A Systematic Review of the Effectiveness of Hearing Technologies on Speech Perception Outcomes for People with a Severe-to-Profound High-Frequency Hearing Loss

Revue systématique sur l'efficacité des technologies de suppléance auditive pour améliorer la perception de la parole chez les personnes présentant une perte auditive sévère à profonde dans les hautes fréquences

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KEYWORDS

COCHLEAR IMPLANTS ELECTRIC ACOUSTIC STIMULATION FREQUENCY LOWERING HEARING AIDS HIGH-FREQUENCY HEARING LOSS REVIEW SPEECH PERCEPTION OUTCOMES

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Abstract

The objective of this study was to determine which hearing technology, among frequencycompression or frequency-transposition hearing aids and cochlear implants, either in conventional electrical or electric acoustic stimulation, is the most effective to improve speech perception for people with a sensorineural, severe-to-profound high-frequency hearing loss. A systematic literature review was conducted. All types of experimental designs were considered. Guidelines from the Scottish Intercollegiate Guideline Network and the Grading of Recommendations Assessment, Development and Evaluation group were used for data collection, bias assessment, and quality of scientific evidence quoting. Seventy-four articles were selected. The scientific quality was low or very low for all outcomes because of serious study limitations, inconsistency, or imprecision across studies. Considering the level of scientific quality, it is not possible to state with certainty which treatment is best for the target population. From the available data, while there is a tendency favouring the electric acoustic stimulation implant as the most effective technology to improve speech recognition on two main outcomes, the use of frequency-lowering hearing aids was also beneficial to some individuals. Overall, the electric acoustic stimulation implant might be the best indication for people with a severeto-profound high-frequency hearing loss, but this would need to be confirmed with further better quality research. In this context, and as the electric acoustic stimulation implant is the most costly and risky alternative, trials with frequency-lowering hearing aids should be considered on an individual basis prior to implantation.

Abrégé

La présente étude avait pour objectif de déterminer, parmi les prothèses auditives avec compression ou transposition fréquentielle et les implants cochléaires en stimulation électrique conventionnelle ou en stimulation électroacoustique, celle qui était la plus efficace pour améliorer la perception de la parole des personnes présentant une perte auditive neurosensorielle sévère à profonde dans les hautes fréquences. Une revue systématique de la littérature a donc été effectuée. Tous les types de devis expérimentaux ont été considérés. La collecte de données, ainsi que l'évaluation du risque de biais et de la qualité des faits scientifiques, ont été effectuées en suivant les lignes directrices proposées par le Scottish Intercollegiate Guideline Network et le groupe Grading of Recommendations Assessment, Development and Evaluation. Au total, 74 articles ont été sélectionnés. La qualité scientifique de l'ensemble des variables à l'étude était faible ou très faible en raison de limitations importantes dans les études, ainsi que d'inconsistances et d'imprécisions dans les effets notés d'une étude à l'autre. Compte tenu du niveau de qualité scientifique, il n'a pas été possible d'affirmer avec certitude quel serait le meilleur traitement pour la population ciblée. Bien que les données disponibles suggèrent une tendance qui favorise l'implant cochléaire en stimulation électroacoustique à titre de technologie la plus efficace pour améliorer la reconnaissance de la parole sur les deux variables principales de comparaison, l'utilisation de prothèses auditives avec abaissement fréquentiel a également été montrée bénéfique pour certains individus. Dans l'ensemble, l'implant cochléaire en stimulation électroacoustique semble être la technologie à privilégier pour les personnes présentant une perte auditive sévère à profonde dans les hautes fréquences, mais ce résultat devra être confirmé par la réalisation d'études supplémentaires et de meilleure qualité. Dans ce contexte et considérant que l'implant cochléaire en stimulation électroacoustique est la technologie la plus coûteuse et la plus risquée, l'essai de prothèses auditives avec abaissement fréquentiel devraient être considérés sur une base individuelle avant l'implantation.

