A scoping review of studies comparing outcomes for children with severe hearing loss using hearing aids to children with cochlear implants.

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ABSTRACT

Objectives

Clinical practice regarding children’s candidature for cochlear implantation varies internationally, albeit with a recent global trend towards implanting children with more residual hearing than in the past. The provision of either hearing aids or cochlear implants can influence a wide range of children’s outcomes. However, guidance on eligibility and suitability for implantation is often based on a small number of studies and a limited range of speech perception measures. No recent reviews have catalogued what is known about comparative outcomes for children with severe hearing-loss using hearing aids to children using cochlear implants. This paper describes the findings of a scoping review that addressed the question ‘What research has been conducted comparing cochlear implant outcomes to outcomes in children using hearing aids with severe hearing-loss in the better-hearing ear?’

The first objective was to catalogue the characteristics of studies pertinent to these children’s candidature for cochlear implantation, to inform families, clinicians, researchers and policy-makers. The second objective was to identify gaps in the evidence base, to inform future research projects and identify opportunities for evidence synthesis.

Design

We included studies comparing separate groups of children using hearing aids to those using cochlear implants, and also repeated measures studies comparing outcomes of children with severe hearing loss before and after cochlear implantation. We included any outcomes that might feasibly be influenced by the provision of hearing aids or cochlear implants. We searched the electronic databases Medline, PubMed and CINAHL, for peer-reviewed journal articles with full-texts written in English, published from July 2007 to October 2019. The scoping methodology followed the approach recommended by the Joanna Briggs Institute regarding study selection, data extraction, and data presentation.
Twenty-one eligible studies were identified, conducted across eleven countries. The majority of children studied had either congenital or pre-lingual hearing loss, with typical cognitive function, experience of spoken language, and most implanted children used one implant. Speech and language development and speech perception were the most frequently assessed outcomes. However, some aspects of these outcomes were sparsely represented including voice, communication and pragmatic skills, and speech perception in complex background noise. Two studies compared literacy, two sound localization, one quality of life and one psychosocial outcomes. None compared educational attainment, listening fatigue, balance, tinnitus, or music perception.

Conclusions

This scoping review provides a summary of the literature regarding comparative outcomes of children with severe hearing-loss using acoustic hearing aids and children using cochlear implants. Notable gaps in knowledge that could be addressed in future research includes children’s quality of life, educational attainment, and complex listening and language outcomes, such as word and sentence understanding in background noise, spatial listening, communication and pragmatic skills. Clinician awareness of this sparse evidence base is important when making management decisions for children with more residual hearing than traditional implant candidates. This review also provides direction for researchers wishing to strengthen the evidence base upon which clinical decisions can be made.
INTRODUCTION

The clarity with which children hear affects how they perceive speech in quiet and noisy settings. Poor sound clarity can limit children’s ability to participate socially and achieve academically, which can lead to poorer quality of life and socio-emotional well-being (Roland et al. 2016). Importantly, being able to hear and accurately discriminate sounds is key to developing spoken language. Friedmann & Rusou (2015) concluded in a review of the literature that there is a critical period for language development within the first year of life. Therefore, when a child has a hearing loss, it is important to offer them technology to optimise sound quality as early in life as possible. As hearing loss worsens in severity from mild through to profound, there comes a cusp at which the sound quality achieved by amplifying sound with conventional acoustic hearing aids (HA) and presenting it to a damaged inner ear is likely to be worse than the clarity a child could experience by replacing the function of cochlear hair cells with electrical stimulation from a cochlear implant (CI). To maximise outcomes, is it necessary to know the cusp at which CIs are likely to outperform HAs for each child.

Clinical CI candidature decisions are made on a case-by-case basis by multidisciplinary teams, within the limits of their own healthcare and funding systems. Each candidate’s audiometric thresholds, speech perception, language development, support network, health, hearing history, prior device use, anatomy and additional needs are taken into account. Speech perception, language development, and additional needs can be difficult to assess in the very young, so audiometric thresholds are especially important in CI candidature decisions for children. However, there is significant variation in estimates of the audiometric cusp at which CIs are expected to outperform HAs for children, and international variation in how these estimates are translated into guidance for clinical practice (Schwartz et al. 2012; Vickers et al. 2016).

In the United States, children can be offered unilateral or bilateral CIs implanted simultaneously or sequentially. Eligibility is based on the better-hearing ear under the age of five years, with bilateral profound hearing loss required under the age of two years and bilateral severe-to-profound hearing loss between the ages of two to five years. From the age of five years, eligibility for unilateral implantation may be based on the worse-hearing ear, in
cases of poor speech discrimination (Varadajan et al. 2021). The American Speech Language Hearing Association (ASHA) defines severe hearing loss as a mean threshold of 71 to 90 dB HL, and profound loss as a mean threshold of ≥ 91 dB HL, averaged over an unspecified number and range of audiometric frequencies (Clark, 1981). In England and Wales, children are considered for either unilateral or simultaneous bilateral CIs if the better-hearing ear meets the National Institute for Health and Care Excellence (NICE) definition of severe-to-profound hearing loss, i.e. thresholds ≥ 80 dB HL at any two frequencies including 0.5, 1, 2, 3 or 4 kHz (NICE, 2019). NICE had concluded that sequential implantation is not a cost-effective use of healthcare resources. Simultaneous or sequential bilateral CIs are permitted in France, where children with moderate or worse hearing loss can be considered candidates on the basis of their worse-hearing ear, but having a mild loss or typical hearing in the better-hearing ear precludes implantation of the worse-hearing ear (Simon et al. 2019). In Belgium, the audiometric criteria for the ear to be implanted depends on the symmetry of the hearing-loss. Three or more thresholds including 0.5, 1, 2, and 4 kHz must equal or exceed 70 dB HL for bilateral losses, or ≥ 85 dB HL in asymmetric losses (Belgisch Staatsblad, 2019). These examples are not exhaustive, but serve to illustrate the variation that exists in how different healthcare systems have established clinical guidance based on the evidence. Comparative studies are important both to inform when a transition from bimodal listening to bilateral implantation is recommended, and also to determine when a child should transition from acoustic amplification alone to listening via either one or two cochlear implants. This review addresses the latter.

Audiometric recommendations proposed by research studies include average unaided thresholds of between 88 to 96 dB HL (Davidson 2006), 80 dB HL or worse (Lovett et al. 2015), and 65 dB HL or greater (Leigh et al. 2016). This lack of agreement in recommendations between studies was influenced, in part, by different choices the authors made regarding how much certainty of benefit was required to recommend CIs over HAs (see Table 2 of Lovett et al. (2015) and Appendix A of Leigh et al. (2016)). Another source of variability was the outcome measure used to derive these recommendations. Davidson (2006) reported that the cusp was dependent on the presentation level used when assessing word perception in quiet. Leigh et al. found different cusps depending on whether phoneme or sentence measures were used, deriving from the same dataset audiometric criteria of 75 dB HL based on sentence perception in quiet (Leigh et al. 2011) and 65 dB HL based on phoneme perception in quiet (Leigh et al. 2016). Lovett et al. (2015) also found a 10 dB
difference in the cusp depending on the type of background noise used during the same word perception test. If the audiometric cusp at which children with CIs out-perform children with HAs can vary so much for different measures of speech perception, it is plausible that estimates of the cusp might also vary between other outcomes, e.g. spatial hearing, quality of life, etc. If so, it is important that other outcomes, that are important to children and their families, are considered when developing candidature recommendations. Failure to do so risks CIs being recommended to improve one outcome, at the cost of creating poorer outcomes in another area of hearing and/or health that was not as well understood or characterized. Conversely, with-holding implantation for one outcome might disadvantage a child in relation to others.

