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Sociodemographic Factors Affecting Loss to Follow-Up After Newborn Hearing Screening: A Systematic Review and Meta-analysis

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Abstract

Objective.—Universal newborn hearing screening (NBHS) has been widely implemented as a part of early hearing detection and intervention (EHDI) programs worldwide. Even with excellent provider knowledge and screening rates, many infants do not receive definitive hearing testing or intervention after initial screening. The objective of this study was to identify sociodemographic factors contributing to loss of follow-up.

Data Sources.—PubMed, Scopus, and CINAHL.

Review Methods.—Per Preferred Reporting Items for Systematic Reviews and Meta-analyses guidelines, the databases were searched from the date of inception through December 28, 2021. Studies containing sociodemographic information on patients who were referred to NBHS were included. Meta-analysis of odds ratios (ORs) was performed comparing rates of sociodemographic variables between patients adherent and nonadherent to follow-up.

Results.—A total of 169,238 infants from 19 studies were included. Low birth weight (OR 1.6 [95% confidence interval, CI 1.2-2.2, $p < .001$), racial minority (OR 1.4 [95% CI 1.2-1.6], $p < .001$), rural residence (OR 1.5 [95% CI 1.1-1.9], $p = .005$), lack of insurance (OR 1 [95%

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Author Contributions

Kelly M. Atherton, contributed to the design, conduct, analysis, and drafting of the manuscript; **Nicolas S. Poupore**, contributed to the design, conduct, analysis, and drafting of the manuscript; **Clarice S. Clemmens**, contributed to the critical editing of the manuscript; **Paul J. Nietert**, contributed to the analysis and critical editing of the manuscript; **Phayvanh P. Pecha**, contributed to the design, conduct, analysis, and critical editing of the manuscript.

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Supplemental Material

Additional supporting information is available in the online version of the article.

CI 1.4-2.5], $p < .001$), and public or state insurance (OR 1.7 [95% CI 1.2-4.2], $p = .008$) were associated with missed follow-up after referred NBHS. Associated maternal factors included low maternal education (OR 1.8 [95% CI 1.6-2.0], $p < .001$), young maternal age (OR 1.5 [95% CI 1.5-1.6], $p < .001$), unmarried maternal status (OR 1.5 [95% CI 1.1-1.9], $p = .003$), and current or former maternal smoking status (OR 1.8 [95% CI 1.4-2.2], $p < .001$).

Conclusion.—Both infant and maternal sociodemographic factors influence follow-up compliance after referred NBHS. Focused efforts should be made by medical providers and policymakers to address these factors to ensure appropriate newborn hearing care and interventions are achieved.

Keywords

early hearing detection and intervention; loss to follow-up; meta-analysis; newborn hearing screening; systematic review

Congenital hearing loss is a relatively frequent congenital anomaly, with an estimated prevalence of moderate or severe bilateral hearing deficit in 1 to 3 per 1000 live births within the well-baby nursery population,[1–3] and 2 to 4 per 100 live births within the neonatal intensive care population.[1,4,5] Unrecognized hearing loss may contribute to delayed development in speech, reading, cognition, and social-emotional development, which can manifest later as poor educational achievement, adverse psychological effects, and lower employment levels.[6–9]

For this reason, universal newborn hearing screening (NBHS) has been widely implemented as a foundational component of early hearing detection and intervention (EHDI) programs. Prompted by the Joint Committee on Infant Hearing (JCIH) Year 2000 and 2007 Position Statements, most states strive to achieve the 1-3-6 protocol, which encourages screening infants by 1 month of age, identifying hearing loss by 3 months of age, and intervention by 6 months of age.[9,10]

Even with excellent provider knowledge and screening implementation at birth, many individuals are lost to follow-up after discharge, never meeting the 3-month diagnosis or 6-month intervention benchmarks. While the JCIH initially recommended a goal of a 95% follow-up compliance rate for those infants who were referred to initial hearing screening,[10] others have suggested lower benchmarks, such as 70%.[11] In practice, studies have reported a wide spectrum of loss to follow-up (LTF) rates following NBHS ranging from 0.31% to 68%.[12–14] One 2016 systematic review by Ravi et al.[13] estimates an average LTF of 20% for single-center studies and 22% for multicenter studies.

Two other systematic reviews by Ravi et al.[15,16] have characterized caregiver and health care professionals' knowledge of and attitudes toward NBHS, and the 2016 systematic review represents the most thorough characterization of reasons for LTF and strategies to reduce LTF to date, with commonly cited factors for LTF including lack of parental knowledge, distance from the testing facility, work constraints, and oppositional parental attitude.[13] However, the important and relevant sociodemographic characteristics that may be contributing to nonadherence after referred NBHS warrant further characterization

through a meta-analysis. Therefore, the overall aim of the present systematic review was to identify and assess infant and maternal sociodemographic factors associated with LTF.

Methods

Search Criteria

Adhering to the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines,[17] we performed a systematic review of studies looking at LTF after NBHS. With the assistance of a professional librarian at our institution who specializes in systematic reviews, detailed search strategies were developed for PubMed (US National Library of Medicine, National Institutes of Health), Scopus (Elsevier), and CINAHL (EBSCO). The concepts “newborn hearing screen” and “sociodemographic factors” were used to develop a search strategy using a combination of subject headings (eg, MeSH in PubMed) and keywords. The PubMed search strategy was appropriately altered for the other 2 databases by maintaining similar keywords and replacing MeSH terms with similar subject headings when available. Databases were searched from the date of inception through December 28, 2021. Appendix 1 describes the detailed search strategies, filters applied, and the number of results for each database. Additionally, the reference lists of relevant articles were manually searched to validate the search strategy to ensure all important articles were identified. The review management software Covidence (Veritas Health Innovation Ltd.) was used for study selection.