People living with a significant hearing loss can generally benefit from the use of hearing aids (HA). However, the effectiveness of HAs for individuals with severe-toprofound sensorineural high-frequency hearing losses (HFHLs) is known to be limited (Ching, Dillon, & Byrne, 1998; Hogan & Turner, 1998). This can be explained by the importance of cochlear damage, causing a severe degradation of frequency resolution capabilities that cannot be compensated by amplification.

Different technological alternatives have been developed to meet the needs of these individuals, such as frequency-lowering HAs-including, among others, frequency-compression and frequency-transposition HAs, or electric acoustic stimulation (EAS) cochlear implants (CIs; Alexander, 2013; Simpson, 2009). Previous research suggests that frequency-lowering HAs are more effective than conventional amplification to support speech recognition for people with a severe-to-profound HFHL. Authors have reported an advantage of frequency-lowering aids ranging from 10% to 20% on different speech recognition tasks for this population (Auriemmo et al., 2009; Bohnert, Nyffeler, & Keilmann, 2010; Glista, Scollie, & Sulkers, 2012; Kuk, Keenan, Korhonen, & Lau, 2009). Frequency lowering remains relatively affordable, and can be reversed (i.e., the feature can be disabled without replacing the HA).

On the other hand, cochlear implantation, either in its conventional electric stimulation or in EAS mode, is nowadays considered as an alternative to amplification for people with a severe-to-profound HFHL and a significant amount of residual hearing in the low frequencies (Sampaio, Araujo, & Oliveira, 2011). The EAS implant is a hearing device that combines a CI with an acoustic HA, allowing simultaneous transmission of low frequencies by conventional acoustic amplification and electric stimulation in high frequencies (von Ilberg, Baumann, Kiefer, Tillein, & Adunka, 2011). Instead of shifting high frequencies to a lower frequency range where there is sufficient residual hearing to process this information, as frequency-lowering HAs would, the EAS implant uses electrical stimulation to directly stimulate the basal portion of the cochlea and ensure an efficient transmission of high-frequency information. CI candidacy criteria have been recently released to include this population considering this technological advance, along with improvements in surgical techniques, electrode designs, and sound processing, which lead to a reduction of implantation related risks and a gain in benefits obtained by implanted patients.

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CIs may not address directly the degradation of frequency resolution capabilities caused by HFHL, but their use is known to improve the perception of high frequencies, which may in turn lead to better speech recognition. Indeed, CIs have been shown to be more effective than HAs to improve speech recognition in patients with a severe-to-profound HFHL (Adunka, Buss, Clark, Pillsbury, & Buchman, 2008; Cullen et al., 2004; Dowell, Hollow, & Winton, 2004). The reported improvements with a CI vary from 20% to 50%; if an EAS processor is used instead of an electric one, patients' performances can improve by another 8% to 17% (Gstoettner et al., 2009; Kiefer et al., 2005; Lorens, Polak, Piotrowska, & Skarzynski, 2008). However, there are several risks associated with cochlear implantation, including the possibility of an irreversible total loss of residual hearing following the surgery, which could greatly limit EAS effectiveness (Talbot & Hartley, 2008). Furthermore, cochlear implantation is a costly intervention in comparison with HAs.

Present Clinical Indications for Frequency-Lowering and EAS Implants

In previous research, proposed clinical indications for EAS were hearing thresholds greater than 80 dB HL at 2000 Hz and above, and thresholds better than 65 dB HL up to 750 Hz, with a maximum word recognition score of 50% to 75% in best fit condition at the ear to be implanted (von Ilberg et al., 2011). The results of a recent international survey (Vickers, De Raeve, & Graham, 2016) suggest that those indication criteria are currently applied in most CI centres around the world. Indications for frequency-lowering HAs appear to be in the same audiometric range. Kuk, Keenan, Peeters, Korhonen, and Auriemmo (2008) suggested that frequency transposition should be used for people with a hearing loss greater than 70 dB HL for high frequencies (above the algorithm cut-off frequency), with thresholds better than 70 dB HL for low to mid frequencies. Another guideline from Scollie et al. (2016) would be to activate frequency lowering only when the gain allowed by the HA is insufficient to reach prescriptive targets in high frequencies, and if the audibility of a calibrated /s/ signal at 65 dB SPL cannot be reached, without frequency lowering. In a clinical setting, those criteria from Scollie et al. (2016) are usually met for most patients with a severe-to-profound HFHL. Therefore, indication criteria for EAS and frequency-lowering HAs are merging, and these technologies are now aiming at improving hearing benefits for the same population.