CI clinics encounter children who differ greatly in terms of audiometric configuration, chronological age, device use, early auditory experience, cognitive function, other complex additional healthcare needs, and exposure to, and development of, spoken language. Aside from audiometric thresholds, it is possible that the cusp between HAs and CIs will be dependent on these other clinical and demographic characteristics. It is difficult for any individual study on CI candidature to make recommendations that are relevant to every possible clinical scenario. However, syntheses and summaries of all available evidence allow us to identify themes and gaps in the literature that provide a good basis upon which to develop general guidance on the candidature of children for CIs.

One seminal systematic review, of literature published up to July 2007, was published in 2009 (Bond et al. 2009a). The authors concluded that unilateral CIs were clinically effective and cost-effective for children with bilateral profound hearing loss. The research studies described in that review indicate that the audiometric cusp for candidacy may now lie somewhere within the range of 65 – 95 dB HL, i.e. severe hearing loss (Davidson, 2006, Leigh et al. 2011 and 2016; Lovett et al. 2015). Bond et al. (2009a) made no recommendations for implantation in children whose better-ear unaided thresholds averaged 70 to 95 dB HL, because of an absence of evidence at that time for what outcomes were likely to improve (Bond, et al., 2009b). The authors also noted the absence of data on quality of life or educational attainment and recommended that these outcomes should also be measured in future studies to improve the evidence upon which CI candidature guidance is based. They also recommended that studies should be carried out to establish the benefits of CIs for children with additional needs, and to determine the location of the audiometric cusp
beyond which CIs would be unlikely to provide clinically meaningful benefits and/or cease to be cost-effective compared to HAs.

Much research has been conducted on cochlear implantation since the latest publication date for studies reviewed by Bond et al (2009a) in July 2007, and both clinical practice and CI technology have evolved. Increasingly, CIs are fitted bilaterally rather than unilaterally, closer in line with HA practice, and age at intervention has tended to decrease, both associated with improved outcomes (Ramsden et al. 2012; Yoshinaga-Itano et al. 2018; Teagle et al. 2019). Manufacturers have also introduced new sound processing algorithms and microphone directionality options (e.g. Lorens et al. 2010; Spriet et al. 2007). Therefore, comparing the outcomes of children using HAs and CIs must be reviewed regularly, because changes in practice and technology might influence the cusp at which implantation should be considered.

De Kleijn et al. (2018) sought to summarise the evidence for audiometric CI criteria in light of these developments in technology and clinical practice. In line with the range of audiometric cusp estimates described by Davidson (2006), Leigh et al. (2011; 2016), and Lovett et al. (2015), de Kleijn et al. searched for the literature on HA users with severe hearing loss. The authors included 10 records comparing outcomes for children with severe hearing-loss in the better-hearing ear using HAs to children using CIs. While the review provides a valuable summary of studies that could be used to define audiometric criteria, the literature search was restricted to studies of speech production, speech perception, receptive language, and auditory performance only. There remains a need to catalogue how other outcomes vary between these groups, including quality of life and educational attainment, as noted by Bond et al. (2009b). Furthermore, provision of HAs or CIs to children with more residual hearing to lose than traditional CI candidates might also affect outcomes that may not be routinely measured in the clinic such as spatial hearing, listening effort and fatigue, psychosocial outcomes, vestibular function, tinnitus, and music perception (Dorman et al. 2016; Fiorillo et al. 2017; Ganek et al. 2020; Killan et al. 2018; Looi 2014; Winn 2007; Wong et al. 2017). While optimizing these outcomes might not be the primary goal when choosing a listening device, they are important outcomes to assess following the provision of listening devices as they can impact children’s quality of life, mental health, social and recreational participation, sleep, and educational attainment (Camarata et al. 2018; Fellinger et al. 2015, Inoue et al. 2013; Smith et al. 2019; Vecchiato et al. 2013).
In summary, the choice whether to offer CIs to a child can affect many aspects of their life. A large range of studies report outcomes for HA and CI users, however there are few comparative studies of children with severe hearing loss who use only acoustic HAs and children using at least one CI, and those available address a limited range of outcomes. There is no review of recent studies addressing a wider range of outcomes than those directly related to speech reception and speech and language development, and with detailed descriptions of the children studied. This gap in the literature has significant implications. It is difficult for clinicians to know to what extent the existing evidence is applicable to each child they consider for implantation. It is also difficult to predict how implantation might affect outcomes that are not included in the studies from which recommendations have been derived. The best methodology to address these problems is a scoping review (Arksey & O’Malley 2005), which is designed to clarify what is known and what is not known and identify areas for future research.

This paper describes the findings of a scoping review that addresses the question ‘What research has been conducted comparing outcomes in children using CIs to outcomes in children using HAs with severe hearing-loss in the better-hearing ear?’ For this review, we used a definition of ‘severe’ inclusive of the different definitions from the World Health Organisation (WHO), ASHA, and British Society of Audiology (BSA) (Clark 1981; WHO 1991; BSA 2018); that is, average unaided hearing thresholds in the better-hearing ear between 61 to 95 dB HL for all participants using HAs. Bond et al (2009a) also found no comparative studies of children using CIs compared to children using HAs with thresholds in this range. Therefore applying this definition of ‘severe’ removed bias in study selection for or against countries using different classification systems, captured all potentially relevant studies published since those included by Bond et al (2009a), and covered the range of criteria proposed by Davidson (2006), Leigh (2011 & 2016) and Lovett (2015).

Toward answering the scoping review question, we defined two objectives:

a. To catalogue the characteristics of studies pertinent to candidature of children with severe hearing-loss for cochlear implantation.
b. To identify gaps in the evidence base regarding comparative outcomes for children with severe hearing-loss using HAs and children using CIs, to inform future research projects and identify opportunities for evidence synthesis.

MATERIALS AND METHODS

This scoping review was designed, conducted, and presented in line with guidance from the Joanna Briggs Institute and the PRISMA extension for scoping reviews (Tricco et al. 2018)

Eligibility criteria

To be included in the review, records needed to contain data from either a group of children with severe hearing-loss who were HA users compared to a group of children using CIs, or data from a group of children with severe hearing-loss assessed before and after they received CIs. Outcomes of interest included all those that could feasibly be influenced by the provision of a CI or HA. Qualitative, quantitative, and mixed methods studies were all included. We aimed to ensure that our review complemented rather than duplicated Bond et al. (2009a). We therefore searched for studies published from July 2007 to the present, immediately following the search by Bond et al (2009a), but overlapping the period considered by de Kleijn et al. (2018) due to the more restricted range of outcomes they considered. Only peer-reviewed records were included. We included both open-access and non-open-access articles. Because of resource limitations, only records with full-texts written in English were included.

Participant inclusion and exclusion

All participants needed to be aged less than 18 years. We applied audiometric eligibility criteria for the children in our HA groups, for both repeated measures and between group comparison studies. We excluded records with only normally-hearing participants, participants who were all profoundly deaf or traditional CI candidates (e.g. described using terms such as “profoundly deaf”, “total deafness”, “severe-to-profound”), and those where all participants had normal or near-normal hearing in one ear; i.e. “single-sided deafness”. Our
protocol defined severe hearing-loss as pure-tone thresholds in the better-hearing ear, averaged across 0.5 to 4 kHz, of 61 to 95 dB HL. During full-text screening, studies were excluded if it was not possible to confirm that all HA users had unaided thresholds within our definition of severe hearing-loss. If it was not possible to determine this from the text, we contacted the corresponding authors and based our decisions upon the responses we received. In the absence of confirmation that any individual participants in the HA group met this criterion (i.e. hearing thresholds were not reported or could not be obtained directly from authors), we included studies where the reported participant characteristics for the average unaided thresholds of the better-hearing ear fell within the range described above. If only group characteristics were reported, we included studies where the group had a mean unaided threshold average within the defined range. If only qualitative descriptions of the degree of hearing loss were given with no supporting audiometric data, we included studies that reported children's hearing fell within the "severe" range. No audiometric inclusion criteria were applied to children in the CI groups of between group comparison studies.