Selection Criteria

The inclusion criteria consisted of studies describing sociodemographic factors in LTF infants who referred their NBHS. Studies were also required to report the same sociodemographic factors in those infants who appropriately followed up so that 2 cohorts, based on follow-up status, were defined for each sociodemographic factor of interest. LTF was defined as never attending an appointment for diagnostic testing. Although EHDI guidelines recommend this visit by 3 months of age, many infants are delayed in follow-up. Rather, this study meta-analysis seeks to examine adherence with follow-up in general, not compliance with EHDI guidelines specifically. Studies that lacked sociodemographic factors (eg, race, insurance status, income), reported on only 1 cohort (followed-up or failed to follow-up), or pertained to hearing screening not related to NBHS (eg, children screened for hearing loss later in childhood or related to a specific disease process) were excluded. Double- or single-blinded randomized controlled trials, double- or single-blinded randomized comparison trials, nonrandomized controlled trials, and prospective or retrospective observational studies were considered for inclusion. We excluded studies that were non-English language, non-human studies, review articles, case reports of less than 4 patients, duplicates, and inaccessible articles.

Titles and abstracts were first independently assessed by 2 reviewers (K.M.A. and N.S.P.) to identify all articles that met the inclusion criteria, and conflicts were resolved by discussion with the senior author (P.P.P.). Two reviewers (K.M.A. and N.S.P.) then assessed the full texts of articles independently to identify those that met all inclusion and exclusion criteria

for the final analyses. Any disagreements or discrepancies were resolved by way of a third author (P.P.P.).

Quality Assessment

A critical assessment of the evidence level of included articles was done using the Oxford Center for Evidence-Based Medicine criteria.[18] Risk of bias was assessed following the Cochrane Handbook for Systematic Reviews of Interventions Version 6.0.[19] The Risk of Bias in Non-Randomized Studies—of Interventions (ROBINS-I) tool was used because all included studies were nonrandomized controlled trials.[20] This tool assesses the bias due to confounding, bias in the selection of participants into the study, bias in the classification of interventions, bias due to deviations from intended interventions, bias due to missing data, bias in the measurement of outcomes, and bias in the selection of the reported results. The risk of bias for each variable was graded as low, unclear, or high. Two authors (K.M.A. and N.S.P.) performed an independent pilot assessment on 3 studies and compared results to check for consistency. Both authors then completed an independent risk assessment on the remaining studies, with all disagreements being resolved by the senior author (P.P.P.).

Data Extraction

Two reviewers (K.M.A. and N.S.P.) independently reviewed the studies included in the data extraction process and compared them for accuracy. Data points included author, year of publication, country of the population studied, study design, sample size, and a number of subjects possessing sociodemographic factors of interest, stratified by follow-up status. We attempted to further categorize those sociodemographic factors as patient-specific (belonging intrinsically to the infant), maternal-specific, or shared, where the latter refers to a factor shared by both mother and infant, or where a variable could have been specific to mother or patient, but was not clearly specified within each article (eg. race).

Variable Definitions

Maternal or infant race/ethnicity was dichotomized into minority and nonminority groups based on the demographic setting of the study. For most studies, nonminority groups consisted of white and non-Hispanic white. Minority groups included Hispanic, black, American Indian, Alaskan Native, Asian, or Pacific Islander. For Prince et al.,[21] which was conducted in Hawaii, racial minority was defined as those representing <20% of the population, including Native Hawaiian or Other, and non-minority races included white, Asian, and Filipino. For Olusanya[22] and Olusanya and Akinyemi,[23] racial minority was defined as not belonging to 1 of the 3 most predominant tribes in Nigeria. Cavalcanti and Guerra[24] and Kanji and Khoza-Shangase[25] categorized subjects as infants of primiparous or multiparous mothers, or as first or not first-child, respectively. Cunningham et al.[26] reported infant birth order as 1st, 2nd/3rd, or ≥4th, while Folsom et al.[27] reported mothers' number of children as 1, 2, or >2. We divided participants into 2 categories: infants of primiparous mothers and multiparous mothers. Birth weight is sometimes regarded as a proxy measure for other sociodemographic factors, and was therefore included as a predictor of LTF.[28]

Low maternal education was defined, with 1 exception, as primary school only, some high school or less, or high school degree/general educational diploma, while high maternal

education was defined as secondary school, some college, college degree, or postgraduate degree. Griz et al.[29] categorized education by middle versus high/graduate school, so in this case, middle school was included as low maternal education, and high/graduate school was included as high maternal education. Maternal age was defined as young if age was <20 years of age, with 1 exception. Christensen et al.[30] categorized maternal age as 13 to 24 years and ≥25 years, so in this case, 13 to 24 years were included as young maternal age, and ≥25 years were included as not young. Two studies initially divided maternal smoking status into 3 groups: current smokers, former smokers, and never smokers.[21,31,32] Of the remaining studies, only 2 differentiated smoking during pregnancy,[30,33] while 1 study simply differentiated lifetime smokers and nonsmokers.[26] For the purposes of this study, subjects were divided into 2 categories: (1) current or former and (2) never smokers.