Previously Published Systematic Reviews

A few systematic reviews have been undertaken on the effectiveness of frequency-lowering HAs and EAS implants, and published articles have addressed those technologies separately. Talbot and Hartley (2008) reported that 92% of the 21 included studies showed an improvement in melody, speech, and sentence recognition with EAS, in comparison with electric stimulation alone, but the authors did not quantify the gain offered by the EAS implant. In a more recent review, which included 27 articles, Incerti, Ching, and Cowan (2013) showed that the improvement in speech recognition with the EAS implant in comparison with electrical stimulation alone was between 6% and 15% for word recognition in quiet and between 4% and 29% for sentence recognition in noise at +10 dB signal-to-noise ratio; when compared to HAs, the reported gain was between 43.8% and 49% for word recognition in quiet. McCreery, Venediktov, Coleman, and Leech (2012) reviewed the available evidence from five studies on frequency-lowering HA effectiveness, with a focus on school-aged children. Results suggested that frequency-lowering HAs can provide a greater benefit than conventional HAs, and this finding was consistent across studies. Due to a great variability in outcome measures used in the reviewed literature, the authors did not specify the extent of the improvement allowed by frequencylowering in percentages, but by using effect sizes when possible. For those three reviews, the evaluation of the scientific quality of the included studies yielded evidence of low to moderate strength.

In summary, people living with a severe-to-profound HFHL now have access to different technological alternatives. To date, a few studies have analyzed available data on frequency-lowering HAs or EAS, but no study has integrated this information to explore which of these alternatives is the most effective to improve speech perception for this population.

Research Question

Among conventional, frequency-lowering, CIs or EAS hearing devices, which technology is the most effective to improve speech recognition for people with a severe-to-profound HFHL?

Method

Design

A systematic literature review was completed following the guidelines by Glasziou, Irwig, Bain, and Colditz (2001) and Higgins and Green (2011).

Eligibility Criteria

All types of experimental designs were considered. To be included, studies had to be based on effectiveness assessments of digital frequencylowering HAs (all types of frequency lowering) or CIs (including EAS and bimodal stimulation) on hearingimpaired participants of all ages with bilateral severeto-profound HFHL. For the purpose of this review, the different types of frequency lowering were classified either as frequency transposition (if the displaced high-frequency signal is superimposed to a base band in lower frequencies) or as frequency compression (if no superimposition of frequency bands occurs during the signal processing). Articles published between 1997 and 2016 in English or French in peer-reviewed scientific journals were considered, along with book chapters and academic theses. Conference abstracts were excluded.

Outcomes

According to the conceptual framework of the Grading of Recommendations Assessment, Development and Evaluation (GRADE) working group (Guyatt, Oxman, Kunz, et al., 2011; Guyatt, Oxman, Schünemann, Tugwell, & Knottnerus, 2011), we identified two main outcomes for this systematic review: speech recognition in quiet and speech recognition in noise. These outcomes were considered critical. Then, all data related to any kind of speech recognition in quiet and in noise were collected. Other speech perception outcomes that have been observed, such as speech detection, discrimination, or identification, were categorized as important but not critical outcomes. Data about subjective benefit, preference, satisfaction, sound quality, and aided puretone detection thresholds were also extracted, as indirect indicators of speech perception; these outcomes were considered important but not critical.