Intervention inclusion and exclusion

Intervention inclusions for the HA group were that children wore at least one acoustic HA and no CI. Intervention inclusions for the CI group were that children used at least one multi-electrode, intra-cochlear hearing implant. The CI group could include children with unilateral CI alone, unilateral CI with a contralateral acoustic HA (bimodal aiding), unilateral or bilateral short arrays for electric-acoustic stimulation (EAS) or bilateral CI. Intervention exclusions for both HA and CI groups included any use of auditory brainstem implants, bone-conduction devices, and vibro-tactile aids.

Outcome inclusion and exclusion

We included any outcome that might plausibly be influenced by the provision of either CIs or HAs, such as listening, language, speech production, reading, music perception, balance, dizziness, tinnitus, educational measures, psychosocial, mental health and quality of life. We excluded studies that did not measure any of the outcomes listed above. Illustrative examples of outcomes not within scope included, but were not restricted to: surgical techniques, development of new tools (e.g. questionnaires), and audits of patient pathways.
Study design inclusion and exclusion

Study designs within scope included observational or interventional studies observing the outcomes of HAs versus CIs. This included peer-reviewed studies in scientific or medical journals reporting randomised controlled trials, quasi-randomised controlled trials, before and after studies, non-randomised controlled trials, cross-over studies, cohort studies, and case control studies. We excluded case studies and case series during title and abstract screening. However, studies that were passed to full text screening were retained if they included data from a sub-group with severe hearing-loss or individual data for participants who met our inclusion criteria. Study designs out of scope included reviews of any kind. We also excluded magazine articles, conference presentations, practice guidelines, expert opinions, book chapters, manufacturers’ articles, predictive modelling and simulation studies, editorials, letters to the editor, workshop summaries, and online training courses.

Information sources

We searched Medline (using OvidSP), the Cumulative Index to Nursing and Allied Health Literature (CINAHL) (using EBSCOhost) and PubMed. Search

We searched for records where titles, abstracts or keywords included terms for “child” AND “hearing aid” AND “cochlear implant”. Search strategies were developed through team discussion and included alternative phrasing for each term. An example search strategy (Medline) is shown in Table 1 (other search strategies are available as supplementary documents). The search results were exported into EndNote, and duplicates removed. The remaining records were exported into Excel. Initial searches were conducted in April 2019, and further update searches were conducted in October 2019 and September 2020. All records were assigned a study code at this point, to enable tracking them through the study selection process.
Table 1: Search terms

<table>
<thead>
<tr>
<th>Search #</th>
<th>Ovid Medline Search term</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>child*.ab,ti.</td>
</tr>
<tr>
<td>2</td>
<td>paediatric.ab,ti</td>
</tr>
<tr>
<td>3</td>
<td>pediatric.ab,ti</td>
</tr>
<tr>
<td>4</td>
<td>CHILD/</td>
</tr>
<tr>
<td>5</td>
<td>ADOLESCENT/</td>
</tr>
<tr>
<td>6</td>
<td>amplif*.ab,ti</td>
</tr>
<tr>
<td>7</td>
<td>&quot;hearing aid*”.ab,ti.</td>
</tr>
<tr>
<td>8</td>
<td>HEARING AIDS/</td>
</tr>
<tr>
<td>9</td>
<td>&quot;cochlea* implant*”.ab,ti.</td>
</tr>
<tr>
<td>10</td>
<td>&quot;cochlea* prosth*”.ab,ti.</td>
</tr>
<tr>
<td>11</td>
<td>COCHLEAR IMPLANTS/</td>
</tr>
<tr>
<td>12</td>
<td>COCHLEAR IMPLANTATION/</td>
</tr>
<tr>
<td>13</td>
<td>1 OR 2 OR 3 OR 4 OR 5</td>
</tr>
<tr>
<td>14</td>
<td>6 OR 7 OR 8</td>
</tr>
<tr>
<td>15</td>
<td>9 OR 10 OR 11 OR 12</td>
</tr>
<tr>
<td>16</td>
<td>13 AND 14 AND 15</td>
</tr>
</tbody>
</table>

Selection of sources of evidence

Study selection was based on the PICOS framework (Population, Intervention, Comparison, Outcome and Study type) and was piloted and refined by CFK and DJH. First, titles and abstracts were screened by CFK, DJH and RK such that each title/abstract was independently screened by two reviewers. Any discrepancies regarding inclusion or exclusion were resolved by discussion between reviewers. Where no consensus could be reached, the final decision regarding inclusion was made by the third reviewer, with the majority verdict being accepted. If there was insufficient information in the title and abstract to establish whether a study met our eligibility criteria, it was passed for full-text screening. The same PICOS framework was applied at the full-text screening stage.

We selected studies including: a) between groups studies, where a group of children with severe hearing-loss using only acoustic HA(s) were compared to another group of children using at least one CI, or HA users with a wider range of hearing-loss were compared to a group of CI users for the purpose of determining the cusp of candidature; and b) repeated measures longitudinal studies, where children with severe hearing-loss were assessed both...
before implantation using only acoustic HA(s) and again after implantation using at least one CI.

Data charting process
A data-charting tool was developed by CFK, DJH and PTK. Data from all eligible studies were charted by CFK, then verified by one of four co-reviewers, DJH, PTK, RHP or BA. Any disagreements were resolved through discussion between CFK and the co-reviewer. During this process, the tool and data extraction were updated in an iterative process.

Data items
To characterise the children, we extracted data on where they had been recruited from, chronological age, age at diagnosis of hearing loss, age at first HA fitting, and age at first cochlear implantation. We also extracted data on cognitive function, communication mode, and the presence or absence of additional difficulties.

To characterise the interventions, we extracted data on the number of children in the HA group using unilateral or bilateral HAs, and the number of children in the CI group(s) who used unilateral, bilateral, bimodal, or short-array implants for EAS in either ear. We extracted data on the outcome measures used in each comparison, and categorised them into the following broad categories: speech perception, speech and language, quality of life, psychosocial, sound localization, listening fatigue, balance, educational, tinnitus and music perception. Within the speech perception category we classified outcome measures into phoneme, word, sentence and supra-segmental speech perception, and also into categories of tests administered in quiet, or in background noise, extracting the type of noise and signal to noise ratio used. Within the speech and language category, we classified outcome measures into phonological skill, receptive language, expressive language, speech production, voice, communication and pragmatics, and generalised auditory performance. To characterise the studies, we extracted participants’ country of residence, publication year, title, objectives, and whether one group of children had been compared before and after cochlear implantation, or two or more separate groups of children had been compared. Figure 1 summarises the selection of studies for data extraction.
Figure 1: Flow diagram of record identification and selection.

We extracted data from the records into Excel, and created summary tables and figures. These included participant characteristics, the interventions studied, outcome measures reported, and study design. Outcomes such as speech perception and speech and language were measured in several studies. These outcome measures were presented in tables, classified into sub-categories so that comparisons could easily be made between studies, and gaps in the literature visualised. Outcome measures used in more sparsely represented areas
such as spatial listening were described in the text. We also catalogued the type of data available from each record, and whether or not statistical comparisons were made between HA and CI outcomes. We catalogued the records by participants’ country of residence, publication year, and study type and size. These areas were first analysed separately, to characterise the available information. To identify gaps in knowledge we then synthesized the results across participant, intervention, outcome measures, and study type. It is beyond the remit of a scoping review to conduct evidence synthesis (Arksey & O’Malley 2005), therefore this was not performed.