Statistical Analysis

Categorical data were compiled using frequencies with percentages. Meta-analysis of odds ratios (ORs) (comparison of rates of sociodemographic variable between patients adherent and nonadherent to follow-up) was performed with Cochrane Review Manager (RevMan) version 5.4 (Nordic Cochrane Centre, Cochrane Collaboration, 2011) for dichotomous sociodemographic variables and SAS v9.4 (SAS Institute) for health insurance, the only trichotomous demographic variable. Both fixed-effects and random-effects models were used depending on the I^2 or heterogeneity test results for each sociodemographic factor.[34,35] If an analysis suggested moderate or high heterogeneity by means of $I^2 > 50\%$, then the fixed-effects model might be invalid.[36] In these cases, the random-effects model would be more appropriate and was therefore utilized.[34,35] A random-effects model incorporates both the random variation within and between different studies to account for the high heterogeneity seen in that particular analysis.[34,35] Mantel–Haenszel ORs were calculated, using inverse variance weights, a feature of RevMan software.[37]

Egger's test with funnel plots was performed to assess the risk of publication bias. In a funnel plot, the effect is plotted on the horizontal axis, and the standard error is plotted on the vertical axis.[38] The vertical line represents the summary effect estimated using a fixed-effects meta-analysis. Around the summary effect are 2 diagonal lines that represent the 95% confidence limits, calculated using the formula effect ± 1.96 standard error for each standard error on the vertical axis. These lines visually depict the expected distribution of studies. In the absence of heterogeneity or selection bias, 95% of all studies should lie below and within the funnel. Furthermore, publication bias results in an asymmetry of the funnel plot and can be visually assessed. A p value of <.05 was considered to indicate statistical significance for all tests.

Results

Study Selection and Characteristics

A literature search of all 3 databases identified 1623 articles, of which 436 duplicate records were removed. Title and abstract screening excluded 1023 articles. Full-text review of the remaining articles excluded 145 articles, leaving 19 remaining articles for inclusion in the final data extraction and analysis.[21–27,29–33,39–45] A diagram outlining the complete search

process is included in Figure 1. A critical assessment of the included studies (Figure 2) indicated an acceptable level of risk of bias for most of the included studies. Potential sources of bias were most evident in bias in the selection of participants for the study, bias due to missing data, and bias in the selection of the reported results. The low risk for publication bias was endorsed by a nonsignificant Egger's test (−0.6 [95% confidence interval, CI −1.6 to 0.5], $p = .243$) and a symmetrical funnel plot with all studies falling within the funnel (Supplementary Figure 1, available online). Of the articles selected for inclusion, 14 were level 3b retrospective case-control or cross-sectional studies, and 5 were level 2b prospective cohort studies based on the Oxford Level of Evidence and were published between 2000 and 2022 from 5 different countries. The descriptive features of these articles are summarized in Table 1.

A total of 169,238 infants were included from all studies, of which 98,300 failed to follow-up after being referred to NBHS. Sample sizes varied considerably, ranging from 41 to 116,513 patients.[40,44]

Infant-Specific and Shared Sociodemographic Factors

Low infant birth weight had higher odds of LTF compared to infants without low birth weight (OR 1.60 [95% CI 1.16-2.21, $p < .001$). Birth weight, as described here, was not reported in connection with prematurity, and the association between these 2 factors is outside the scope of this study. The rural location of residence had higher odds of LTF compared with the urban location of residence (OR 1.47 [95% CI 1.12-1.91], $p = .005$). Regarding health insurance, patients with uninsured or self-pay status had higher odds of being lost to follow-up (OR 1.75 [95% CI 1.41-2.46], $p < .001$) than privately insured patients, as did patients with public or state insurance (OR 1.72 [95% CI 1.19-4.15], $p = .008$). Infants with a minority racial classification and infants of mothers with a minority racial classification had higher odds of being LTF (OR 1.40 [95% CI 1.21-1.62, $p < .001$), compared to subjects who were of a nonminority race. Of all the sociodemographic factors analyzed here, race was the factor reported by the highest number of included studies (14/19). Nine studies reported maternal race,[22,23,30,32,33,39,40,44,45] 3 reported infant race,[27,31,41] and 2 did not specify if the reported race belonged to the infant or mother.[21,26] Racial and ethnic categories varied between studies, but most frequently were categorized as white, black, Hispanic, Asian, and other. Less frequently reported groups included American Indian/Alaskan Native,[26,27,39,41] Native Hawaiian/Pacific Islander,[21,26,39,40] and Filipino.[21] These different classifications and the sometimes separation of race and ethnicity within studies made a comparison of individual racial categories difficult. Forest plots of the dichotomous factors are displayed in Supplementary Figures 2–4 and Table 1, available online summarizes results across studies' health insurance categories.

Maternal-Specific Sociodemographic Factors

Unmarried maternal marital status had higher odds of LTF (OR 1.46 [95% CI 1.14-1.87], $p = .003$), while low maternal education level was associated with higher odds of LTF (OR 1.79 [95% CI 1.56-2.04], $p < .001$). Young maternal age demonstrated higher odds of LTF (OR 1.52 [95% CI 1.45-1.59], $p < .001$), and current or former maternal smoking status was associated with higher odds of LTF (OR 1.75 [95% CI 1.38-2.23], $p < .001$). Maternal

primiparous status, or firstborn infant status, showed no significant difference in odds of LTF (OR 0.70 [95% CI 0.23-2.14], $p = .53$) compared to maternal multiparous status. Forest plots of these variables are displayed in Supplementary Figures 5–9, available online

In summary (and as listed in Table 2), factors associated with significantly increased odds of being LTF include having public/state insurance or being uninsured, low maternal education, current or former maternal smoking status, low infant birth weight, young maternal age, rural location of residence, unmarried maternal status, uninsured or self-pay status, and racial minority classification.