Search Strategy

The following databases and search engines were consulted: Cochrane Library, Medline, Embase, CINAHL, PsycINFO, ProQuest, and Web of Science. Each of these data sources was searched using keywords combined as follows: [(hearing aids) OR (cochlear implant)] AND [speech AND (perception OR recognition OR intelligibility OR detection)] AND [((hearing loss) OR (deafness) OR (hearing impaired persons)) AND (high frequency)]. Google Scholar, reference lists of included studies, and registers of controlled trials (www. clinicaltrials.gov; www.isrctn.com; www.who.int/ictrp/en/) were also searched and contacts were made with key researchers to identify other peer-reviewed published studies that might not have been pointed out by conventional databases' consultation.

Data Collection and Procedures

Titles of studies identified with the literature search were reviewed to exclude those that did not meet inclusion criteria. Then, a second validation of inclusion criteria was realized by reviewing the abstracts of the selected articles. This led to a final list of studies that were included in the complete review process. These studies were read to collect the data pertaining to targeted outcomes, and to critically appraise their scientific quality.

The selection of the studies to be included in the review, the validation of inclusion criteria, and the critical appraisal of the studies were done independently by two reviewers on the first 10 titles. Then, the inter-judge agreement was verified. If disagreements were present between reviewers, they were solved by mutual agreement. Successive review rounds on an additional 10 titles were realized until an inter-judge agreement greater than 90% was reached.

Data collection was supported using extraction forms and checklists from the Scottish Intercollegiate Guidelines Network (Harbour & Miller, 2001; Scottish Intercollegiate Guidelines Network, 2017). Collected data were summarized by outcome in a format inspired by GRADE's evidence profile and summary of findings tables (Guyatt, Oxman, Akl, et al., 2011). In those tables, the reported estimated effects for each outcome represents the range of extracted effects across studies; no averaging was done because of the wide variety of outcome measures, in different languages, that were used across studies.

Critical Appraisal

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The GRADE framework was adopted to appraise the scientific quality of the reviewed evidence (Balshem et al., 2011; Guyatt, Oxman, Schünemann, et al., 2011). This framework was developed by an international group of expert guideline developers, and has been endorsed by organizations such as Cochrane, the National Institute for Health and Care Excellence, Scottish Intercollegiate Guidelines Network, and the World Health Organization. It was recently used in audiology by the authors of a systematic review on the effectiveness of early sign and oral language intervention for spoken language development in children with a hearing loss (Fitzpatrick et al., 2016).

According to the GRADE approach, the scientific quality of reviewed data was rated on a 4-point scale (high, moderate, low, and very low), by outcome and across studies. An initial rating of quality was done for each outcome based on the research design used in individual studies. Next, the initial quotation of each outcome was adjusted if the presence of limitations was observed in individual studies following the risk of bias assessment. Then, data were scanned to detect inconsistency and imprecision of the estimated effect, indirectness of outcome measures, and publication bias. If these global biases were identified and considered serious, the quality rating of concerned outcomes was downgraded. Finally, strengths in studies were also considered, namely if the effect on a particular outcome was large across studies. If strengths were observed, the quality rating of the outcome was rated up.

The risk of bias assessment was based on GRADE's (Guyatt, Oxman, Vist, et al., 2011) and Dollaghan's (2007) recommendations. Studies were checked to detect lack of blinding, randomization, control or allocation concealment, incomplete accounting of patients and outcome events, selective outcome reporting, too short follow-up, unvalidated outcome measures, carryover effects, recruitment bias, sample size, adequacy of statistical analyses, funding sources, author's conflicts of interest, and any other source of potential bias that drew the reviewer's attention. The GRADE 3-point scale for risk of bias was used (low, serious, and very serious). Each study received a risk of bias rating by outcome, meaning that a single study may have received more than one rating, depending on the number of outcomes assessed.

