Barriers to and Facilitators of Early Hearing Detection and Intervention in the United States: A Systematic Review

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Objectives: Early hearing detection and intervention (EHDI) is guided by the 1-3-6 approach: screening by one month, diagnosis by 3 mo, and early intervention (EI) enrollment by 6 mo. Although screening rates remain high, successful diagnosis and EI–enrollment lag in comparison. The aim of this systematic review is to critically examine and synthesize the barriers to and facilitators of EHDI that exist for families, as they navigate the journey of congenital hearing loss diagnosis and management in the United States. Understanding barriers across each and all stages is necessary for EHDI stakeholders to develop and test novel approaches which will effectively reduce barriers to early hearing healthcare.

Design: A systematic literature search was completed in May and August 2021 for empirical articles focusing on screening, diagnosis, and EI of children with hearing loss. Two independent reviewers completed title and abstract screening, full-text review, data extraction, and quality assessments with a third independent reviewer establishing consensus at each stage. Data synthesis was completed using the Framework Analysis approach to categorize articles into EHDI journey timepoints and individual/family-level factors versus system-level factors.

Results: Sixty-two studies were included in the narrative synthesis. Results revealed that both individual/family-level (e.g., economic stability, medical status of the infant including middle ear involvement) and system-level barriers (e.g., system-service capacity, provider knowledge, and program quality) hinder timely diagnosis and EI for congenital hearing loss. Specific social determinants of health were noted as barriers to effective EHDI; however, system-level facilitators such as care coordination, colocation of services, and family support programs have been shown to mitigate the negative impact of those sociodemographic factors.

Conclusions: Many barriers exist for families to obtain appropriate and timely EHDI for their children, but system-level changes could facilitate the process and contribute to long-term outcomes improvement. Limitations of this study include limited generalizability due to the heterogeneity of EHDI programs and an inability to ascertain factor interactions.

Key words: Early intervention, EHDI, Social determinants of health.

Abbreviations: CASP= Critical Appraisal Skills Programme; D/HH = deaf or hard-of-hearing; EHDI = early hearing detection and intervention; EI = early intervention; LTD = loss/lost to documentation; LTFU = loss/lost to follow-up; NICU = Neonatal Intensive Care Unit; SDOH = social determinants of health; WIC = Special Supplemental Nutrition Program for Women, Infant, and Children.

IDEA

Inclusion, Diversity, Equity, Accessibility Article.

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INTRODUCTION

The incidence of permanent, congenital hearing loss is estimated to be 1–3 per 1000 infants (Williams et al. 2015). Undiagnosed and late-diagnosed congenital hearing loss have significant developmental implications related to speech/language, cognition, academic achievement, and quality of life (Vohr et al. 2008; Pimperton & Kennedy 2012; Pimperton et al. 2016; Yoshinaga-Itano et al. 2017, 2018). National standards for early hearing detection and intervention (EHDI) have been provided and updated by the Joint Committee on Infant Hearing since 1971. Current national benchmarks, which are supported by the World Health Organization (2021) for implementation world-wide, include screening for hearing loss in all infants before 1 mo of age, diagnosis by 3 months, and enrollment in early intervention (EI) services by 6 months (JCIH 2019). Completion rates for infant newborn hearing screening in the United States have rapidly increased since the inception of state-specific universal newborn hearing screening programs (CDC 2022). Unfortunately, rates for diagnostic follow-up and EI–enrollment lag behind screening rates despite nearly 20 years of EHDI program evolution for most states (Subbiah et al. 2018).

Over the past 30 years, an extensive corpus of research has been produced examining the various factors that contribute to, or hinder, successful EHDI. Many early studies focused on effective screening techniques to inform programs in evidence-based practice to facilitate early identification of risk for hearing loss (Jacobson & Morehouse 1984; Hosford-Dunn et al. 1987; Norton et al. 2000). As programs evolved, so did research into areas spanning diagnosis, management, EI, family experience, and programmatic aspects of EHDI. Most recently, equity in EHDI was addressed by a systematic review by Kingsbury et al. (2022), which called on professionals within the EHDI system to provide culturally responsive care and advocate for policy changes that can effectively address disparities in outcomes. Understanding the complex factors identified through previous research regarding EHDI and formulating approaches that effectively address these factors will be critical in the continued evolution of EHDI programs to help facilitate equitable EI for children who are born deaf/hard-of-hearing (D/HH).

The purpose of this systematic review was to critically examine the barriers and facilitators that exist for families as they navigate the journey of congenital hearing loss diagnosis and management in the United States. Understanding these combined factors across all stages is necessary for EHDI stakeholders to develop and test novel approaches that will effectively reduce barriers to early hearing healthcare.

MATERIALS AND METHODS

This systematic review was registered with Prospero (CRD42021257187). The review was completed according to

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the Preferred Reporting Items for Systematic Review and Meta-analyses (PRISMA) guidance.

Search Strategy

A systematic search of PubMed, EMBASE, CINAHL, APA PsychINFO, and Google Scholar was completed on May 9, 2021, and August 27, 2021, for articles containing medical subject heading terms of hearing loss/diagnosis, early medical intervention, loss-to-follow-up, time-to-treatment, or early hearing detection and intervention. The search strategy is detailed in Appendix A in Supplemental Digital Content 1, http://links.lww.com/EANDH/B77.

Study Selection Criteria and Data Extraction

Covidence systematic review software (www.covidence.org) was used to manage three stages of study selection and evaluation: (1) screening, (2) full-text review, and (3) data extraction. Inclusion and exclusion criteria were established before the title and abstract screening and were agreed upon by all authors. Inclusion criteria were as follows: empirical, peer-reviewed articles in English completed in the United States with a population focus on infants birth-to-three years of age, and published between 1990 and 2021. Articles that did not specifically address barriers or facilitators to EHDI were excluded. For this review, articles specific to the United States were targeted for the purpose of concentrating on one country with a national guideline for EHDI (i.e., 1-3-6) but lacking a standard for program execution across states. In addition, the United States is without universal health insurance coverage, which adds a dimension that is different from other developed countries. Studies from other countries engaging in EHDI may be very helpful in providing insight for potential successful facilitators; however, the United States presents its own unique challenges and to define those challenges, studies within the United States were the only ones included in this review.