Discussion

Current literature addressing LTF after referred NBHS lacks a concerted focus on the effect of individual sociodemographic predictors. Therefore, the goal of this study was to perform a meta-analysis of existing sociodemographic data in the literature that covers LTF in the setting of EDHI. This work expands upon the most recent systematic review by Ravi et al.[13] that identified other nonsociodemographic reasons for LTF, primarily unfavorable attitudes and low priority of hearing concerns by parents. Our aim was to identify underlying factors that contribute to these more factual reasons for LTF. We found that infant-specific, maternal-specific, and shared sociodemographic are all associated with LTF after referred NBHS.

The findings of this meta-analysis largely agree with studies of compliance pertaining to other recommendations or interventions within the infant and pediatric population. In these contexts, sociodemographic factors contributing to noncompliance have included race, gender, insurance status, geographic location, residence in areas of high unemployment, lower parental education, and inadequate prenatal care.[46,47]

Sociodemographic factors are defined differently and possess contextual nuance within the cultures and societies they represent. Thirteen out of 19 studies included in our meta-analysis were conducted in the United States, and overall, most of the included studies originated from high-income countries, which may experience different contributing factors to LTF than lower-income countries. In this meta-analysis, young maternal age, education level, and minority race were defined differently by the individual studies included, especially those conducted outside the United States, indicating that these factors have varying culturally defined implications on behavior and access to resources. The impact of racial or ethnic minorities is complicated to study, given how identity is defined and classified in different cultures. All studies with individually significant findings concerning race were conducted in the United States, indicating that race is an important factor affecting follow-up rates in this country, though may not play as significant a role or maybe underrecognized in other countries. It is also possible that “minority” racial groups would be better characterized as underrepresented groups since some groups may constitute a large proportion of the population but may experience disproportionate access to care and resources. Race may have a confounding relationship with other risk factors for LTF, such as education, income, or insurance coverage, depending on the sociocultural climate of an individual country or region.

Other important regional and cultural considerations concern disseminating and implementing NBHS and EDHI. As NBHS is a new concept within the last 30 years, its implementation worldwide has progressed at different rates and has been met with different degrees of acceptance. The studies included here range from 2002 to 2021, representing almost 20 years of implementation. Overall, the NBHS literature indicates improving rates of LTF over this timeframe, as more efficient systems for follow-up and interventions to prevent LTF have been implemented. Several studies identified during the systematic review portion of this study identify parental refusal or oppositional attitude as reasons for LTF. Congenital hearing loss is viewed differently based on cultural attitudes toward disability and related interventions, with some cultures not viewing congenital hearing loss as a condition needing treatment, or as a potential social limitation.[48–50] These beliefs constitute a recognized, but difficult to qualitatively assess factor impacting parents' choice not to follow-up after referred NBHS.

Considering the strength of association for the variables examined in our study, no one factor emerged with noticeably larger odds of LTF. This may indicate the multilevel effect of sociodemographic characteristics, rather than the role of any 1 factor in driving LTF. Sociodemographic factors convey the physical and societal environment in which patients live and, therefore, help explain fundamental underlying reasons for LTF. It should be noted that factors affecting compliance and LTF in the infant and pediatric populations can be distinctly different than the adult population considering that infants are entirely reliant on parental or caregiver support for their medical care.

For instance, low maternal education can often indicate a lower level of health literacy,[51–53] which can, therefore, affect awareness of the importance of timely diagnosis of congenital hearing loss. Similarly, younger and unmarried mothers may lack sufficient social support, making it more difficult to attend follow-up appointments in the face of competing priorities such as work or additional childcare responsibilities.

Those living in rural locations often need to travel further distances to appointments and have markedly less access to public transportation.[54] This additional travel time and the need for reliable personal transportation pose notable barriers to care. Additionally, fewer care providers, clinics, and hospitals equipped for testing and follow-up are located in rural locations.[54] Insurance coverage is region and country-specific, but public insurance or uninsured status is typically associated with a lower income level.[55,56] NBHS-related costs are highly variable,[57] with private insurance coverage determinations decided by individual states in the United States.[58] While most insurance plans in the United States cover initial NBHS, follow-up visits are subject to copayments, and uninsured individuals may be responsible for all associated charges, requiring significant out-of-pocket costs. For those families already facing financial hardship, uncertainty about the cost of screening, additional testing, follow-up, and possible treatment for hearing loss may preclude their participation in EDHI programs.

Our study further endorses that infant and maternal sociodemographic factors need to be important considerations for programs aimed at reducing LTF rates. However, addressing sociodemographic variables can be challenging. Most measures taken to reduce LTF have

addressed institutional or systematic shortcomings by emphasizing database management and scheduling reminders, improving documentation, implementing tracking systems, and developing virtual or tele-audiology opportunities for follow-up.[13,59–61] Innovations in technology have improved the record-keeping and follow-up capabilities of clinics and hospitals, yet these interventions do not specifically target those at disproportionate risk of LTF. Some measures have partially addressed sociodemographic barriers, such as increasing parental education, providing financial support, and utilizing community health care workers,[13] but again may not target those individuals at the highest risk. Given the finite nature of time, personnel, and resources possessed by EDHI programs, measures to reduce follow-up may be more efficient and yield better results when applied to those most at risk of LTF, such as those possessing the sociodemographic characteristics identified by this study.

