INPLASY PROTOCOL

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Conflicts of interest:
None declared.

Permanent deafness in children not identified via universal newborn hearing screening

Harris, SA[1]; Beckman, A[2]; Munro, KJ[3].

Review question / Objective: **1. Investigate how children with permanent hearing loss were identified if it was not via universal newborn hearing screening (UNHS). 2. Identify the age of identification, aetiology and developmental outcomes for these children.**

Background: **Hearing loss is one of the most common disabilities affecting the global population. According to the World Health Organisation (WHO), approximately 466 million people suffer from disabling hearing loss, which equates to over 5% of people worldwide (WHO, 2021). In newborns, significant permanent hearing loss (e.g ≥40 dB HL in the better hearing ear) occurs in 1-3 of every 1000 births, far exceeding the incidence of other common conditions newborns are routinely screened for (Butcher et al 2019). Universal Newborn Hearing Screening (UNHS) has contributed significantly to the early diagnosis of hearing loss in many children (Harrop-Griffiths, 2016). Findings from a 2012-2013 study showed that the age of hearing loss diagnosis is "now well within a baby's first six months of life" (Wood, Sutton and Davis, 2015).**

Despite the success of UNHS, it is acknowledged that not all hearing loss will be detected. Some children will acquire deafness later in life through trauma or illnesses such as bacterial meningitis. Other types of acquired deafness can include syndromic or genetic conditions associated with progressive hearing loss. Some children will be missed because screening sensitivity is reported to be 51% in detecting children with all degrees of permanent hearing loss (Watkin and Baldwin, 2011). Studies that have investigated the percentage of deaf children not identified via UNHS vary significantly in their findings between 1% (Connelly et al, 2005), to 51% (Watkin and Baldwin, 2011).

INPLASY registration number: This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 15 May 2023 and was last updated on 15 May 2023 (registration number INPLASY202350064).

INTRODUCTION

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Despite the success of UNHS, it is acknowledged that not all hearing loss will be detected. Some children will acquire deafness later in life through trauma or illnesses such as bacterial meningitis. Other types of acquired deafness can include syndromic or genetic conditions associated with progressive hearing loss. Some children will be missed because screening sensitivity is reported to be 51% in detecting children with all degrees of permanent hearing loss (Watkin and Baldwin, 2011). Studies that have investigated the percentage of deaf children not identified via UNHS vary significantly in their findings between 1% (Connelly et al, 2005), to 51% (Watkin and Baldwin, 2011).

Rationale: The purpose of this scoping review is to investigate studies which have investigated deafness in children screened but not identified via UNHS . In clinic we regularly come across children whose aetiology is such that we would have expected them to be identified at birth and yet they were not. We have decided upon a scoping review as this will help to determine the scope or coverage of the literature related to the research question (Munn et al, 2018). It will also assist in collating and mapping the available evidence and in identifying key characteristics related to the research question. In addition, it will aim to form a precursor to a later systematic review.

METHODS

Strategy of data synthesis: Data from the identified studies will be summarised in a spreadsheet which will include publication details (title, author(s), publication year, journal, contact detail(s), study design, participant demographics, funding and any other relevant data). Ideally meta-analysis will be possible, however, it is unlikely sufficient number of studies exist to explore further research topics.

Eligibility criteria:
PICO inclusion:
Participants;
Diagnosed with permanent hearing loss
All sexes
Children who had newborn hearing screening
Interventions;
N/A
Outcomes;
Outcomes of UNHS including age of detection, intervention, average thresholds, aetiology and methodology
Developmental outcomes including language, speech perception, literacy, social development, interpersonal skills and quality of life
PICO exclusion:
Participants;
No UNHS
Normal hearing or non-organic hearing loss
Temporary conductive hearing loss. Downs syndrome, cleft palate or other high risk of persistent otitis media with effusion covered by separate NICE guidance
Auditory processing disorder
Studies of children over 16 years in age . If a study includes both children and adults it will be excluded unless the findings were reported separately
Hearing loss identified using an unrecognised testing methodology(e.g. Visual Reinforcement Audiometry or Pure Tone Audiometry) (BSA recommended

procedures))
Interventions;
Deaf children identified via UNHS
Children diagnosed with permanent hearing loss who were identified using protocols other than those acknowledged by BSA.
Outcomes;
Not reporting key audiological outcomes
Not reporting key developmental outcomes
Did not specify the total number of children confirmed to have hearing loss to capture the percentage of those identified via UNHS. Should the studies offer a reason for the failure to diagnose the condition these studies will be included.