During the screening phase, titles and abstracts were assessed by two independent reviewers of a four-person review team (UF, CD, DH, LH) for potential relevance. The managing reviewer (UF) resolved any conflict (e.g., one reviewer selected "yes" or "maybe" and the other selected "no") and involved a third reviewer if required to resolve discrepancies. For the full-text review stage, two independent reviewers screened all potentially relevant full-text articles (i.e., all articles tagged as "yes" or "maybe" during the screening stage). Disagreements were resolved by consensus or a third member of the review team.

For the data extraction stage, five reviewers (UF, CD, JC, DH, and LH) extracted predetermined data from each study, including: study aim(s), study design, population characteristics, inclusion/exclusion criteria, intervention details, outcome measures, details of control or comparison groups (if applicable), overall results/conclusions, barriers identified, and facilitators identified. Each reviewer independently extracted data and rated quality using the Critical Appraisal Skills Programme (CASP 2019) cohort study template modified to fit the review aims and specific study characteristics. The CASP checklists help reviewers evaluate the validity, results, and clinical relevance of each article. In addition, the study's level of evidence was identified according to the Oxford categorization scheme (Oxford Centre for Evidence-Based Medicine 2009). A third reviewer evaluated the data and completed consensus when data were found to conflict between the first two reviews.

Data Analysis

Quality ratings, including level of evidence, were summarized and reviewed for key areas of quality from the modified CASP, including risk for bias in subject recruitment or data analysis, applicability of results, and consistency with other evidence. Authors then ranked the importance of each of the 12 quality indicators from the modified CASP to select key quality ratings used to identify the highest-quality studies. Rankings resulted in the following factors judged to be of greatest importance: focused aim, acceptable recruitment, outcomes measured without bias, identified confounds, and confounds accounted for in the design. Articles with reviewer ratings that met all five of these factors were assigned an overall CASP rating of "1" (primary articles with higher level of evidence) while those with one or more of these factors found to be lacking by reviewers were assigned an overall CASP rating of "2" (secondary articles with relatively lower level of evidence). Data from articles were analyzed using the Framework Analysis approach (Ritchie & Spencer 1994; Goldsmith 2021) to categorize studies into four different time periods along the EHDI timeline (specifically, screening, diagnosis, management, EI), as well as the entire journey. The approach also differentiated between studies that addressed individual/family factors vs. system-level factors that served as either barriers or facilitators. For the purposes of this review, "management" refers to provision of hearing technology (e.g., hearing aid or cochlear implant). Enrollment in EI and provision of speech/language therapy was classified in the EI category. Individual/family factors refer to demographic characteristics (e.g., race/ethnicity, insurance status, medical status of the infant, and distance to major healthcare centers), whereas system-level factors refer to functionality and logistical characteristics of the EHDI system itself. Study conclusions were compared for agreement or disagreement on barriers and facilitators for each step of the EHDI timeline. Some factors applied to more than one step along the EHDI timeline (e.g., access to healthcare facilities or family knowledge of next steps for completion of timepoints), whereas others applied to a specific step (e.g., infant prematurity delaying screening completion or financial barriers impacting management via devices).

RESULTS

The initial literature search yielded 672 studies and after 108 duplicates were removed, 564 studies remained. An additional 395 articles were not relevant to the primary aim after title/abstract screening and the remaining 169 articles underwent a full-text review. During full-text review, 107 were excluded for various reasons (62 did not address barriers related to EHDI, 33 were not empirical, 7 were not completed in the United States, 2 were abstracts only, 2 had incorrect patient populations, and 1 was a wrong setting). Ultimately, 62 articles met criteria for extraction of data and quality/bias reviews (Fig. 1). A master table of extracted articles including level of evidence, overall CASP rating, study design, population, number of participants, outcome measure(s), comparison(s), intervention(s), EHDI time point(s), factor level, barriers, and facilitators identified can be found in Appendix B in Supplemental Digital Content 1, http://links.lww.com/EANDH/B77.

Levels of evidence and quality ratings were analyzed to identify articles with higher quality to include in further analysis. Of the 62 articles, only one was a level 1b randomized control trial by Bush et al. (2017) while most other articles (n = 27, 43.5%) were level 2b cohort studies or level 3b cohort studies (n = 10, 16.1%). Finally, there were eight level 4 studies and 16 level 5 studies that generally represented survey-based methods of expert opinion. Based on the five modified CASP factors with a level of evidence rating of 3b or higher, 20 (32.3%) articles were identified as having higher levels of quality (indicated in Appendix B by + in Supplemental Digital Content 1, http://links.lww.com/EANDH/B77). Analysis of study results yielded themes across the entire EHDI journey as well as themes specific to individual EHDI timepoints. Due to the heterogeneity of study designs and factors, a meta-analysis of results was not possible and therefore an evidence-based narrative synthesis was completed. What follows is a summary of thematic findings for barriers to, and facilitators of, successful completion at each step of the EHDI process. Barriers and facilitators specific only to screening, diagnosis, management, or EI enrollment are separately discussed. A visual summary of the findings can be found in Figure 2 while a table of barriers and facilitators identified by each article can be found in Appendix B in Supplemental Digital Content 1, http://links.lww.com/EANDH/B77.

Barriers to Effective EHDI

Entire EHDI Journey

Individual/Family Factors

Overall, there is an overarching cascading effect for each step in the EHDI process in which late screening leads to late diagnosis, and in turn, late management and EI enrollment (Spivak et al. 2009; Chapman et al. 2011; Holte et al. 2012; Krishnan & Van Hyfte 2014; Tran et al. 2016a; Shanker et al. 2019). Due to this cascading effect, Holte et al. (2012) reported that only 32% of babies who referred on the newborn hearing screening actually meet all three 1-3-6 benchmarks. The most prominent theme for individual/family factors impacting the entire EHDI journey was social determinants of health (SDOH). SDOH include conditions in the environment that can have a significant impact on health, functioning, and quality of life (Healthy People 2030). Domains within SDOH include economic stability, education access and quality, health care access and quality, neighborhood and built environment, and social and community context. Within each of these domains, conditions such as low income, limited education, limited access to healthcare due to distance or lack of transportation, etc. presented as barriers to meeting EHDI milestones in 16 of the 20 (80%) higher level of evidence studies.

