

## Early Detection of Congenital Hearing Loss in Infants: A Systematic Review Protocol to Identify the Most Effective and Efficient Screening Methods for Use in Low-Resource Settings

INPLASY202560019

doi: 10.37766/inplasy2025.6.0019

Received: 4 June 2025

Published: 4 June 2025

Mardiyanti, Nurhaeni, N; Wanda, D; Agustini, N; Huda, MH; Pradana, AA.

### Corresponding author:

Mardiyanti mardiyanti

mardiyanti.mardiyanti81@gmail.com

### Author Affiliation:

University of Indonesia.

### ADMINISTRATIVE INFORMATION

**Support** - This study receives no specific financial support from public, commercial, or not-for-profit funding agencies. The review will be conducted as part of doctoral academic requirements in the Doctoral Program in Nursing Science, Faculty of Nursing, Universitas Indonesia. All expenses related to literature access, data management, and dissemination will be self-funded by the principal investigator.

**Review Stage at time of this submission** - Preliminary searches.

**Conflicts of interest** - The authors declare no conflicts of interest related to the conduct, authorship, or publication of this protocol. This review is conducted independently for academic purposes as part of doctoral training in nursing science.

**INPLASY registration number:** INPLASY202560019

**Amendments** - This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 4 June 2025 and was last updated on 4 June 2025.

### INTRODUCTION

**R** **Review question / Objective** P (Population):  
Infants or newborns

I (Intervention): Hearing screening implemented by nurses or community health workers

C (Comparison): Standard screening techniques (e.g., Otoacoustic Emissions [OAE], Auditory Brainstem Response [ABR])

O (Outcomes): Screening effectiveness, sensitivity, specificity, cost-effectiveness, feasibility, acceptability, referral rate, and coverage.

**Rationale** Hearing loss is one of the most common congenital conditions affecting newborns

worldwide, with significant long-term impacts on speech, language, cognitive, and socio-emotional development if undetected and untreated. Early detection and intervention within the first six months of life—ideally through Universal Newborn Hearing Screening (UNHS)—have been shown to significantly improve developmental outcomes.

While UNHS programs have been successfully implemented in many high-income countries using otoacoustic emissions (OAE) and auditory brainstem response (ABR) technologies, these tools remain largely inaccessible in many low- and middle-income countries (LMICs), including Indonesia, due to high costs, limited equipment, and lack of trained audiologists. Consequently, delayed diagnosis of hearing loss is common, particularly in resource-constrained settings, which

limits timely intervention and increases the burden on families and healthcare systems.

In recent years, alternative screening approaches have emerged, including the use of parental-report questionnaires, low-cost mechanical devices, and innovations involving artificial intelligence (AI) and smartphone-based technology. These new tools offer potential for wider implementation by non-specialist health workers, such as nurses or community health staff, but their effectiveness, diagnostic accuracy, feasibility, and cost-efficiency remain inconsistently reported across studies.

Therefore, a systematic review is urgently needed to synthesize the evidence on the comparative effectiveness and feasibility of both conventional and emerging screening methods for early detection of infant hearing loss, especially in LMICs. This review will support evidence-based decision-making for selecting appropriate tools and strategies that can be scaled for national programs in Indonesia and similar settings.

**Condition being studied** Congenital hearing loss is a common neurodevelopmental condition affecting approximately 1–3 per 1,000 live births globally, with higher prevalence in low- and middle-income countries (LMICs). It can be caused by genetic factors, infections during pregnancy (e.g., rubella, cytomegalovirus), perinatal complications such as asphyxia or hyperbilirubinemia, and postnatal factors including ototoxic medications. Hearing loss in infancy, if not detected and managed early, can severely delay speech and language development, impact cognitive and emotional growth, and reduce educational and social outcomes later in life.

Early detection through newborn hearing screening—ideally within the first month of life—allows timely intervention such as hearing amplification, language therapy, and parent counseling. Standard screening tools include Otoacoustic Emissions (OAE) and Auditory Brainstem Response (ABR), which are effective but often expensive and dependent on specialist personnel. In many LMICs, including Indonesia, these tools are underutilized due to limited resources.

## METHODS

**Search strategy** comprehensive electronic search was conducted across six major databases: PubMed, IEEE Xplore, ProQuest, ScienceDirect, Scopus and ClinicalKey.