For the purpose of this review, the sample size of a research project was considered too small when fewer than 15 participants completed the study. This choice was based on the minimum number of participants that is generally accepted for parametric statistical

analyses and beyond which the use of non-parametric analyses is recommended (Rosner, 2006). However, when a justification was presented to demonstrate that the sample size was sufficient (such as an a priori sample size or a posteriori power calculation), the "small sample size" risk of bias was not attributed, regardless of the sample size. Statistical analyses were considered incomplete, non-optimal, or flawed when no analyses were provided; when parametric instead of non-parametric methods were used on a small sample size without any justification; when no post hoc analyses or adjustment for multiple comparisons after an analysis of variance, or no analysis of variance, were made when the research design used would have called for it; if an alpha threshold greater than .05 was used; and when errors were made in interpreting p-values. Also, a too short follow-up period was defined as a trial shorter than eight weeks for frequency-lowering HAs, and 12 months following initial activation for EAS and CI technologies. These choices were made based on previous research showing significant improvements in speech perception after six to 10 weeks with frequency lowering (Auriemmo et al., 2009; Ellis & Munro, 2015; Glista et al., 2012; Kuk et al., 2009), and a more important and faster progression of speech perception during the first 12 months following implantation (Blamey et al., 2013).

Results

Seventy-four research articles were included¹. The literature search strategy flow chart is presented in Figure 1. From those 74 articles, 14 assessed the effectiveness of frequency-compression HAs, covering years 2005 to 2016, seven were on frequency-transposition HAs (from 1997 to 2016), 51 explored the EAS implant (from 1999 to 2016), and two covered conventional CIs in electrical stimulation (from 2007 to 2016). No randomized controlled trial was found. The most frequent research designs used were non-randomized trials (using within-subjects repeated measures) and cross-sectional studies. No study comparing the effectiveness of frequencytransposition or frequency-compression HAs and of the EAS implant directly with each other was found. Assessments were generally made in comparison with conventional HAs and also, in the case of the EAS implant, in comparison with the conventional CI in electrical stimulation.

Scientific Quality Analysis

According to the GRADE approach, the quality of the reviewed evidence was low or very low for all outcomes, meaning that "our confidence in the effect estimate is limited: the true effect may be substantially different from the estimate of the effect" (Balshem et al., 2011, p. 404). While this review provides evidence of the present state of knowledge, the level of the scientific evidence calls for caution when interpreting the results. A summary of the quality analysis, by outcome and across studies, is presented in tables 1 to 5.



Figure 1. Literature Search Strategy Flow Chart.

Table 1. Quality Assessment and Summary of Findings: Frequency-Transposition HA vs. Conventional HA

No. of studies (participants)	Limitations	Inconsistency	Indirectness	Imprecision	Publication bias	Particular strengths	Range of reported absolute effect across studies	Quality
Outcome: % monos	yllable recognition ir	nquiet						
4 (17)	Serious	Serious	Not serious	Serious	Not detected	No	0 to +9.1% *	Very low
Outcome: % phoner	ne recognition in qu	iet						
1(6)	Serious	Serious	Not serious	Serious	Not detected	No	+6.6% *	Very low
Outcome: % conson	ant recognition in q	uiet						
3 (23)	Very serious	Serious	Not serious	Serious	Not detected	No	0 to +25% *	Very low
Outcome: % conson	ant recognition in n	oise (+5 dB SNR)						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	+17%	Very low
Outcome: % vowel re	ecognition in quiet							
1 (10)	Very serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % fricative	e recognition in quie	it						
2 (13)	Very serious	Serious	Not serious	Serious	Not detected	No	-15 to +20% *	Very low
Outcome: % fricative	e recognition in nois	e (+5 dB SNR)						
1(8)	Serious	Notrelevant	Not serious	Not serious	Not detected	No	+15%	Very low
Outcome: % stop red	cognition in quiet							
2 (13)	Serious	Serious	Not serious	Serious	Not detected	No	-12.5 to 0% *	Very low
Outcome: % stop red	cognition in noise (+	5 dB SNR)						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % affricat	e recognition in quie	et						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % affricat	e recognition in nois	se (+5 dB SNR)						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % approx	imant recognition in	quiet						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % approx	imant recognition in	noise (+5 dB SNR)						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % nasal re	ecognition in quiet							
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % nasal re	ecognition in noise (-	+5 dB SNR)						
1(8)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: /s/ detect	tion in quiet							
1(5)	Very serious	Not relevant	Not serious	Not serious	Not detected	No	NS	Very low