Lack of *economic stability* places infants at higher risk for either not completing the EHDI process or having delays in

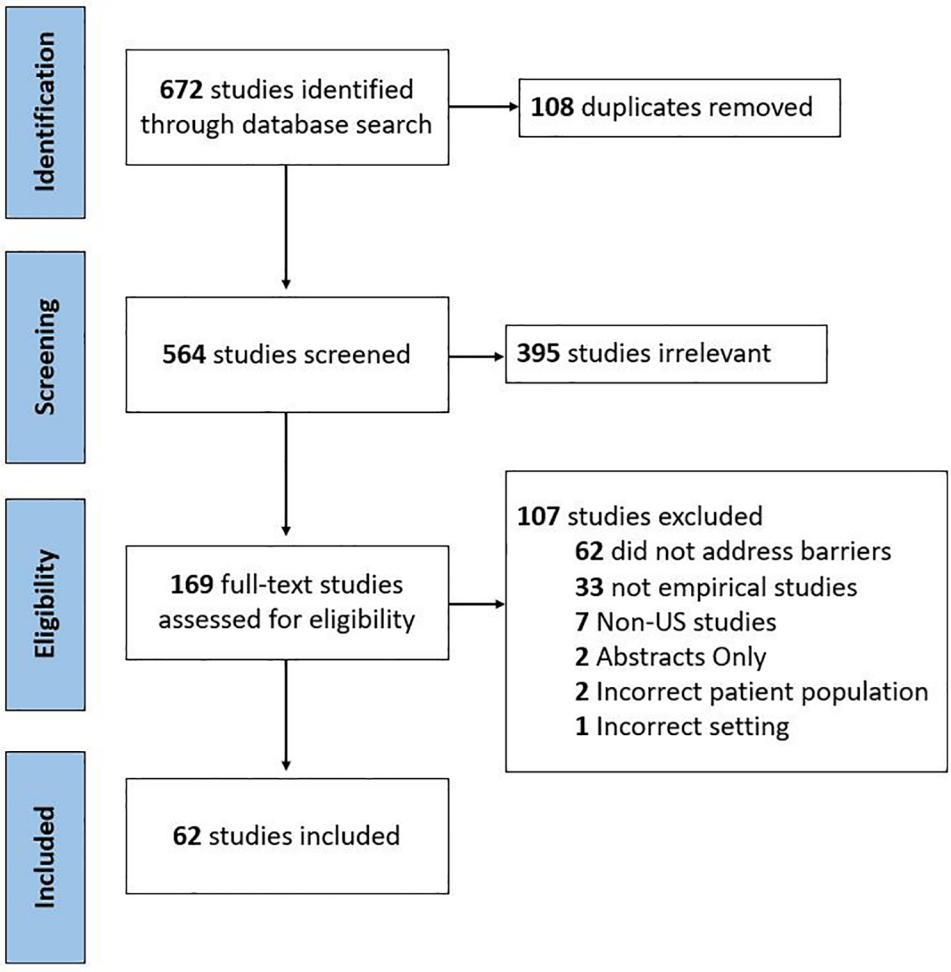

Fig. 1. PRISMA Flowchart of the systematic review.