Stakeholder consultation

We also carried out the optional stage of the scoping review methodology recommended by Arksey and O’Malley (2015) that involved seeking stakeholder feedback on the results of the review. We approached nine experts, covering a range of relevant professions and expertise, for comments on a full manuscript of preliminary findings that included 18 studies published up to October 2019. They were asked to comment on the appropriateness of our interpretation of the data, the real-world relevance of the findings, discussion points they felt should be added and suggestions for future research that we had not identified. Responses were received from two stakeholders, one with expertise in researching outcomes in children using HAs and CIs who also works for a government agency advising on the commissioning of healthcare services, and the second a Paediatric Audiologist with expertise in HA fitting and CI referral.

RESULTS

Characteristics of studies relevant to CI candidacy for severely deaf children

The following sections catalogue the records in terms of the children who participated, interventions studied, outcomes and outcome measures used, data and analyses of these outcomes, and study designs.
## Supplemental Table 1. Participant characteristics

<table>
<thead>
<tr>
<th>First Author (Publication year)</th>
<th>Additional Difficulties</th>
<th>Onset of hearing loss</th>
<th>Recruited from</th>
<th>Average better ear unaided thresholds of HA users (dB HL)</th>
<th>Communication mode</th>
<th>Cognitive function</th>
</tr>
</thead>
<tbody>
<tr>
<td>Baudonk (2010)</td>
<td>All congenital</td>
<td>University</td>
<td>All HA users 70 - 90 in the better-hearing ear</td>
<td>All used Dutch oral communication</td>
<td>All had minimal intelligence quotient of 80.</td>
<td></td>
</tr>
<tr>
<td>Baudonk (2011)</td>
<td>All prelingual</td>
<td>University</td>
<td>HA group mean = 83.33. No range stated</td>
<td>All enrolled in oral/aural rehabilitation programs.</td>
<td>Normal non-verbal intelligence.</td>
<td></td>
</tr>
<tr>
<td>Ching (2015)</td>
<td>All under 3 years</td>
<td>Population based cohort (LOCHI study).</td>
<td>For all eligible individuals, average in the better-hearing ear &gt;60 and described as “severe” (Fig.2)</td>
<td>All used English as primary form of communication, alone, with sign or with another spoken language.</td>
<td>Cognitive ability at or slightly above age-appropriate levels.</td>
<td></td>
</tr>
<tr>
<td>Eriks-Brophy (2013)</td>
<td>All before 6 months.</td>
<td>AVT programmes</td>
<td>Participants HT28 = 73.3, HT01 = 70, HT17 = 63.3, HT07 = 85, HT29 = 61.7</td>
<td>All used English on a regular basis and all children enrolled in AVT programmes.</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>Fitzpatrick (2012)</td>
<td>Known or presumed under 3 years.</td>
<td>Children’s Hospital</td>
<td>Group pure tone average 68.7 (SD=8.5)</td>
<td>More than 90% enrolled in spoken language rehabilitation programmes.</td>
<td>Non-verbal intelligence in the average range.</td>
<td></td>
</tr>
<tr>
<td>Hammer (2016)</td>
<td>NS</td>
<td>Schools for deaf children and an Audiology programme</td>
<td>Group mean of 75 for eligible sub-group of 4 year old HA users</td>
<td>All monolingual speakers of Dutch, using auditory / oral communication.</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>Kawar (2019)</td>
<td>All prelingual bilateral sensorineural hearing loss</td>
<td>Deaf and hard of hearing treatment centres under the Ministry of Education</td>
<td>Described as “severe”</td>
<td>All preferred oral communication in Arabic, some exposed to sign language but none used this regularly.</td>
<td>Typical academic performance, no diagnosed learning disabilities or behavioural issues or significant developmental delay</td>
<td></td>
</tr>
<tr>
<td>Study</td>
<td>HA Group</td>
<td>CI Group</td>
<td>Schools, Early Intervention Centres and a CI Centre</td>
<td>Sub-group in Group Comparison Described as “Severe” (2011). Included HA Users with Wide Range of Hearing Impairment for Regression Analyses (2011, 2016).</td>
<td>All English as a Primary Language, Minority of Participants Total Communication.</td>
<td>Normal to Borderline Cognitive Status (Not &gt; 1SD from the Mean).</td>
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<tr>
<td>Leigh (2011) and Leigh (2016)</td>
<td>HA group: NS; CI group: all prelingual.</td>
<td></td>
<td></td>
<td>Sub-group in group comparison described as “severe” (2011). Included HA users with wide range of hearing impairment for regression analyses (2011, 2016).</td>
<td>All English as a primary language, minority of participants total communication.</td>
<td>Normal to borderline cognitive status (not &gt; 1SD from the mean).</td>
</tr>
<tr>
<td>Meister (2015)</td>
<td>NS</td>
<td>Audiology Centres.</td>
<td>Home and School NS</td>
<td>All in sub-group classed as “severe” &gt;60 and ≤80</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>Most (2007)</td>
<td>All prelingual.</td>
<td>NS</td>
<td>Sub-group average = 77 (SD=4.96)</td>
<td>All used spoken language as main form of communication.</td>
<td></td>
<td>No reading difficulties reported by teachers.</td>
</tr>
<tr>
<td>Rezaei (2017)</td>
<td>CI group: 75% congenital, 25% prelingual. HA group: 70.83% congenital, 29.16% prelingual</td>
<td>NS</td>
<td>Group mean = 88.33</td>
<td></td>
<td></td>
<td>Nonverbal IQ within the normal range</td>
</tr>
<tr>
<td>Trevisi (2016)</td>
<td>NS</td>
<td>Audiology Centres.</td>
<td>Participants 20, 24, 26 and 29 met ASHA definition of severe</td>
<td>Some verbal, some sign, some verbal with sign, some very limited communication abilities.</td>
<td></td>
<td>Heterogenous and often severe disabilities including intellectual delay.</td>
</tr>
<tr>
<td>Wong (2017)</td>
<td>NS</td>
<td>Population based cohort (LOCHI study).</td>
<td>All individuals within range 61 - 80</td>
<td>Majority spoken language, some spoken language with sign, one child sign only, some unknown.</td>
<td>Severe HA users nonverbal IQ = -.027(SD=1.18); CI group: 0.024(1.12) (Mean Z scores).</td>
<td></td>
</tr>
<tr>
<td>Lovett (2015)</td>
<td>NS</td>
<td>Hospitals, educational services and charities.</td>
<td>Included HA users with wide range of hearing impairment for regression analyses</td>
<td>All learning spoken English as a first or bilingual language.</td>
<td></td>
<td>Nonverbal IQ whole HA group = 104.7 (SD=12.1); CI group = 107.5 (SD=10.4).</td>
</tr>
<tr>
<td>Study</td>
<td>N=1</td>
<td>N=1</td>
<td>University based hospital centre.</td>
<td>Participants: 2 = 90.00; 4 = 92.50; 5 = 85.00</td>
<td>All used English as the primary spoken language and enrolled in programmes with an emphasis on spoken language.</td>
<td>NS</td>
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<td>----------------------------------------------------------------------------------</td>
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</tr>
<tr>
<td>Gantz (2016)</td>
<td>congenital, diagnosed aged 4 years, N=1 unknown</td>
<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Gratacap (2015)</td>
<td>N=6 congenital, N=1 fluctuating</td>
<td>CI centre.</td>
<td>Participants: BA = 87; CM = 71; PJ = 88; AA = 95; NA = 95; JA = 87; LM = 93</td>
<td>All received speech therapy.</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>Meredith (2017)</td>
<td>All post-lingual progressive</td>
<td>CI centre.</td>
<td>Participants: 3 = 75.00; 4 = 88.75; 5 = 82.5; 6 = 87.50; 8 = 87.5</td>
<td>Normal early speech and language development by parent report.</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>Park (2019)</td>
<td>NS</td>
<td>NS</td>
<td>CI centre at university hospital.</td>
<td>Arm 1 Participants: 3 = 82.5; 6 = 95; 7 = 82.5; 8 = 83.75; 19 = 91.25; 20 = 95</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>Tzifa (2013)</td>
<td>N=2 congenital, N=2 progressive, N=1 acquired</td>
<td>Hospital CI centre.</td>
<td>Participants: 1 = 76.25; 2 = 66.25; 3 = 68.75; 4 = 66.25; 6 = 77.5</td>
<td>Use of spoken language implied in the text.</td>
<td>NS</td>
<td></td>
</tr>
</tbody>
</table>