The search strategy was structured using the PICOS framework. The main keywords and MeSH terms included:

Population terms: “newborn”, “infant”, “neonatal screening”, “hearing loss”

Intervention terms: “hearing screening”, “otoacoustic emissions (OAE)”, “auditory brainstem response (ABR)”, “artificial intelligence”, “machine learning”, “parental questionnaire”, “behavioral observation audiometry”

Comparison terms: “standard screening”, “conventional ABR”, “OAE”

Outcome terms: “accuracy”, “sensitivity”, “specificity”, “cost-effectiveness”, “feasibility”, “referral rates”

Search limits applied:

Publication years: 2000–2025

Language: English

Article type: Peer-reviewed journals, full-text articles

Population: Infants aged 0–12 months

Reference management and screening were performed using citation software (EndNote), with selection conducted in two phases: title/abstract screening followed by full-text review.

**Participant or population** This review will include studies involving infants or newborns aged 0 to 12 months, regardless of gender, who undergo hearing screening in clinical, community, or primary care settings, particularly in low- and middle-income countries (LMICs) or regions with limited access to universal newborn hearing screening (UNHS).

The target population includes:

Healthy term infants

Preterm infants

Infants at high risk for hearing loss (e.g., history of neonatal intensive care, hyperbilirubinemia, congenital infections, craniofacial anomalies, family history of hearing loss).

**Intervention** This review will evaluate various hearing screening methods and technologies used to detect congenital hearing loss in infants, particularly those that can be implemented in low-resource settings by nurses, midwives, or non-specialist health workers. The interventions include both conventional and innovative screening approaches, such as:

Objective screening tools, Subjective and Technology-assisted innovations.

**Comparator** The comparators in this review will include standard or conventional hearing screening methods that are widely used and recognized as gold standard practices, particularly in well-resourced healthcare settings. These include:

Otoacoustic Emissions (OAE): either Transient Evoked OAE (TEOAE) or Distortion Product OAE (DPOAE)

Auditory Brainstem Response (ABR): either Automated ABR (AABR) or Clinical ABR performed by audiologists.

These standard methods will be compared to alternative interventions evaluated in this review, such as simplified tools, AI-based screening, or community-based programs. The comparison will focus on evaluating:

Diagnostic accuracy (sensitivity, specificity)  
Feasibility for nurse-led or community-based use  
Cost-effectiveness  
Referral and follow-up rates

The aim is to determine whether alternative tools can match or outperform conventional methods in low-resource settings.

**Study designs to be included** Randomized Controlled Trials (RCTs); non-randomized controlled trials; Quasi-experimental studies; Diagnostic accuracy studies; Prospective and retrospective cohort studies; Cross-sectional studies.

**Eligibility criteria** Inclusion Criteria:

Studies published in peer-reviewed journals from 2000 to 2025

Articles available in full-text and preferred in English

Studies that involve infants aged 0–12 months, regardless of risk status

Studies that report on screening accuracy (e.g., sensitivity, specificity, predictive values), feasibility, acceptability, or cost-effectiveness

Studies where screening is conducted by non-specialist personnel, including nurses, midwives, or trained community health workers.

**Information sources** This review will draw on multiple sources to ensure a comprehensive retrieval of relevant studies. The primary electronic databases to be searched include PubMed/MEDLINE, IEEE Xplore, ProQuest, ScienceDirect, Scopus, ClinicalKey.

In addition to electronic databases, the review will include:

Hand-searching reference lists of included studies and relevant reviews

Contacting study authors (if necessary) to obtain missing data or clarify study details

Search of trial registries (e.g., ClinicalTrials.gov, WHO ICTRP) for ongoing or unpublished studies  
Grey literature from relevant organizations such as the World Health Organization (WHO), especially for implementation guidelines and technical reports on newborn hearing screening in LMICs.

**Main outcome(s)** The primary outcomes of this review are focused on evaluating the performance and implementation feasibility of hearing screening methods for infants in low-resource settings. The main outcomes include:

Diagnostic Accuracy  
Sensitivity and specificity of each screening method in detecting congenital hearing loss in infants  
Positive predictive value (PPV) and negative predictive value (NPV)  
False positive and false negative rates  
Accuracy will be measured against a gold standard reference (e.g., ABR or diagnostic audiology evaluation)  
Feasibility of Implementation  
Usability by nurses or non-specialist health workers  
Screening coverage (percentage of eligible infants screened)  
Referral rate and follow-up completion  
Screening duration per infant or per session  
Training requirements and ease of use  
Cost-Effectiveness  
Acceptability.

**Additional outcome(s)** Outcomes will be extracted as reported by each study, generally at the point of screening and/or during the follow-up diagnostic phase, typically within the first 6 months of life.