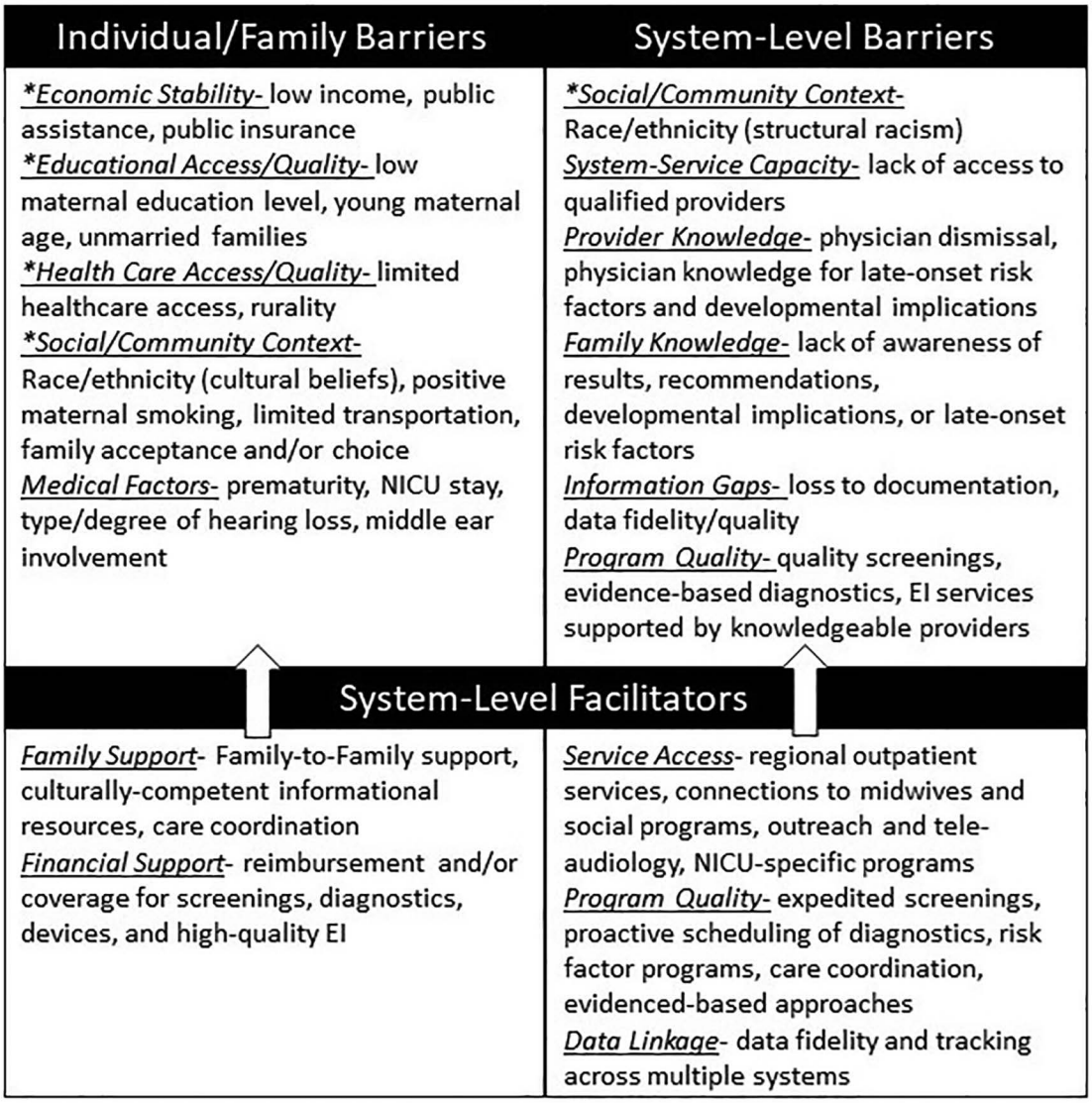

Fig. 2. Visual summary of barriers and facilitators spanning individual/family factors and system-level factors. *Denotes those factors falling under the SDOH categorization. SDOH, social determinants of health.

their completion across all steps in the journey, for example, if they came from families with low income (Hoffman et al. 2011; Bush et al. 2015b; Elpers et al. 2016; Cunningham et al. 2018; Roman et al. 2021) or who used public assistance programs or public insurance (Auerbach et al. 2013; Cunningham et al. 2018; Zeitlin et al. 2021). Public insurance was a significant factor specifically at the diagnostic step of the EHDI journey (Liu et al. 2008; Deem et al. 2012; Auerbach et al. 2013; Crouch et al. 2017; Meyer et al. 2020; Sapp et al. 2021) while financial concerns arose for funding hearing technology during the management step (Holte et al. 2012; Muñoz et al. 2013). Families with public insurance are less likely to secure hearing aids for their child despite timely diagnosis (Spivak et al. 2009; Auerbach et al. 2013).

Educational access and quality have an impact on EHDI completion rates; families with lower levels of educational attainment experience loss-to-follow-up or delayed EHDI progress (Christensen et al. 2008; Gaffney et al. 2014; Crouch et al. 2017; Cunningham et al. 2018; Thomson & Yoshinaga-Itano 2018; Deng et al. 2020; Roman et al. 2021), diagnosis (Liu et al. 2008; Holte et al. 2012; Gaffney et al. 2014; Bush et al. 2015b; Deng et al. 2020; Zeitlin et al. 2021), management (Bush et al. 2015b), and EI enrollment (Gaffney et al. 2014). Holte et al. (2012) found that infants of parents with lower levels of education attainment were on average diagnosed 7 months later than those with higher levels of education attainment, placing these infants at significantly higher risk for long-term speech/language, cognitive, and educational delays and differences. Young parental age has also been associated with less success in timely EHDI follow-up; however, there was significant variability across studies of what constitutes "young" (e.g., 15 to 19 years, <20 years, and <25 years; Christensen et al. 2008; Gaffney et al. 2014; Crouch et al. 2017; Cunningham et al. 2018; McInerney et al. 2020; Meyer et al. 2020). In addition, unmarried parental status has been associated with loss-to-follow-up for EHDI (Bush et al. 2017; Thomson & Yoshinaga-Itano 2018).

Access to and quality of healthcare in rural areas is significantly poorer than in more metropolitan areas of the country,

leading to protraction of the EHDI process (Liu et al. 2008; Hoffman et al. 2011; Bush et al. 2013, 2015; Crouch et al. 2017). Access to health care is highly related to *neighborhood and built environment factors* (e.g., housing, transportation, and safety) because home location often dictates distance from metropolitan centers due to limited transportation options for attending needed medical appointments impacting follow-up rates (MacNeil et al. 2007; Liu et al. 2008; Muñoz et al. 2011; Larsen et al. 2012; Auerbach et al. 2013; Bush et al. 2013, 2014; Hunter et al. 2016; Cunningham et al. 2018; Meyer et al. 2020; Sapp et al. 2021). In addition, maternal smoking during pregnancy is predictive of difficulty following-up with recommended EHDI phases (Christensen et al. 2008; Liu et al. 2008; Razak et al. 2021).

Lastly, *social and community contexts* contributed to completion of the EHDI process. In the United States, being of a minority race is associated with increased disadvantage across each SDOH domain (Phelan & Link 2015). For the purposes of this review, we take the current-day mainstream scientific position that there are no biological or genetic bases for racial and ethnic categories, but rather that race and ethnicity are social constructs (Long & Kittles 2003; Serre & Paabo 2004; Roberts 2013; Templeton 2013; Maglo et al. 2016; Yudell, Roberts, DeSalle, & Tishkoff 2016). As such, we categorized race and ethnicity into the social and community context domain to examine the systemic inequities and discrimination that produce health disparities (Yudell et al. 2016). Race and ethnicity are related to delayed care or loss-to-follow-up for multiple populations of color. "Non-white" families (Crouch et al. 2017; p. 42), specifically Hispanic/Latino (Christensen et al. 2008; Thomson & Yoshinaga-Itano 2018; Juarez et al. 2020), non-Hispanic Black (Zeitlin et al. 2017; Deng et al. 2020), Native American (Gaffney et al. 2014), and Hmong families (Meyer et al. 2020) are at higher risk for delays in completing EHDI steps for early identification and management. Social and community contexts can be multifaceted and subject to cultural differences related to ethnicity (e.g., families making decisions consistent with their own culture versus adopting Western medicine approaches), contexts related to the larger issues of systematic racism inherent in society and healthcare, or interactions that can be present with other SDOH factors like economic stability and built environment. Ultimately, the difficulties with navigating healthcare in America is a result of the interaction between cultural differences and system-level processes that can hinder families of certain groups effectively accessing the healthcare they need. Unfortunately, differential effects of culture versus structural racism or other SDOH have not been specifically studied in EHDI systems.