Key: Additional difficulties: Did the study include children with known additional difficulties that might affect listening and or language development?; AVT = Auditory Verbal Therapy; LOCHI = Longitudinal Outcomes of Children with Hearing Impairment Study; NS = not stated. Individual participants’ unaided thresholds averaged over 0.5 to 4 kHz.
Participants

Supplemental table 1 catalogues key characteristics of the children studied, including the inclusion or exclusion of children with additional difficulties, onset of hearing loss, where children were recruited from, and descriptions of their communication mode and cognitive function. Most studies excluded children with additional difficulties. Exceptions were Wong et al. (2017) where children with and without additional needs were recruited into a population based cohort study, and Trevisi et al. (2016) who studied children with CHARGE syndrome, all of whom had additional difficulties. Most studies that reported cognitive function represented children with typical non-verbal intelligence. The children with CHARGE syndrome studied by Trevisi et al. (2016) communicated using a variety of spoken and signed methods. Otherwise, most studies were of children who primarily used spoken language. Most children were reported to have had congenital or early-onset hearing loss, although it was usually not possible to distinguish congenital severe or profound losses from congenital hearing-loss of a milder degree that later progressed to severe or profound levels.
Figure 2. Intervention and assessment timelines
Figure 2 illustrates participants’ ages at intervention and assessment, where this information was available. For between groups studies, age at first HA fitting for the HA group and age at first implantation for the CI group are plotted side-by-side. For the repeated measures studies, only age at first implantation is plotted. Age at HA fitting ranged from a few months to 7 years. Age at first implantation ranged from under 1 year to 16 years of age, with the children in repeated measures studies typically implanted later than those from between groups studies. Ages at assessment ranged from 2 to 17 years. Less discrepancy in age at assessment was seen between the study design types, reflecting the shorter duration of CI use typically experienced by children in repeated measures studies. Some studies pooled data for the CI and HA groups, for age at first hearing aid fitting (Ching et al. 2015; Eriks-Brophy et al. 2013) or age at assessment (Eriks-Brophy et al. 2013; Kawar et al. 2019; Most et al. 2007). These are plotted as HA group data, with dashed lines indicating the range, where available. Baudonk et al. (2010) did not report a mean or distribution for age at first HA fitting, but reported that their HA group all received their first device before two years of age. Kawar et al (2019) reported that all children studied had been fitted with some device by the age of 1 year. All other data missing from Figure 2 implies that data for ages at intervention or assessment were not available from the article (e.g. Trevisi et al. 2016).
<table>
<thead>
<tr>
<th>First author and publication year</th>
<th>Country</th>
<th>Study Type</th>
<th>CI fitting</th>
<th>CI group size</th>
<th>Severe HA group size</th>
<th>Speech perception</th>
<th>Speech and language</th>
<th>Quality of Life</th>
<th>Psychosocial</th>
<th>Sound localization</th>
<th>Listening fatigue</th>
<th>Balance</th>
<th>Educational</th>
<th>Tinnitus</th>
<th>Music perception</th>
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<tbody>
<tr>
<td>Baudonk (2010)</td>
<td>Belgium</td>
<td>BG</td>
<td>NS</td>
<td>N = 29</td>
<td>N = 15</td>
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<td></td>
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</tr>
<tr>
<td>Baudonk (2011)</td>
<td>Belgium</td>
<td>BG</td>
<td>biCl</td>
<td>N = 13</td>
<td>N = 10</td>
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<td>Canada</td>
<td>BG</td>
<td>uniCl</td>
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<td>N = 5</td>
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<tr>
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<td>Belgium</td>
<td>BG</td>
<td>uniCl</td>
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<td>N = 15</td>
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<tr>
<td>Jallu (2019)</td>
<td>India</td>
<td>BG</td>
<td>NS</td>
<td>N = 15</td>
<td>N = 13</td>
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<tr>
<td>Kawar (2019)</td>
<td>Israel</td>
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<td>NS</td>
<td>N = 19</td>
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<td>BG &amp; RS</td>
<td>uniCl</td>
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<td>G S</td>
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<td>Meister (2015)</td>
<td>Germany</td>
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<td>uniCl</td>
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<td>Most (2007)</td>
<td>Israel</td>
<td>BG</td>
<td>NS</td>
<td>N = 10</td>
<td>N = 15</td>
<td>G S</td>
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<tr>
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<td>Italy</td>
<td>BG</td>
<td>NS</td>
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<td>Wong (2017)</td>
<td>Australia</td>
<td>BG</td>
<td>NS</td>
<td>N = 110</td>
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<tr>
<td>Lovett (2015)</td>
<td>U.K.</td>
<td>RS</td>
<td>biCl</td>
<td>N = 28</td>
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<tr>
<td>Gantz (2016)</td>
<td>U.S.A.</td>
<td>RM</td>
<td>uniEAS</td>
<td>N = 3</td>
<td>IS</td>
<td>IS</td>
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<tr>
<td>Gratacap (2015)</td>
<td>France</td>
<td>RM</td>
<td>uniCl3</td>
<td>N = 7</td>
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<tr>
<td>Meredith (2017)</td>
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<td>RM</td>
<td>uniCl4</td>
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<td>Park (2019)</td>
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<td>UniEAS</td>
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<tr>
<td>Tzifa (2013)</td>
<td>U.K.</td>
<td>RM2</td>
<td>uniCl</td>
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<td></td>
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<td>biCl</td>
<td>N = 2</td>
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</tr>
</tbody>
</table>
Key: CI = multi-channel cochlear implant; uniCI = unilateral CI; biCI = bilateral CI; Bim = bimodal; NS = not specified; uniEAS = unilateral electric-acoustic cochlear implant; BG = between groups study; RS = regression study; RM = repeated measures study; G = group data available; I = Individual data points available, numerical and/or graphical; S = statistical comparison(s) made, either between two groups or before and after CI; R = Regression analyses. In the outcome columns, shaded cells indicate that an outcome was measured during the study. White cells indicate that an outcome was not measured. ¹Up to N=43 children using HAs with mild to profound hearing impairment were included in the regression analyses, however N for children with severe hearing impairment was not stated. ²These data are available from Supplemental Digital Content 2 (http://links.lww.com/EANDH/A165). ³Children across the wider study received either bilateral CIs, bimodal fitting or unilateral CIs. Fitting cannot be determined for the eligible children with severe hearing loss, however they were all assessed post-operatively via one CI used alone. ⁴Children across the wider study received either bilateral CIs or unilateral CIs. Fitting cannot be determined for the eligible children with severe hearing loss, however they were all assessed post-operatively via one CI used alone. ⁵Other outcome areas were assessed pre-operatively for some participants, but are not included as they were not repeated post-operatively.
Table 2 provides a map of available data, including numbers of participants, study type, size, interventions, outcome areas assessed, and the type of data available within each outcome area. Studies had been conducted across many countries, with children recruited from a variety of hospital, educational and university settings. Thirteen studies comprised one or more separate groups of HA and CI users and four studies were of children with severe hearing loss assessed before and after implantation using the repeated measures design. A mixture of group and individual data were available from the between groups studies, while only individual data points were available from repeated measures studies. There was a tendency for HA groups to be smaller than CI groups. Wong et al. (2017) included the largest group of 54 children fitted with HA(s) who had severe hearing loss. Visual inspection of the figures in Lovett et al. (2015) suggests that around 23 children studied met our definition of severe hearing-loss. All other studies had 21 or fewer HA participants meeting our severe hearing loss definition.