Source of evidence screening and selection: Identification of studies relevant to the research question will be achieved by searching electronic databases of published literature which will include EMBASE, Cochrane Library, PubMed, SCOUPS, PSYCINFO and Google Scholar. To identify ongoing or concluded studies grey literature searching will be undertaken using ClinicalTrails.gov, International Clinical Trials Registry Platform (ICTRP WHO), search portal, targeted sites from Canadian Agency for Drugs and Technologies in Health (CADTH), Grey Matters and the preprint server for health sciences (Medrxiv). Google Scholar will be used to identify further grey literature . Should a high number of studies be identified via Google Scholar only the first 200 hits sorted by relevance will be screened for inclusion. Studies will be sorted by relevance to the research objectives.
Studies which report on developmental issue s and quality of life outcomes of deaf children not identified via UNHS will prove particularly useful in addressing the two objectives as this information may help inform best clinical practice.
The study flow will detail the total number of accepted records identified, included and excluded and the reasons for their exclusions, Reference lists of the included studies and their citations will be tracked . Reference lists of relevant published clinical guidelines, including the British Society of Audiology, British Academy of Audiology and Public Health England will be reviewed for additional studies to include. The reference lists of all included research will be catalogued by authors and title and screened by both the primary author and offered to the secondary authors for their opinion on inclusion . Grey literature including magazine articles and conference presentations will be included. Audiology magazines (E.g. The BAA Magazine and BSA Audacity) will be searched using online archives of professional body websites including British Academy of Audiology and British Society of Audiology.

Data management: Records retrieved by the search will be exported to reference management software for automated removal of duplicates. Further manual assessment will then be performed to identify and remove any remaining duplicates.
The title and abstract of all identified studies will be screened by SH to determine eligibility for inclusion. Potentially eligible studies will be assessed against the inclusion and exclusion criteria by SH. KM will audit 10% of studies independently from SH. Where there is consensus with both independent authors, studies will be included but where there is discrepancy a discussion will be held to resolve the issues. Any disagreement that cannot be resolved by discussion will be considered by an independent researcher . AB will review 10% of accepted studies in full (denoted by availability) to independently audit practice . Any disagreement that cannot be resolved by discussion will be considered by an independent researcher .
Data from the identified studies will be summarised in a spreadsheet which will include publication details (title, author(s), publication year, journal, contact detail(s), study design, participant demographics, funding and any other relevant data). Ideally meta-analysis will be possible, however, it is unlikely sufficient number of studies exist to explore further research topics.

Reporting results / Analysis of the evidence: As recommended by PRISMA guidelines (Page et al, 2021) a study flow diagram will illustrate the screening pathway and inclusion and exclusion process highlighting the total number of studies excluded at each stage.

Presentation of the results: Data will be analysed and presented in a way suitable to the information obtained. It is envisaged that one the scope has been completed a service evaluation template will be formed to analyse local provision data.

Language restriction: Yes only abstracts transcribed into English will be included.

Country(ies) involved: United Kingdom.

Keywords: Hearing loss; hearing disorders; hearing problems; hearing difficult; hearing impairment; audiology; newborn hearing; otoacoustic emission; auditory brainstem respons.

Dissemination plans: The information will be shared with colleagues across regional and national paediatric audiology specialist interest groups, namely the British Society of Audiology and Southwest groups. Further the information will be discussed at the regular Children's Hearing Services Working Group meeting which includes stakeholders from patients, families of children and early interventionists (e.g. speech therapists and teachers of the deaf) with a view to creating a new key performance indicator for quality monitoring.

Contributions of each author:

Author 1 - Stuart Harris - Author 1 will screen identified studies. Author 1 will analyse accepted studies. Author 1 will draft manuscript following data analysis.
Email: stuart.harris1980@nhs.net
Author 2 - Adam Beckman - Author 2 will review 10% of accepted papers to audit author 1's decision making process.
Author 2 will support author 1 and 3 in manuscript draft and editing.
Author 2 will facilitate any discrepancies between author 1 and 3 in the review process.
Author 3 - Kevin Munro - Author 3 will review 10% of identified papers to audit author 1's decision making process. Author 3 will support author 1 and 2 in manuscript draft and editing. Author 3 will facilitate any discrepancies between author 1 and 2 in the review process.
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