In addition to SDOH factors medical issues and pre-term birth (Chapman et al. 2011; Muñoz et al. 2011; Deem et al. 2012; Larsen et al. 2012; Findlen et al. 2019), low birth weight (Tran et al. 2016b; Crouch et al. 2017; Meyer et al. 2020), and history of prolonged neonatal intensive care unit (NICU) stay (Chapman et al. 2011; Deem et al. 2012; Giordano et al. 2015; Stuart 2016; Deng et al. 2020; Razak et al. 2021) protract the entire EHDI process. However, hospital programs with strong NICU audiology teams can moderate the negative effects of medical factors on timing of screening, diagnosis, and management (Awad et al. 2019). Transient middle ear dysfunction contributes to delays in diagnosis and timely management (Muñoz et al. 2011; Holte et al. 2012; Larsen et al. 2012; Awad et al. 2019; Findlen et al. 2019). Higher maternal depression ratings after birth are associated with higher likelihood of loss-to-follow-up (Zeitlin et al. 2019, 2021). Lastly, a general lack of adherence to follow-up recommendations leads to delays or non-completion of EHDI steps (Muñoz et al. 2011; Stuart 2016; Findlen et al. 2019). Lack of adherence to follow-up recommendations may result from parental responsibilities (i.e., work/school schedule and childcare) that present practical barriers to completing follow-up visits (MacNeil et al. 2007; Elpers et al. 2016; Hunter et al. 2016; Shanker et al. 2019; Razak et al. 2021).

System-Level Factors

Several system-level factors hinder families' progression through the EHDI process. Shulman et al. (2010) identified four main barriers to follow-up and intervention: lack of system-service capacity, lack of provider knowledge, challenges to families obtaining services, and information gaps. Lack of service-system capacity refers to limitations in the number of adequately trained providers at each EHDI step. Lack of trained pediatric audiologists for diagnosis has been noted to be a key issue, but this limitation can also extend past the birth-to-three stage into the early elementary years, including the educational setting (Ward et al. 2019). System-wide limitations in access to providers with experience serving children who are D/HH impacts not only EHDI benchmarks, but more importantly limits children from meeting their full potential (Hoffman et al. 2011; Behl et al. 2016).

Lack of provider knowledge has often manifested as physician dismissal of the seriousness of a newborn hearing screening referral or physicians taking the "wait and see" approach (Brown et al. 2006; Holte et al. 2012; Elpers et al. 2016). Key themes across studies included incomplete training for physicians in EHDI needs, contributing to referral delays and lack of understanding about the importance of follow-up. Indeed, 79% of surveyed physicians responded that their training did not prepare them to meet the needs of children who are D/HH (Brown et al. 2006). In addition, there is significant variability in the EHDI knowledge base of general family practice physicians versus pediatricians (Ross & Visser 2012; Bush et al. 2015a). This variability is related to practice setting and pediatric volume (Ross & Visser 2012), resulting in families receiving differential care depending upon what type of primary provider or practice setting they choose.

Family awareness of screening results and need for follow-up is also reported as a barrier (Bradham et al. 2011; Larsen et al. 2012; Bush et al. 2015b, 2017; Elpers et al. 2016; Juarez et al. 2020), with one study indicating particular lack of understanding of screening results and need for follow-up in fathers (Juarez et al. 2020) and another noting confusion about the entire process by extended family members, specifically grandparents (McNee & Jackson 2012). Although this could be classified as a characteristic of families, lack of knowledge really reflects a failure of the EHDI system as a whole to educate families about results, implications, and the importance of follow-up. In a survey by Larsen et al. (2012) parents reported a lack of understanding about actionable steps following diagnosis of hearing loss, including hearing aid management (45%), medical referrals (55%), EI (36%), and parent support (62%).

Screening-Specific Factors

System-level factors for screening completion and outpatient rescreening follow-up depend upon high-quality, evidence-based approaches as recommended by JCIH. In particular, multiple rescreenings are unnecessary, and delay diagnosis and intervention (Holte et al. 2012; Tran et al. 2016b). Variability in quality across screening programs relates to screener competency (training and volume of births), overall screening completion rates, and staff turnover (Christensen et al. 2008; Larsen et al. 2012; Rao et al. 2002; Sanchez-Gomez et al. 2019). Lack of involvement/oversight from an audiologist (Thomson & Yoshinaga-Itano 2018) and adequate time allowed for program quality control are key factors in program quality (Rao et al. 2002). Data fidelity (e.g., erroneous family demographics) may preclude effective outreach/follow-up (Sanchez-Gomez et al. 2019).

Diagnosis-Specific Factors

Individual and family factors related to delayed or lack of diagnosis after referral on newborn hearing screening include missed/canceled appointments (Awad et al. 2019), limited sleep-state for reliable and valid ABR testing (Holte et al. 2012; Findlen et al. 2019), and presence of either a mild degree of hearing loss (Awad et al. 2019) or progressive hearing loss (Findlen et al. 2019). Infants who passed the newborn hearing screening but have risk factors for late-onset or progressive hearing loss are less likely to complete recommended audiologic monitoring (Gehring & Jones 2017; Stich-Hennen & Bargen 2017; McInerney et al. 2020). Despite most NICU graduates having risk for delayed-onset hearing loss, many families are not informed or do not understand the need for follow-up after initial screening (Gehring & Jones 2017). Stich-Hennen et al. (2017) studied implementation of a two-class system for monitoring infants who pass newborn hearing screening but have risk factors for delayed-onset hearing loss. Less than half of the children who should have received follow-up returned for care, and for those with the highest risk (predominantly syndromic and children with cleft palate) about 25% ultimately had an educationally significant hearing loss. McInerney et al. (2020) found a higher percentage of families of at-risk infants complying with follow-up recommendations (86%), although only 7.9% of those returning completed recommended follow-up through age 24 mo.

System-level factors hindering effective diagnosis include equipment failure at the time of assessment (Holte et al. 2012) or noisy test/need for repeat testing (Larsen et al. 2012; Shanker et al. 2019). Inadequate access to evaluations occurs due to inconvenient appointment times (MacNeil et al. 2007), lack of appointment availability, not knowing where to go for an assessment (Larsen et al. 2012), and wait times between screening and diagnostic appointment (Hoffman et al. 2011). Multiple studies reported variability in diagnostic test batteries, leading to the need for multiple tests to confirm hearing loss (Windmill & Windmill 2006; Muñoz et al. 2011; Findlen et al. 2019). Given that repeated testing contributes to loss-to-follow-up (Zeitlin et al. 2021), an evidence-based, effective, and efficient test battery is critical at the diagnostic stage. Finally, some of the LTFU reported by state EHDI programs can be attributed to loss-to-documentation, as some diagnostic centers may be completing testing but not reporting results to their state EHDI program (Soto et al. 2016).