**Interventions**

Most children using CIs were implanted unilaterally (Table 2). Five records did not state whether children received one or two CIs. (Baudonck et al. 2011) compared their HA group to one group of unilaterally implanted children, and to a separate group of bilaterally implanted children.

**Outcomes**

Table 2 provides an overview of the outcomes that were assessed. Speech perception and speech and language outcome measures are catalogued in detail in Tables 3 and 4 respectively.
Table 3: Speech perception outcome measures

<table>
<thead>
<tr>
<th>Study</th>
<th>Speech perception in quiet (Presentation level)</th>
<th>Speech perception in noise (Presentation level; Signal to noise ratio)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Phonemes</td>
<td>Words</td>
</tr>
<tr>
<td>---------------------</td>
<td>----------</td>
<td>-------</td>
</tr>
<tr>
<td>Fitzpatrick (2012)</td>
<td>PBK (70 dB SPL)</td>
<td>PBK (70 dB SPL)</td>
</tr>
<tr>
<td>Leigh (2011, 2016)</td>
<td>PBK or CNC (65 dB SPL)</td>
<td>PBK or CNC (65 dB SPL)</td>
</tr>
<tr>
<td>Most (2007)</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Lovett (2015)</td>
<td>-</td>
<td>CAPT (50 dB A)</td>
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<tr>
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<td></td>
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</tr>
<tr>
<td>Gantz (2016)</td>
<td>-</td>
<td>CNC (60 dB A)</td>
</tr>
<tr>
<td>Gratacap (2015)</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Park (2019)</td>
<td>-</td>
<td>CNC (60 dB A)</td>
</tr>
<tr>
<td>Meredith (2017)</td>
<td>LNT (45 dB HL)</td>
<td>PBK (45 dB HL)</td>
</tr>
</tbody>
</table>

Key: ATT = IHR-McCormick Automated Toy Discrimination Test; CAPT = Chear Auditory Perception Test; CCT = Consonant Confusion Task; CNC = Consonant-Nucleus-Consonant test; HeSPAC = Hebrew Speech Pattern Contrasts (Intonation and Pattern Contrasts sub-tests); HINT-C = MPT = Minimal Pairs Test (extended version); PBK = Phonetically Balanced Kindergarten Test; WPPT = Word Pattern Perception Test; NS = not stated. Presentation levels are described using the units from the original manuscripts. ¹These data are available from Supplemental Digital Content 2 (http://links.lww.com/EANDH/A165).
<table>
<thead>
<tr>
<th>Study</th>
<th>Phonological skills</th>
<th>Receptive language / comprehension</th>
<th>Expressive language</th>
<th>Speech production</th>
<th>Voice</th>
<th>Communication / Pragmatics</th>
<th>Auditory Performance</th>
</tr>
</thead>
<tbody>
<tr>
<td>Baudonk (2010)</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>Consonant production&lt;sup&gt;1&lt;/sup&gt;</td>
<td>-</td>
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<tr>
<td>Baudonk (2011)</td>
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<td>-</td>
<td>-</td>
<td>Intelligibility</td>
<td>PESP&lt;sup&gt;2&lt;/sup&gt;</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Ching (2015)</td>
<td>CTOPP: sound matching</td>
<td>PPVT-4</td>
<td>-</td>
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<tr>
<td>Eriks-Brophy (2013)</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>GFTA</td>
<td>KLPA-2</td>
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<tr>
<td>Fitzpatrick (2012)</td>
<td>CTOPP: memory and analysis CTOPP: rapid naming</td>
<td>CELF: Core language score PPVT-III</td>
<td>CELF: Core language score</td>
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<td>Finite verb morphology&lt;sup&gt;3&lt;/sup&gt;</td>
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<td>Jallu (2019)</td>
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<td>Morpho-syntactic error; Complex sentences; MSAE</td>
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<td>Meister (2015)</td>
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<td>-</td>
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<td>Rezaei (2017)</td>
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<td>-</td>
<td>Intelligibility</td>
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<tr>
<td>Lovett (2015)</td>
<td>-</td>
<td>CELF: Standard score&lt;sup&gt;4&lt;/sup&gt; PLS-4: Standard score&lt;sup&gt;4&lt;/sup&gt;</td>
<td>CELF: Standard score&lt;sup&gt;4&lt;/sup&gt; PLS-4: Standard score&lt;sup&gt;4&lt;/sup&gt;</td>
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<td>GFTA</td>
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<td>APP</td>
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<td>Tzifa (2013)</td>
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<td>SIR</td>
<td>-</td>
<td>-</td>
<td>CAP</td>
<td>-</td>
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<td>PLS-4 (auditory comprehension)</td>
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<td>PEACH</td>
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</table>
CASL = Comprehensive assessment of spoken language; CELF = Clinical Evaluations of language fundamentals test [Semel & Wiig, 2006];

1 Consonant errors, distortions, substitutions and omissions, final consonant deletion, cluster reductions, liquid gliding, stopping, devoicing, assimilation of sounds, substitutions of /n/ by /m/ and of /s/ by /ʃ/.

2 Grade, roughness, breathiness, astenicity, strain, instability, hypernasality, hyponasality and cul-de-sac.

3 Mean length of utterance, finite verb production, Subject-verb agreement errors / omissions.

4 These data are available from Supplemental Digital Content 2 (http://links.lww.com/EANDH/A165).
In terms of speech perception, the majority of assessments were conducted in quiet (Table 3). Fewer studies addressed speech perception in background noise and presentation levels, type of noise, and signal to noise ratios differed. The speech materials used also varied and included phonemes, words, sentences, and supra-segmental features.

Various outcome measures were used to assess speech and language outcome skills (Table 4). Speech production was the most frequently assessed speech and language outcome, reported in seven articles. Expressive language was the next most frequently assessed speech and language outcome, reported in five. Phonological skills, receptive language comprehension, and proxy reports of general auditory performance were each reported in more than one article. Only one study assessed voice (Baudonck et al. 2011).

Quality of life scores for 38 individuals using HAs, some of whom had severe hearing-loss, are plotted in a scatterplot alongside a histogram of scores from 22 children with BiCIs in the Supplemental Digital Content 2 file of Lovett et al. (2015). Psychosocial outcomes were reported in one article only (Wong et al. 2017). Personal and group interaction and social behaviours were assessed with the social sub-scale of the Child Development Inventory. The Strengths and Difficulties Questionnaire (Goodman 1997) was used to assess emotion, conduct, hyper-activity and peer problems.

With regard to spatial listening, Lovett et al. (2015) also presented left-right discrimination and sound-source localization results in their Supplemental Digital Content 2 file. Left-right discrimination was assessed using loudspeakers situated at 30 degrees to the left and to the right of the children. Sound-source localization was assessed using 5 loudspeakers spaced at 30 degree intervals from -60 to +60 degrees azimuth. Localization accuracy before and after implantation were reported for three eligible individuals in one study only (Gantz et al. 2016), using the Everyday Sounds Localization Test (Dunn et al. 2005), presented at 60 dB(A) from an array of eight loudspeakers arranged in an arc of approximately 108 degrees. Accuracy was determined by calculating the average root mean square error in degrees (Gantz et al. 2016).
The only educational outcome assessed was literacy, which was reported in two studies. Fitzpatrick et al. (2012) assessed word and pseudo-word reading skills via the Wechsler Individual Achievement Test version 2 (Wechsler 2001). Outcomes for HA and CI users were also compared on the Gray Silent Reading Test (Wiederholt et al. 2000) and the spelling sub-test of the Peabody Individual Achievement Test (revised) (Markwardt 1998). Pre- and post-implantation scores on the Woodcock Reading Mastery Test (Woodcock 1998) were reported by Gantz et al. (2016).