One innovative approach to the issue of LTF is currently being investigated by Matthew Bush et al.[62] at the University of Kentucky, where this team is currently implementing a community-engaged patient navigation intervention at state-funded EHDI clinics. This program involves patient navigators, who are individuals trained to help patients navigate the health care system and overcome personal, interpersonal, and environmental barriers to treatment. If shown to be successful in this initial study, patient navigation could represent a viable option for interventions targeted at mothers and infants with specific sociodemographic risk factors.

Limitations of this study include a high level of variability in terms of the sociodemographic factors reported in the included studies. Fourteen of 19 studies included in this meta-analysis were retrospective case-control or cross-sectional designs, many of which relied on the hospital, state, or regional databases, which are convenient sources of large numbers of participants but limit generalizability. Additionally, these databases collect different information and often contain large amounts of missing data which cannot be addressed due to deidentification. While all of the studies included here utilized some form of universal NBHS, individual protocols differed in terms of timing and location of testing, provider type administering screening, and recommended follow-up procedure. Some studies utilized a 1-step screening process prior to diagnostic evaluation, while some used a 2-step screening, creating 2 opportunities for LTF before the 3-month EDHI benchmark. Future studies should delineate between nonadherence with follow-up by 1 month of age (the benchmark set by EDHI) and complete LTF. LTF, as defined within our meta-analysis, refers to the latter since included studies did not draw this distinction.

The type of audiology testing used varies in the universal NBHS literature, as it did in the included studies, likely due in part to resource availability. Large tertiary academic institutions often have access to new and functional equipment, whereas small or rural locations may struggle with few, old or faulty equipment that cannot be readily repaired, further exacerbating the effect of rural locations on LTF. Lastly, not all sociodemographic factors contributing to LTF were able to be analyzed due to a limited number of studies reporting on these factors. For instance, socioeconomic status, which captures a combination of education, social class, and income, was only reported in 2 studies,[24,29] and could provide a more nuanced measure of sociodemographic profile than education or income

alone. Place (hospital vs outside of the hospital) and mode of delivery (vaginal vs cesarean section)[22,23,32] were variably reported, and potentially reflect underlying factors such as transportation availability and cultural practices, as well as sociodemographic factors such as wealth and education. Interestingly, 1 study, in particular, identified LTF rates in Appalachian versus non-Appalachian regions within Kentucky.[54] This unique approach to regional geographic landscapes and recognized sociodemographic differences based on those areas provides useful and specific information for the communities addressed but is not widely applicable to the general NBHS population.

In order to accurately assess the actual LTF rate and the factors influencing it, NBHS protocol must move toward standardization. Overall, the heterogeneity of results observed in the individual studies included here emphasizes the need for standardized definitions, reporting methods, common terminologies, and protocols associated with NBHS programs to increase the generalizability of future results.[13]

Conclusion

This systematic review and meta-analysis identify significant sociodemographic characteristics of infants and their mothers who may be at high risk not only for nonadherence with EHDI 1-3-6 guidelines but for complete LTF after NBHS. These factors can be used to identify infants who may be at high risk for LTF and therefore provide increased counseling or services to help combat that risk. Without appropriate follow-up, these infants will progress to school age with “unknown” reasons for NBHS failure, potentially causing detrimental learning and developmental delays. Focused efforts should be made by medical providers and policy-makers to address these factors to ensure appropriate newborn hearing care and interventions are achieved.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Appendix 1

Database Search Strategies

PubMed search strategy:

((“newborn hearing screen*“[tw] OR “neonatal hearing screen*”[tiab] OR “infant* hearing screen*”[tiab] OR NBHS[tiab] OR “early hearing detection and intervention”[tw] OR EHDI[tiab]) AND (“follow up”[tiab] OR “follow up rate”[tiab] OR “loss to follow