Management-Specific Factors

Type and degree of hearing loss significantly impacts the provision of hearing technology, with milder degrees of hearing loss (Muñoz et al. 2013; Awad et al. 2019), unilateral hearing loss (Spivak et al. 2009; Auerbach et al. 2013; Muñoz et al. 2013), conductive hearing loss (Spivak et al. 2009; Auerbach et al. 2013), delayed onset (Spivak et al. 2009), or progressive hearing loss (Findlen et al. 2019) having the most protracted management processes. Failure to recommend hearing aids for mild or unilateral hearing loss also protracts the hearing aid fitting process (Holte et al. 2012). For these less severe forms of hearing loss, family acceptance of the need for amplification is a factor (Holte et al. 2012; Muñoz et al. 2013). Although cost is associated with barriers at each step of the EHDI process, hearing aid cost is a particular economic burden due to the lack of, or variable and complex insurance coverage (Holte et al. 2012; Muñoz et al. 2013). For system-level factors, lack of support from primary care providers for hearing aid recommendations (Holte et al. 2012) and lack of physician medical clearance appointments delays hearing aid management (Krishnan & Van Hyfte 2014; Findlen et al. 2019).

Early Intervention Enrollment–Specific Factors

Similar to management, the type and degree of hearing loss impacts EI enrollment for children with diagnosed hearing loss. Children with lesser degrees of hearing loss or unilateral hearing loss have delayed or lower EI enrollment (Liu et al. 2008; Holte et al. 2012; Tran et al. 2016b; Crouch et al. 2017). Timing of diagnosis is also noted to have a cascading effect on enrollment in EI, resulting in delays. However, even children diagnosed by three months of age can experience delays in EI enrollment (Tran et al. 2016b). Family acceptance and the need to process the diagnosis also plays a part timely EI enrollment (Holte et al. 2012). For system-level factors, families living in rural or suburban areas often struggle with limited EI services (too few qualified EI providers or lack of hearing-specific intervention) (Liu et al. 2008; Holte et al. 2012; Giordano et al. 2015).

Evidence to the Contrary

Although there was a large corpus of evidence supporting the barriers that SDOH pose to early hearing health care, there were some studies that reported results to the contrary. Awad et al. (2019) examined factors contributing to JCIH (2019) benchmarks for diagnosis, management via hearing aid fitting, and EI enrollment in a large metropolitan hospital setting with additional clinic sites in adjacent areas. Their findings suggested that missed or canceled appointments and middle ear involvement contribute to delays in the EHDI timeline, while insurance status was not a contributing factor. In an urban safety-net hospital, Razak et al. (2021) reported that several sociodemographic factors (maternal education level, maternal age, insurance status, or ethnicity) were unrelated to diagnostic completion rates, although NICU stay and maternal smoking were significant factors in the 17% LTFU rate. Finally, Smith et al. (2019) studied socioeconomic factors in a large population of children with and without public insurance across multiple sites, finding no significant differences in hearing aid uptake, compliance, or speech/language outcomes across children who had SDOH considerations versus those who did not.

Facilitators of Effective EDHI

Across studies discussed earlier that address SDOH, many pointed to favorable SDOH as facilitators, including higher family income levels, private insurance status, relatively older parental age and higher parental level of education. Because favorable SDOH represent some of the only individual/family-level facilitators that influence healthcare more generally, system-level facilitators will be the focus of this section.

Entire EHDI Journey

One main facilitator that emerged from studies that impacted the entire EHDI journey was the provision of family support (Shulman et al. 2010). Bradham et al. (2011) highlighted the strengths of family-based organizations as having a national presence but with state and local collaborations to provide relevant information and resources to families. Ward et al (2019) further endorsed the need for family-based organizations to support families but underscored the importance of matching families based on their similar needs (e.g., location, family choice for language modality, children with additional special needs). Finally, Zeitlin et al. (2019) demonstrated the impact that social and family supports had on follow-up by showing that access to education, healthcare providers, and family members to help with the process can mitigate the negative impact of maternal depression on follow-up. A secondary aspect of family support includes providing resources for financial support. Christensen et al. (2008) noted that hospitals that provided free outpatient rescreenings showed higher follow-up rates, while Rao et al. (2002) demonstrated that hospitals that incorporated screenings as a part of "standard of care" were able to achieve better reimbursement rates that reduced the financial burden for families. Finally, the major barrier to effective management of hearing loss via hearing technology is financial. Munoz et al. (2013) suggested that implementing loaner hearing aid programs and assisting with identifying financial support for families will facilitate the timely fitting of hearing aids to maximize early access to sound.

Related to family support is the aspect of care coordination/navigation. In the only randomized control trial represented in the final set of 62 articles reviewed, Bush et al. (2017) reported that the provision of a care navigator for facilitating EHDI for rural families was associated with improved adherence to follow-up recommendations, earlier diagnosis, and an increase in parents' baseline knowledge of infant hearing loss and recommendations. Care coordination with pediatricians (Christensen et al. 2008; Seeliger et al. 2016; Zeitlin et al. 2021), with diagnostic testing sites (Seeliger et al. 2016; Al-Mulki & Todd 2020Al-Mulki & Todd 2020) and using hospital Child Find services to coordinate care has also been shown to improve diagnostic follow-up (Deem et al. 2012). Proactively scheduling screening and diagnostic follow-up appointments at the time of need instead of shifting the responsibility to parents for calling to schedule at a later time significantly improves completion of follow-up (Krishnan & Van Hyfte 2014; Thomson & Tran et al. 2017; Yoshinaga-Itano 2018; Zeitlin et al. 2021). Lastly, partnering with community services like the Women, Infant, and Children (WIC) Program allows for better communication of needed follow-up and the potential for colocating follow-up services at appointments that are already being completed for infants who were lost to follow-up (Hunter et al. 2016; Zeitlin et al. 2021).