**Gaps in the evidence base**

Children with additional needs, older children, teenagers, and those using signed communication were sparsely represented in the eligible articles. Other notable gaps were the absence of any comparisons between children using CIs to children with severe hearing loss using HAs related to listening fatigue, balance, tinnitus, and music perception (Table 2). There were also notable gaps in outcomes related to spatial hearing, as no studies reported speech perception in spatially-separated noise or spatial release from masking. Within educational outcomes, there were no comparisons of numeracy, or achievement in examinations. Tables 3 and 4 reveal that other specific aspects of speech perception and speech and language development such as phoneme perception in background noise and pragmatic language skills were also not assessed.

Several further outcomes were measured in only one or two studies. These included quality of life, psychosocial outcomes, sound localization, perception of supra-segmental features, sentence perception in noise, phonological skills, and voice. By cross-referencing Figure 2 with Supplemental Table 1 and Tables 2 to 4 it is possible to identify the populations and interventions involved with the measurement of these sparsely represented outcomes. For example, psychosocial outcomes were only assessed by Wong et al (2017), in large groups of both CI users and HA users with severe hearing loss, all under approximately 6 years of age. Therefore, there is a gap in our knowledge regarding comparison of psychosocial outcomes in older children with severe hearing loss using HAs and their implanted peers.
Literacy was compared between a group of children with severe hearing loss and HAs to a unilaterally-implanted CI group (Fitzpatrick et al., 2012). It was also measured before and after implantation for a small sub-set of individuals with severe hearing loss within a larger study of children who received short electrode arrays to provide electric-acoustic stimulation (Gantz et al. 2016). Figure 2 and Supplemental Table 1 show that most children from both of these studies were known or presumed to have had hearing loss from an early age. However, they received their CIs later, on average, than is current standard practice. This is likely because of limited availability of neonatal hearing screening for the cohort of children assessed by Fitzpatrick et al. and the considerable low-frequency residual hearing present in the children studied by Gantz et al. Therefore, there is a gap in the knowledge regarding both literacy and educational attainment for children with severe hearing-loss using HAs compared to children who received early identification and bilateral intervention, with either two CIs or bimodal fitting.

While word perception in quiet was the most frequently assessed speech perception outcome, cross-referencing Table 3 with Table 2 revealed that only Lovett et al. (2015) reported this outcome for a group of children who all used bilateral CIs. Meredith et al. (2017) reported repeated measures data for six children receiving a unilateral CI. Fitzpatrick et al. (2012) presented group comparisons of only unilateral CI users and the majority of children studied by Leigh et al. (2011; 2016) wore unilateral CIs. Last, Gantz et al. (2016) reported before and after results for children receiving unilateral EAS implants. Therefore, there is a gap in our knowledge regarding word perception in quiet for children using bimodal fitting or bilateral CIs compared to children with severe hearing loss using only HAs, especially for those older than 7 years. Similarly, sentence perception in quiet was measured by Leigh et al. (2011; 2016) and in quiet and noise by Fitzpatrick et al. (2012). Cross-referencing Table 3 with Table 2 revealed that, between these studies, only five bilaterally-implanted children were represented. The reader is invited to use the information presented in this scoping review to identify further gaps in knowledge that may be of particular interest to them.
A great number of studies have reported outcomes for children using HAs or CIs in isolation. However, the current review has identified only a limited number of studies reporting direct comparisons between these interventions, which constitute an important form of evidence upon which to base clinical guidance. This review describes those comparative studies that included children with severe hearing-loss using HAs. By cataloguing what is known and what is not, we have presented a map of data available to support future meta-analyses and evidence syntheses, aid researchers in planning future studies, and inform families of deaf children, policy makers, and practitioners.

It is beyond the remit of a scoping review to conduct evidence synthesis or assess study quality (Arksey & O’Malley 2005). Accordingly, we have not catalogued the outcomes of any statistical comparisons made within the studies, and we advise readers to refer to the original source documents for this information. However, the information provided by the current review can be used to determine whether there is sufficient similarity in how outcomes have been assessed to warrant a formal synthesis of evidence being conducted in a subsequent review. The data map (Table 2) illustrates that few outcomes have been assessed by multiple studies limiting the potential scope for such syntheses. Outcomes such as quality of life, psychosocial outcomes, fatigue, balance, tinnitus and music perception were only reported in one eligible article or none at all. Localization accuracy and literacy were both assessed in two articles but in the case of both outcomes, one study was a repeated measures design and the other a between groups design. Additionally, localization test methods differed between the two studies in loudspeaker number and separation, and the two studies were further distinguished by reporting data from different interventions (bilateral CIs in Lovett et al. 2015; EAS in Gantz et al. 2016). Hence for these outcomes there are few articles available, and significant methodological differences between studies that limit the potential for evidence synthesis. It is possible that a search strategy designed for a systematic review could find additional articles, but the results of the current scoping review suggests that the weight of evidence on these outcomes will be limited until more research is published. The remaining outcomes of speech perception and speech and language development are those addressed in the systematic review of group comparison studies by de Kleijn et al. (2018).
De Kleijn et al., concluded that the heterogeneity of populations studied and inconsistency in the outcome measures used precluded meta-analysis of the comparative studies they identified. While we have identified additional group comparisons published after de Kleijn et al.’s searches (Jallu et al., 2019, Kawar et al., 2019, Park et al., 2019 and Meredith et al., 2017), the heterogeneity in the outcome measures and populations studied remains. We suggest that more consistent approaches to data collection, for example by developing core outcome sets or establishing other forms of international consensus on choice of outcomes, would aid future evidence syntheses in addition to greater consistency in how study populations are defined by inclusion and exclusion criteria. Scoping reviews such as this are complimentary to systematic reviews such as that of de Kleijn et al. (2018) in part because they can catalogue a wider range of outcomes, and study designs, and in doing so identify gaps for further research to address, and opportunities for formal meta-analyses to be conducted where there is evidence of comparable outcome data.

We identified studies from a range of countries, reporting outcomes from children recruited from a variety of hospital, educational and university settings. Despite the large number of records published that compared outcomes for children using HAs to children using CIs, only 21 were eligible for inclusion in the review. The most frequent cause for exclusion was that even large studies rarely reported outcomes for children with unaided thresholds of between 61 to 95 dB HL using HAs in isolation. Grouping these children together with children with better hearing is likely to improve the overall performance of the HA group on aural / oral outcomes, whereas grouping them with children using HAs with profound deafness will likely have the opposite effect. Some studies grouped all HA users’ results together across the whole range of hearing loss. We did retain any comparative studies where correlations were made between outcomes of CI users and HA users with a wide range of audiometric thresholds (Leigh 2011 & 2016, Lovett 2015), but most studies that included children using HAs with a wide range of losses only presented group summary data. Doing so increases the variance in the HA group data. Each of these group summary approaches therefore decreases the likelihood of identifying differences between HA and CI users that could be relevant and important in defining the audiometric cusp of suitability for CIs. The capacity of future research to contribute to audiometric criteria development might also be strengthened by
characterising children in terms of device acceptance following fitting and daily device use, in addition to age at fitting. These data were not charted, as it was not readily available for any of the studies included.