Effect measures:

Diagnostic metrics (e.g., sensitivity, specificity) will be reported as proportions with 95% confidence intervals

Cost-effectiveness will be reported in local currency or standardized international units

Feasibility and acceptability will be summarized narratively and, where applicable, using frequency-based descriptive statistics.

**Data management** All retrieved citations will be imported into a reference management software (e.g. EndNote) for de-duplication and organization. Screening results (titles, abstracts, full texts) and data extraction will be managed using a standardized Excel form developed based on the review objectives and PICOS criteria. Discrepancies between reviewers will be resolved by consensus or a third reviewer.

**Quality assessment / Risk of bias analysis** The quality of included studies will be assessed using appropriate critical appraisal tools depending on the study design:

QUADAS-2 for diagnostic accuracy studies

JBICritical Appraisal Tools for observational and quasi-experimental studies

Cochrane Risk of Bias tool (RoB 2.0) for randomized trials,

Assessment will be performed independently by four reviewers. Results will inform interpretation but not be used as exclusion criteria.

**Strategy of data synthesis** A narrative synthesis will be performed to compare and summarize the effectiveness, accuracy, feasibility, and cost-efficiency of each screening method. Where sufficient homogeneity exists in study design and outcome measures, a meta-analysis of diagnostic accuracy (sensitivity, specificity) may be conducted using random-effects models.

Heterogeneity will be explored through subgroup analysis based on:

Type of screening method (OAE, ABR, AI-based tools, etc.)

Screening provider (nurse vs. audiologist)

Setting (hospital vs. community).

**Subgroup analysis** Where data are available, subgroup analyses will be performed to explore potential sources of heterogeneity and contextual differences in screening performance. The planned subgroups include:

Type of Screening Method, Type of Personnel Delivering the Screening, Setting (hospital or community).

**Sensitivity analysis** Sensitivity analysis will be conducted to test the robustness of the review findings by examining the impact of:

Study quality or risk of bias

Excluding studies rated as high risk of bias (based on QUADAS-2, JBI, or RoB 2.0 tools)

Study design

Comparing results from randomized controlled trials (RCTs) versus observational studies

Publication year

Screening personnel

Excluding studies conducted solely by specialists to assess how results hold in nurse- or lay provider-led settings.

**Language restriction** No language restriction.

**Country(ies) involved** Indonesia.

**Other relevant information** This systematic review protocol is part of a doctoral research project under the Doctoral Program in Nursing Science at the

Faculty of Nursing, Universitas Indonesia.

This protocol follows the PRISMA 2020 guidelines and, where applicable, the PRISMA-DTA for reviews of diagnostic accuracy studies.

**Keywords** newborn infant screening; hearing loss; sensitivity; specificity; accuracy.

**Dissemination plans** The findings of this systematic review will be disseminated through multiple academic and professional channels to maximize its impact, particularly in advancing evidence-based newborn hearing screening in low-resource settings. The dissemination plan includes: Publication in a peer-reviewed journal (International journal of nursing studies; BMC pediatrics, Belitung Nursing Journal), Integration into doctoral dissertation.

The review forms part of the candidate's PhD dissertation, which focuses on developing a model of early hearing detection led by nurses in Indonesia.

#### **Contributions of each author**

Author 1 - Mardiyanti Mardiyanti - Conceived the review topic, developed the research question and objectives, formulated the search strategy and eligibility criteria, and drafted the protocol manuscript. Will lead the literature screening, data extraction, quality assessment, and synthesis.

Email: mardiyanti.mardiyanti81@gmail.com

Author 2 - nani indonesia - Provided critical supervision in the design and methodological rigor of the review protocol. Contributed to refining the inclusion criteria, advised on risk of bias assessment tools, and reviewed all protocol drafts for scientific and academic accuracy.

Email: nani-n@ui.ac.id

Author 3 - Dessie wanda - Provided critical supervision in the design and methodological rigor of the review protocol. Contributed to refining the inclusion criteria, advised on risk of bias assessment tools, and reviewed all protocol drafts for scientific and academic accuracy.

Email: dessie@ui.ac.id

Author 4 - Nur Agustini - Contributed to Literature screening, data extraction, quality assessment, and synthesis.

Email: nur.agstn@gmail.com

Author 5 - Mega Hasanul Huda - Contributed to Literature screening, data extraction, quality assessment, and synthesis.

Email: megahasanulhuda@gmail.com

Author 6 - Anung Ahadi Pradana - Contributed to Literature screening, data extraction, quality assessment, and synthesis.

Email: ahadianung@gmail.com