up”[tiab] OR “lost to follow up”[tiab] OR “delay* identification”[tiab])) OR ((“Hearing Tests”[Mesh] OR “hearing disorders”[MeSH]) AND (“Neonatal Screening”[Mesh] AND “Follow-Up Studies”[Mesh])) OR ((“newborn hearing screen*“[tw] OR “neonatal hearing screen*”[tiab] OR NBHS[tiab] OR “early hearing detection and intervention”[tw] OR EHDI[tiab]) OR (“Hearing Tests”[Mesh] AND “Neonatal Screening”[Mesh]) AND (“Health Care Disparities”[Mesh] OR “Health Status Disparities”[Mesh] OR disparit*[tw] OR inequalit*[tw] OR inequit*[tw] OR “culturally competent care”[Mesh] OR “culturally competent”[tiab] OR “Ethnic Groups”[Mesh] OR ethnic*[tiab] OR “Continental Population Groups”[Mesh] OR “Race Factors”[Mesh] OR race[tiab] OR racial[tiab] OR “Racism”[Mesh] OR “African Continental Ancestry Group”[Mesh] OR “African Americans”[Mesh] OR “African Americans”[tiab] OR Africa*[tiab] OR Asian*[tiab] OR immigrant*[ti] OR “Indians, north American”[Majr] OR lndia*[tiab] OR Japanese[tiab] OR Caucasian*[tiab] OR Hispanic*[tiab] OR Latin*[tiab] OR Mexican[tiab] OR “Minority Groups”[Mesh] OR “Socioeconomic Factors”[Mesh] OR “social determinants of health”[Majr] OR socioeconomic[tiab] OR sociodemographic[tiab] OR “Poverty”[Mesh] OR “Poverty Areas”[Mesh] OR poverty[tiab] OR “Health Services Accessibility”[Mesh] OR “Rural Population”[Majr] OR “Rural Health Services”[Majr] OR “Rural Health”[Majr] OR rural*[tiab] OR geograph*[tiab] OR proximity[tiab] OR “Vulnerable Populations”[Mesh] OR “Medically Underserved Area”[Mesh] OR underserved[tiab] OR “Insurance, Health”[Mesh] OR insurance[tiab] OR insured[tiab] OR “Medically Uninsured”[Mesh] OR uninsured[tiab] OR underinsured[tiab] OR “Medical Assistance”[Mesh] OR “Medicaid”[Mesh] OR Medicaid[tiab] OR “Medicare”[Mesh] OR medicare[tiab] OR “Communication Barriers”[Majr] OR “Translating”[Majr] OR language[ti] OR non-English[ti] OR “speaking”[ti]))

Scopus (Elsevier) search strategy:

((TITLE(“newborn hearing screen*” OR “neonatal hearing screen*” OR “infant* hearing screen*”))OR (TITLE-ABS-KEY(“newborn hearing screen*” OR “neonatal hearing screen*” OR “infant hearing screen*” OR NBHS OR “early hearing detection and intervention” OR EHDI OR “neonatal hearing screen*” OR “infant* hearing screen*” OR “neonatal hearing screen*”)))AND ((TITLE-ABS-KEY(“follow up” OR “follow up rate” OR “loss to follow up” OR “delay* identification”))OR (TITLE(“newborn hearing screen” OR “neonatal hearing screen” OR “infant hearing screen”))OR (TITLE-ABS-KEY(“newborn hearing screen*“ OR “neonatal hearing screen*“ OR “infant* hearing screen*“ OR NBHS OR “early hearing detection and intervention” OR EHDI OR “neonatal hearing screen” OR “infant hearing screen” OR “neonatal hearing screen*”))) AND (TITLE-ABS-KEY(“health care disparit*” OR “health status disparit*” OR disparities OR inequal* OR inequit* OR “culturally competent care” OR “cultural* competen*” OR “ethnic groups” OR ethnic OR “race factors” OR racial OR racism OR immigrant OR minority OR poverty OR socioeconomic OR sociodemographic OR “social determinants of health” OR “health services accessibility” OR rural OR “rural population” OR “rural health services” OR geograph* OR proximity OR “vulnerable populations” OR “medically underserved area” OR underserved OR insurance OR insured OR “medically uninsured” OR uninsured OR underinsured OR “medical assistance” OR Medicaid OR medicare

OR “communication barriers” OR translat*) OR (TITLE(non-English OR speaking OR language)))

CINAHL search strategy:

((newborn hearing screen* OR neonatal hearing screen* OR infant* hearing screen* OR NBHS OR “early hearing detection and intervention” OR EHDI) AND (follow up OR follow up rate OR loss to follow up OR lost to follow up OR delay* identification)) OR ((MH”Hearing Screening+” OR MH”Hearing Disorders+”) AND (MH”Neonatal Assessment+” OR MH”Infant, Newborn”) AND (MH”After Care+”)) OR (((newborn hearing screen* OR neonatal hearing screen* OR infant* hearing screen* OR NBHS OR “early hearing detection and intervention” OR EHDI) OR (MH”Neonatal Assessment” OR OR MH”Hearing Screening”)) AND (MH”Health Care Disparities” OR MH”Health Status Disparities” OR disparit* OR inequalit* OR inequit* OR ethnic* OR MH”Cultural Competence+” OR MH”Transcultural Care+” OR MH”Ethnic Groups” OR MH”Race Factors” OR race OR racial OR MH”Racism+” OR MH”Cultural Bias” OR MH“Black Persons+” OR “African Americans” OR Africa* OR Asian* OR immigrant* OR MH”Native Americans” OR India* OR Japanese OR Caucasian* OR Hispanic* OR Latin* OR Mexican OR MH”Minority Groups” OR MH”Socioeconomic Factors” OR MH”Social Determinants of Health” OR socioeconomic OR sociodemographic OR MH”Poverty” OR MH”Poverty Areas” OR poverty OR MH”Health Services Accessibility” OR MH”Rural Population+” OR MH”Rural Health Services” OR MH”Rural Health Centers” OR rural* OR geograph* OR proximity OR MH”Special Populations” OR MH”Medically Underserved Area” OR MH”Medically Underserved” OR underserved OR MH”Insurance, Health” OR insurance OR insured OR MH”Medically Uninsured” OR uninsured OR underinsured OR MH”Medicaid” OR Medicaid OR MH”Medicare” OR medicare OR MH”Communication Barriers” OR “Interpreter Services” OR language OR non-English OR “speaking”))

