.

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2023



NENI DIRECTORY

Håfa Adai & Welcome!

Newborn hearing screening is extremely important as it helps to determine whether infants have hearing loss. Guam EHDl works to assure that all babies born on Guam get screened for hearing loss by 1 month. If a baby does not pass their hearing screen, a Diagnostic Audiological Evaluation (DAE) will be conducted by 3 months of age. A baby with hearing loss will be enrolled in early intervention services by 6 months of age.

Universal newborn hearing screening is mandated by [Guam Public Law 27-150](#).



Early identification of hearing loss is key to providing appropriate intervention services to promote language development.

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Early Intervention