Our inclusion of a broad range of outcomes enables this review to identify gaps in the evidence base for comparisons of children with average unaided hearing thresholds from 61 to 95 dB HL using HAs and children using CIs. Most studies assessed speech perception, speech production and / or language development. We have catalogued differences in the participant characteristics, and interventions and outcome measures these studies used, that may account for discrepancies in their findings. For example, in terms of participant characteristics, long-term outcomes for children born profoundly deaf are influenced by age at intervention (Yoshinaga-Itano & Sedey, 2010; Ching et al. 2018; Yoshinaga-Itano et al. 2018). Some between groups comparisons, albeit a minority, included congenitally-deaf CI users with a wide range of chronological age at implantation, some as late as during their fourth year of life (Figure 2). This likely increased variability in CI group outcomes. Including late-implanted CI users with congenital deafness limits the relevance of these studies to determining candidature for children born with severe hearing-loss or children with acquired or progressive losses, who have better access to sound via HAs during their early years. Future research in CI users with acquired or progressive losses could clarify how comparative outcomes are influenced by hearing device type, while minimizing the potentially confounding effect of early auditory deprivation.

There was little information on the outcomes of older children and teenagers in the literature. Children assessed via repeated measures before and after implantation tended to be older than those in between groups comparisons (Figure 2) but the numbers of children with severe hearing-loss participating in repeated measures studies was small (Supplemental Table 1). It is plausible that outcomes for HA users with severe hearing-loss and CI users might diverge during later childhood and adolescence. Therefore, studies comparing outcomes for older children and teenagers would make an important addition to the literature, and could also improve the availability of some of the missing outcomes that are easier to assess in older children and teenagers (e.g. tinnitus, spatial release from masking and localization). Further research into comparative outcomes for children with additional health or learning needs is
also needed, since these children were underrepresented in the literature despite representing a higher proportion of children with hearing loss than in the general population (Birman et al. 2012; Szymanski et al. 2012). No records included whole groups of children who used sign language as their primary form of communication.

This review also highlights gaps in the evidence base in terms of interventions. Simultaneous, or short-interval sequential, cochlear implantation is becoming the standard of children’s care in more areas of the world (Ramsden et al. 2012; Teagle et al. 2019). Only one study presented data for a HA group compared to one group of unilateral CI users and a separate group of bilateral CI users (N. Baudonck et al. 2011). There is a need for more studies comparing outcomes for HA users with severe hearing loss to bilaterally implanted and bimodal CI users. Only one study reported outcomes on EAS (Gantz et al. 2016), likely because EAS is commonly used for steeply sloping losses where thresholds averaged across the frequency range often fall within the moderate range. Also, the risk of progressive hearing loss means that EAS is not offered to children as often as full array insertion and accordingly there are fewer studies of this intervention.

In line with the scoping review methodology, we charted data on a wide range of outcomes. It is possible that the relative importance of outcomes may vary between and among children, parents, clinicians, researchers and funders. However, our stakeholder consultation indicated that each outcome domain included in this review is valid in this population and relevant to the question of comparative outcomes with HAs or CIs. It is notable that this scoping review identified similar gaps in knowledge as those identified by Bond et al (2009b) when reviewing evidence for the candidature of children with profound hearing loss for CIs. These gaps included measures of quality of life and educational attainment, and outcomes for children with additional needs. These gaps now exist with regard to the candidature of children with severe hearing loss, and are arguably even more urgent to address, now that children with more residual hearing are presenting for CI assessments. With more residual hearing, challenging and more complex outcomes also become increasingly relevant including spatial listening, speech perception in noise, voice, communication, pragmatic skills and music perception. An awareness of comparative outcomes in listening fatigue, tinnitus and balance would also provide a more holistic background against which
candidature decisions could be made. While all of these outcomes were investigated across
the many records captured by our initial literature searches, none were eligible for inclusion
as they did not include groups of HA users with severe hearing loss in isolation.

This review did not address comparative outcomes for all children outside traditional
audiometric CI candidacy. Some repeated measures studies we excluded from the current
review included children whose hearing loss in the implanted ear was moderate pre-
operatively, rather than severe, and who therefore did not meet our inclusion criteria. This led
to the exclusion of some studies evaluating EAS in children with good low-frequency
hearing. In addition, some excluded studies concerned children with asymmetric or single-
sided deafness, where hearing in the ear contralateral to a CI was normal, or the loss was mild
or moderate rather than severe. These groups could be the focus of future reviews. Our
research objectives were focussed on the transition from only acoustic amplification to at
least one CI, rather than on those considering transition from unilateral to bilateral CI. For
this reason, the definition of our CI group encompassed both children using one CI only and
children using a CI and contralateral HA (bimodal). An alternative, complementary approach
would be to review comparisons of children listening bimodally to those listening via
bilateral CIs. This would be of particular relevance to healthcare systems and clinical
scenarios that assess children for cochlear implantation on the basis of their worse-hearing
ear.

Our stakeholders confirmed that the range of outcomes included are relevant and meaningful
to families of children with hearing loss. They also suggested ways in which our
interpretation of the data could be improved. First, by discussing the size, number, variation
in methods, and lack of standardization of outcome measures and result reporting for studies
in this field, and the implications of this for evidence synthesis. Second, they suggested more
discussion of factors such as early device acceptance and daily usage on children’s outcomes,
in addition to the factors we had charted. Last, they alerted us to extra data available via the
LIMITATIONS

A limitation of this study was the financial constraints that prevented access to translation services, resulting in the exclusion of studies without full-texts in English. Because of this, we may have omitted studies written in other languages that are relevant to the objectives of this review.

CONCLUSIONS

This scoping review catalogued recent literature comparing outcomes for children with severe hearing-loss using HAs to those of children using CIs. While several studies were eligible for inclusion, there remain significant gaps in the evidence base for comparative outcomes in these groups. To address these gaps, more studies are needed that include descriptions of outcomes for HA users with severe hearing-loss in isolation from those with other degrees of hearing loss, and compare the outcomes of these children to outcomes for children using CIs who have had more comparable early auditory experience. Studies using interventions such as bilateral digital HA fitting, bilateral CIs and bimodal fitting would ensure relevance to current best practice. Further research is also needed to compare a broader set of outcomes for children with severe hearing-loss to children with CIs, including speech perception in complex background noise, spatial listening, quality of life, listening effort, balance, dizziness, tinnitus, voice, communication, pragmatic language skills, music perception, and educational attainment.

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REFERENCES


https://doi.org/10.1080/14670100.2019.1655868


Vecchiato G, Maglione AG, Scorpecci A, Malerba P, Graziani I, Cherubino P, et al. (2013). Differences in the perceived music pleasantness between monolateral cochlear implanted and
normal hearing children assessed by EEG. Annual International Conference Of The IEEE
Engineering In Medicine And Biology Society, 2013, 5422-5.
https://doi.org/10.1109/EMBC.2013.6610775, 10.1109/EMBC.2013.6610775
candidacy. Cochlear Implants Int, 17 Suppl 1, 36-41. DOI: 10.1080/14670100.2016.1155809
Corporation.
Winn, S. (2007). Employment outcomes for people in Australia who are congenitally deaf:
has anything changed? Am Ann Deaf, 152, 382-390.
https://www.jstor.org/stable/26234465?seq=1
Wong, C. L., Ching, T. Y. C., Cupples, L., et al. (2017). Psychosocial development in 5-year-
old children with hearing loss using hearing aids or cochlear implants. Trends Hear, 21,
2331216517710373. doi: 10.1177/2331216517710373
conditions of children with hearing loss. Clin Exp Otorhinolaryngol, 5 Suppl 1, S73-75. DOI:
10.3342/ceo.2012.5.S1.S73
through early hearing detection and earlier cochlear implantation. Otol Neurotol, 39, 1256-
1263. DOI: 10.1097/MAO.0000000000001976
development in the presence of severe-to-profound hearing loss: A closer look at children
with cochlear implants versus hearing aids. Otol Neurotol, 31, 1268-1274. DOI:
10.1097/MAO.0b013e3181f1ce07