To address the issue of loss-to-documentation (LTD) as opposed to LTFU, Sanchez-Gomez et al. (2019) demonstrated that linking EHDI data with other sources of information (e.g., Vital Statistics) can help with locating families who have been LTFU or LTD. Soto et al. (2016) linked Medicaid billing data and was able to identify a subset of infants who were LTD as opposed to LTFU. These studies provide tangible solutions to the need identified by Shulman et al. (2010) to improve data systems for effective data tracking and surveillance of follow-up activities.

Screening-Specific Factors

Familiarity with screening sites has been found to positively impact completion of outpatient rescreening recommendations. Returning to the birth nursery, having on-site primary care clinics, and completing outpatient rescreenings at pediatrician well-checks or WIC clinics have been shown to facilitate outpatient screening completion (Christensen et al. 2008; Hunter et al. 2016; Thomson & Yoshinaga-Itano 2018). Hunter et al. (2016) reported that babies served by WIC whose families were low income and included high numbers of Black or Hispanic families were 75% more likely to follow-up and 90% more likely to complete diagnostic testing when offered follow-up screening at WIC, compared with nonintervention rates.

Diagnosis-Specific Factors

The quality of the diagnostic assessment and the preparation of families for the diagnostic appointment can significantly improve diagnostic assessment completion and timing. Facilitation of quality testing can be achieved by using an evidence-based test battery with proactive scheduling (scheduling the diagnostic at the time of referred screening or scheduling follow-up testing before families leave medical facilities) (Findlen et al. 2019; Sapp et al. 2021; Zeitlin et al. 2021), using more experienced clinicians, and providing preappointment instructions to families to maximize the natural sleep state (Awad et al. 2019; Shanker et al. 2019; Zeitlin et al. 2021). Ultimately, these approaches can lead to fewer appointments needed for confirmation of hearing loss and earlier diagnosis.

Increasing access to diagnostic testing through outreach and teleaudiology services can also reduce LTFU. Sapp et al. (2021) completed an outreach study in which educational audiologists were trained and supported to provide diagnostic services in underserved areas of Iowa. Implementation of this intervention resulted in a 10% reduction in LTFU and decrease in age at diagnosis by almost 1 mo. Teleaudiology has also been used to provide services to areas of states with limited access to diagnostic services. Results suggest that high-quality diagnostic assessments are possible with teleaudiology with concurrent high parental satisfaction (Dharmar et al. 2016; Stuart 2016; Meyer et al. 2020Meyer et al. 2020). Unfortunately, audiologists' perception of telehealth effectiveness and lack of insurance coverage are barriers to wide-spread implementation (Prins et al. 2021). Finally, implementing a process to effectively evaluate infants who pass the newborn hearing screening but have risk factors for late-onset or progressive hearing loss is needed. Adhering to JCIH (2019) risk factor monitoring guidelines with both parent and physician education can serve to facilitate ongoing monitoring of this group of infants that are often LTFU (Stich-Hennen & Bargen 2017; McInerney et al. 2020).

Early Intervention Enrollment–Specific Factors

Aside from the facilitation of on-time EI enrollment through early screening and early diagnosis (Tran et al. 2016b; Roman et al. 2021), there were few studies that directly addressed the

facilitation of EI enrollment for infants and children who are D/HH. Crouch et al. (2017) reported that infants are more likely to be enrolled in EI if they present with a profound degree of hearing loss, presumably due to the significant impact that degree of hearing loss can have on development. However, additional research is needed around EI enrollment in this population.

DISCUSSION

This systematic review set out to examine and synthesize the barriers to and facilitators of EHDI in the United States with the aim to inform program stakeholders about what factors can and should be addressed to help children who are D/HH meet their full potential. Although Holte et al. (2012) reported that only 32% of infants in their study completed each of the 1-3-6 benchmarks by the targeted timeline, this number shrinks to less than 20% for national-level data (Subbiah et al. 2018), indicating continued difficulty with meeting EHDI expectations and suggesting a significant need to address barriers. Many of the barriers identified through this review have been reported by EHDI programs in other developed or developing countries (Lai et al. 2014; Ravi et al. 2016; Fitzpatrick et al. 2017; Naidoo & Khan 2022; Waterworth et al. 2022). A common theme across the journey for these US studies was that specific SDOH served as significant barriers, consistent with a recent review by the American-Speech-Language-Hearing Association Special Interest group for EHDI (Kingsbury et al. 2022).

Although individual/family-level factors related to the negative impact of SDOH represent a wide-spread problem in healthcare (Braveman & Gottlieb 2014), this systematic review was able to identify that system-level changes can mitigate the negative impact of SDOH factors and facilitate EHDI services for children. A key aspect of studies that did not report negative influences of SDOH on meeting EHDI milestones (Awad et al. 2019; Smith et al. 2019; Razak et al. 2021) is that they took place at large medical centers with diverse patient populations and likely have system-level approaches to patient care in place that mitigate the negative impact of SDOH on pediatric healthcare. For example, the Awad et al. (2019) study was completed in a metropolitan area with 8 adjacent satellite sites to improve regional access to services. The Smith et al. (2019) study cited the possibility that a regional EI program allowed families with lower socioeconomic status to adequately access resources for management and habilitation of hearing loss. Finally, the Razak et al. (2021) study was carried out at an urban safety-net hospital with the specific mission to provide services to the community regardless of insurance status or ability to pay, eliminating the cost concern that many studies report as a barrier across the EHDI benchmark time points. These studies collectively suggest that instituting system-level facilitators of EHDI, such as regional programs and care coordination to facilitate access and uptake of services, can potentially mitigate the barriers that accompany SDOH.