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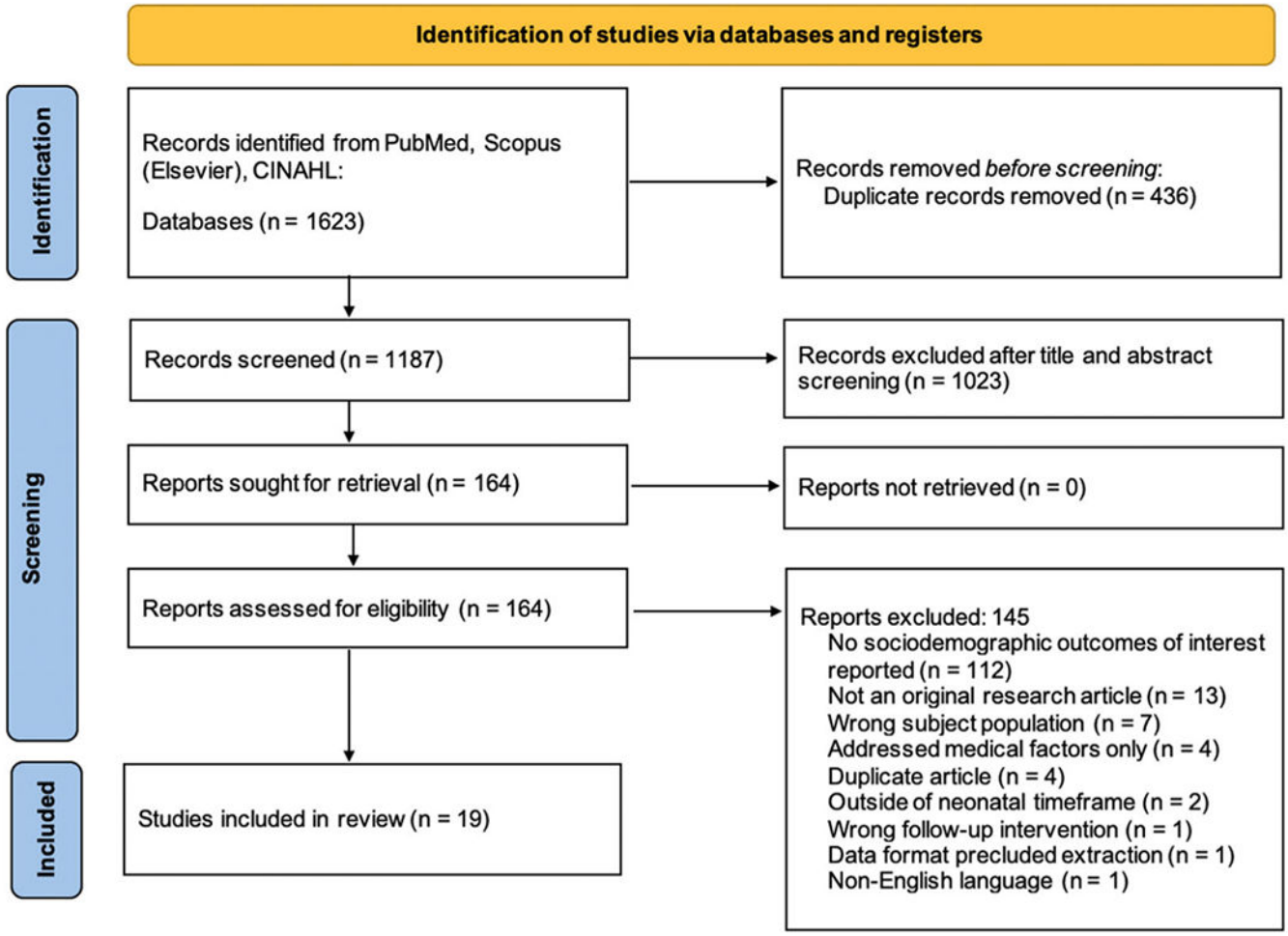

Figure 1.
Preferred Reporting Items for Systematic Reviews and Meta-analyses flow diagram of study selection.