The overarching theme of system-level facilitators identified in this review is having systems that meet the family where they are. Reviewed articles described a number of different ways to facilitate the EHDI journey. Care coordination/navigation has been shown to be highly effective in shepherding families through medical care for children with complex medical needs and to be cost effective by preventing delays in treatment (Antonelli et al. 2008; Ruggiero et al. 2019). Two secondary articles in this review demonstrated that lack of EHDI resources specific to care coordination can contribute to increased LTFU (Hoffman et al. 2011; Smith et al. 2019). Bush et al. (2017) demonstrated the effectiveness of care coordination for rural families seeking EHDI services specifically, however other articles have shown effectiveness for families in urban and suburban areas as well (Deem et al. 2012; Tran et al. 2017; Al-Mulki & Todd 2020). As such, care coordination may be one facilitator that is effective regardless of family geographic location. Care coordination could further solve an overarching issue within the EHDI system in that screening, diagnosis, and EI occur in different physical places and by different entities. Screening usually occurs at the birthing facility/nursery as a part of inpatient services, but secondary outpatient screenings can occur in community clinics or physician offices depending upon the program. Diagnosis often occurs through outpatient clinics that can be affiliated with large hospital systems or small private practices. Finally, EI can take the form of in-home services overseen by a government agency or center-based programs with private practitioners. Care coordination/navigation can prevent families being LTFU at multiple places along this disjointed system. In addition, improved systems to link data between entities overseeing and involved in screening, diagnosis, and EI should be studied and implemented to address loss-to-documentation.

Related to care coordination is the idea of colocating services. This review found that pairing follow-up services with other medical appointments (Christensen et al. 2008) or social services (Hunter et al. 2016; Zeitlin et al. 2021) contributed to reducing LTFU. This is consistent with previous reports from the Vermont EHDI Program, which facilitated a quality improvement study to reduce LTFU for outpatient rescreening by partnering with physicians throughout Vermont to complete needed follow-up in each county (Jordan & Hazard 2015). Parents have endorsed that colocating services is helpful in facilitating care following diagnosis of hearing loss in addition to care coordination (Fitzpatrick et al. 2008). One aspect of colocating services at the screening level that warrants additional research includes ensuring infants who are birthed at home can be provided the services they need. Palmer et al. (2019) showed that providing focused education and access to equipment to midwives successfully increased the odds of a home-birthed baby having a completed newborn hearing screening by 1 mo of age. For completion of diagnostic assessment, teleaudiology can also be successful in serving families where they are for diagnosis. Additional evidence of the successful implementation of teleaudiology services for diagnosis of late-onset hearing loss in rural school-aged children was recently published (Emmett et al. 2022). There is a need to address insurance coverage issues and audiologists' perception of this service delivery model to expand teleaudiology for EHDI follow-up (Prins et al. 2021).

Family support through high-quality parent education from well-qualified providers and increased access to family-to-family interaction is needed to overcome barriers to EHDI. Families report hearing-specific medical information, therapy-related information including types of therapy and locating services, and prognosis about communication development is essential when navigating a new diagnosis of hearing loss in infants (Fitzpatrick et al. 2008). Key to high-quality parent education is the role of qualified providers with expertise in the unique needs of young D/HH children and their families (JCIH 2013; Moeller et al. 2013). Specific to parent-to-parent support, Henderson et al. (2014) calls on EHDI programs to establish parent-to-parent

support as a high priority. Parent-to-parent support should be incorporated within and complementary to EHDI programs, and professionals serving parents should be informed of peer parent support programs (Henderson et al. 2014). Further, families strongly communicated the need for parent support from other parents who have experienced the journey, particularly in the form of local parent support groups (Haddad et al. 2019) and that parent-to-parent support should be an integral part of any EHDI program. Clear directives for follow-up, unbiased information about management and communication options, and information in language families understand was supported by secondary articles in this review (Bush et al. 2013, 2015; Muñoz et al. 2013; Behl et al. 2016; Elpers et al. 2016; Sax et al. 2019). However, we identified few articles that definitively examined the efficacy of parent education materials and parent-to-parent support, speaking to the need for additional research in these areas.

Finally, Smith et al. (2019) recommended implementing a national standard for screening, diagnosis, and EI to facilitate favorable outcomes for all children by decreasing the variability that exists within and across states. Screening systems vary across states and programs, with some adopting an inpatient-only approach while others use an inpatient-to-outpatient rescreen protocol. There is some evidence that an inpatient-only approach can facilitate screening completion and reduce LTFU for the diagnostic phase (Roman et al. 2021), however, there is also evidence from annual Centers for Disease Control and Prevention EHDI data (CDC 2022) that states with an inpatient-to-outpatient rescreening process can also be successful with screening and diagnosis completion. Variability in service provision is also a barrier leading to LTFU at the diagnostic and EI enrollment phases. Although evidence-based practice exists for diagnostic assessments to reduce variability and facilitate effective early diagnosis (Hatton et al. 2012; Sutton et al. 2013; Hyde et al. 2016; JCIH 2019; American Academy of Audiology 2020), additional research is needed to understand if there are more efficacious screening and EI enrollment approaches to successfully complete these benchmarks. This is critically important considering continued evidence that earlier EI enrollment, before six months of age is a critical predictor of spoken language outcomes (Grey et al. 2022) and kindergarten readiness (Meinzen-Derr et al. 2020). Even earlier EI, by three months of age, is now being advanced to optimize global language development (Walker et al. 2022).

Limitations

There are several limitations of this systematic review. Generalizability across EHDI programs or to other countries may be limited given that this review focused only on the unique US system and each state executes their EHDI program differently. Complex interactions among factors that might affect EHDI follow-up cannot be accounted for, and this limits our ability to understand significant vs. non-significant factors and how they may compound barriers or work together to mitigate barriers. Finally, most of the articles focused on the screening and diagnostic phases of EHDI, with relatively limited research available for management and EI enrollment and execution. This limits our ability to understand additional factors that may serve as barriers or facilitators at those time points. Overall, additional high-quality research is needed to evaluate factors especially at the EI–enrollment phase as well as efficacy for various approaches to family support across the EHDI journey.

CONCLUSION

Effective early hearing detection and intervention is essential for children who are born D/HH to meet their full potential. Many barriers exist for families to obtain appropriate and timely services, but system-level changes could facilitate the process and contribute to long-term outcomes improvement. Rigorous evaluation of system-level/programmatic facilitators, including care coordination, family support, and data linkage opportunities, is necessary to understand how EHDI programs can effectively improve the quality of their systems. Additional research is needed to identify the most effective solutions to facilitate EHDI world-wide in consideration of regional and country-level circumstances.

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