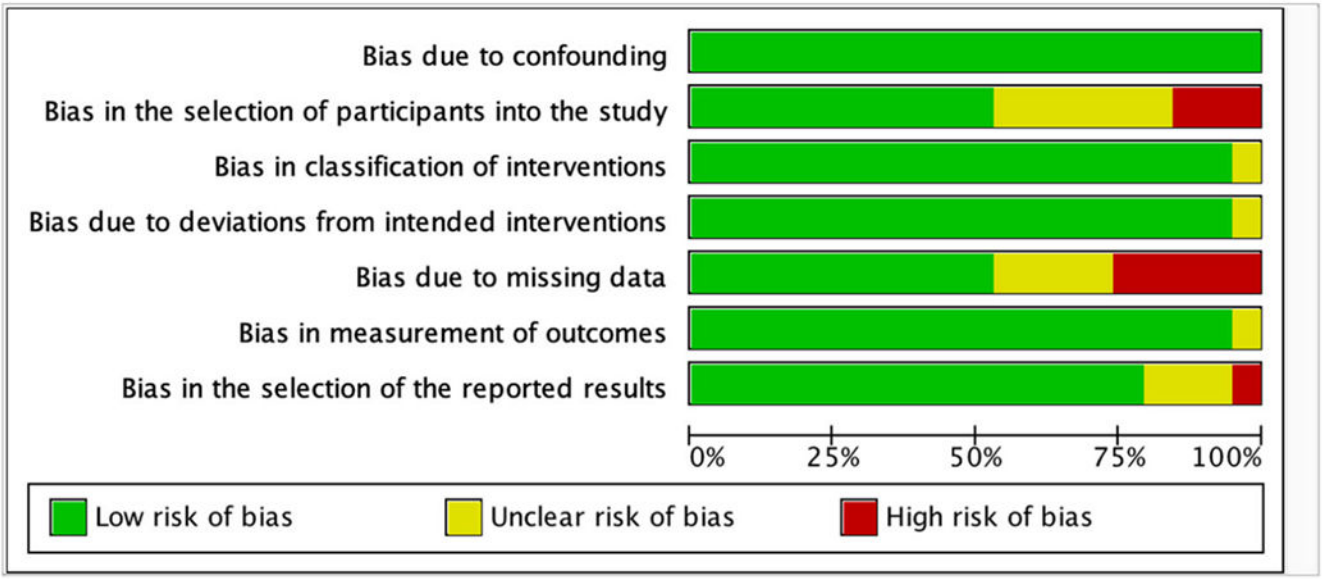

Figure 2.
Distribution of bias risk for multiple domains.

Table 1.

Descriptive Features of Studies Included in the Meta-analysis

References	Origin of population	OLE	Study design	Total number of patients (n)	Number of patients failed to follow-up (n)	Conclusions
Cavalcanti et al.[24]	Brazil	3b	Cross-sectional	577	224 (38.8%)	Low income, low education, multiparity, and lack of prenatal care are associated with NBHS nonadherence
Cheung et al.[32]	United States	3b	Case-control	443	217 (49.0%)	Maternal age, gravidity, and smoking status are associated with LTF in extended-stay NICU newborns
Christensen et al.[30]	United States	3b	Case-control	8124	1462 (16.0%)	Low Apgar scores and birth weight are associated with higher risk of hearing loss
Cunningham et al.[26]	United States	3b	Case-control	13,904	2482 (17.9%)	Low income, rural, and minority infants are at risk of LTF after referred NBHS
Deng et al.[39]	United States	3b	Cross-sectional	8829	2929 (33.2%)	Minority racial groups, low maternal education, and NICU stay are associated with delay in diagnostic testing for hearing loss
Folsom et al.[27]	United States	2b	Cohort	4911	2111 (43.0%)	Low socioeconomic status and increased social risk are strong predictors of poor follow-up compliance
Gaffney et al.[40]	United States	3b	Case-control	116,513	57946 (49.7%)	Health information technologies should be adopted to increased follow-up rate
Griz et al.[29]	Brazil	2b	Cohort	1035	886 (85.6%)	Low maternal education, low income, and rural location contribute to low compliance in outpatient follow-up
Kanji and Khoza-Shangase[25]	South Africa	2b	Cohort	325	109 (33.5%)	LTF is contextual, and risk-based surveillance programs are needed
Liu et al.[33]	United States	3b	Case-control	1492	157 (10.5%)	Limited resources should be directed to those infants at highest risk of LTF
Olusanya[22]	Nigeria	3b	Cross-sectional	551	56 (10.2%)	Maternal and infant sociodemographic and medical factors are not related to postdischarge LTF
Olusanya and Akinyemi[23]	Nigeria	3b	Cross-sectional	285	148 (51.9%)	Delivery in a hospital (vs outside a hospital) is associated with LTF for second-stage screening
Prince et al.[21]	United States	3b	Case-control	1013	176 (17.4%)	LTF risk may be related to cultural differences
Razak et al.[31]	United States	3b	Case-control	197	34 (17.3%)	LTF is higher among mothers who smoke, are multiparous and have infants at high risk of hearing loss
Sapp et al.[41]	United States	3b	Case-control	671	249 (37.1%)	Rural follow-up rates improved with diagnostic testing by educational audiologists
Vohr et al.[42]	United States	3b	Case-control	2761	294 (7.4%)	Medicaid, no insurance, NICU status, and out-of-state residence are associated with LTF
Wenjin et al.[43]	China	2b	Cohort	3425	1013 (29.6%)	Rural populations have a higher LTF rate than urban populations
Zeitlin et al.[44]	United States	2b	Cohort	41	11 (26.8%)	Race and access to health care professionals are predictive of LTF
Zeitlin et al.[45]	United States	3b	Cross-sectional	4141	434 (10.5%)	Race, maternal age, and parity are predictive of LTF

Abbreviations: LTF, loss to follow-up; NBHS, newborn hearing screening; NICU, neonatal intensive care unit; OLE, Oxford Level of Evidence.

Table 2.

Summary Odds Ratios for Each Sociodemographic Factor Examined

Sociodemographic factor	Odds ratio	95% CI	*p* value
Low maternal education	1.79	1.56-2.04	<.001[a]
Current or former maternal smoking status	1.75	1.38-2.23	<001[a]
Public/state versus private insurance	1.75	1.41-2.46	<.001[a]
Uninsured/self-pay versus private insurance	1.72	1.19-4.15	.008[a]
Low infant birth weight	1.60	1.16-2.21	<.001[a]
Young maternal age	1.52	1.45-1.59	<.001[a]
Rural location	1.47	1.12-1.91	.005[a]
Unmarried maternal status	1.46	1.14-1.87	.003[a]
Racial minority	1.40	1.21-1.62	<.001[a]
Primiparous maternal status	0.70	0.23-2.14	.53

Abbreviation: CI, confidence interval.

[a]Significant *p* value.