Outcome: Speech recognition threshold for sentences in noise

2(14)	Serious	Not serious	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: Speech	recognition threshold	for monosyllables in a	quiet					
1 (1)	Serious	Not relevant	Not serious	Not serious	Not detected	No	-15 dB	Very low
Outcome: Aided p	ure-tone detection th	resholds in quiet (high	frequencies)					
2 (11)	Not serious	Not serious	Serious	Not serious	Not detected	No	-60 to -20 dB	Low
Outcome: Environ	mental sound detecti	on						
1 (10)	Serious	Not relevant	Serious	Serious	Not detected	No	Improved	Very low
Outcome: Benefit								
2 (15)	Serious	Not serious	Serious	Serious	Not detected	No	Improved**	Very low
Outcome: Subject	ive preference for free	quency lowering						
2 (15)	Serious	Serious	Serious	Not serious	Not detected	No	N.S.	Very low

Note. * = Reports of deleterious effects on some individuals; ** = for some individuals. HA = Hearing aids; N.S. = Not significant; SNR = Signal-to-noise ratio.

Table 2. Quality Assessment and Summary of Findings: Frequency-compression HA vs. Conventional HA

No. of studies (participants)	Limitations	Inconsistency	Indirectness	Imprecision	Publication bias	Particular strengths	Range of reported absolute effect across studies	Quality
Outcome: /s/ detec	tion threshold in qu	iiet						
2(16)	Serious	Serious	Not serious	Serious	Not detected	No	-10 to 0 dB *	Very low
Outcome: /sh/ dete	ction threshold in q	luiet						
2 (16)	Serious	Not serious	Not serious	Serious	Not detected	No	-10 to 0 dB *	Very low
Outcome: % senten	ce recognition in qu	uiet						
3 (12)	Serious	Serious	Not serious	Serious	Not detected	No	0 to +24% *	Very low
Outcome: % senten	ce recognition in no	oise (+10 dB SNR)						
2(9)	Serious	Serious	Not serious	Serious	Not detected	No	-7 to 0% *	Very low
Outcome: % word re	ecognition in quiet							
1(6)	Very serious	Not relevant	Not serious	Serious	Not detected	No	+12.7% *	Very low
Outcome: % monos	yllable recognition	in quiet						
1(6)	Serious	Serious	Not serious	Serious	Not detected	No	0 to +16%	Very low
Outcome: % phone	me recognition in q	uiet						
4 (33)	Serious	Serious	Not serious	Serious	Not detected	No	0 to +6% *	Very low
Outcome: % consor	nant recognition in o	quiet						
10 (91)	Serious	Serious	Not serious	Serious	Not detected	No	0 to +20% *	Very low
Outcome: % consor	nant recognition in r	noise (0 dB SNR)						
1(7)	Serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % vowel r	ecognition in quiet							
3(43)	Notserious	Serious	Not serious	Serious	Not detected	No	-5.6 to +5% *	Very low
Outcome: % fricativ	e recognition in qui	et						
2(22)	Serious	Serious	Not serious	Serious	Not detected	No	0 to +10% *	Very low
Outcome: % /s/ dete	ection in quiet							
2 (16)	Serious	Not serious	Not serious	Not serious	Not detected	No	0 to +21%	Very low
Outcome: % /s/ and	/sh/ discrimination	in quiet						
1(6)	Serious	Not relevant	Not serious	Not serious	Not detected	No	0 to +20%	Verylow

Outcome: Speech recognition threshold for sentences in noise

4 (36)	Serious	Serious	Not serious	Serious	Not detected	No	-7.4 to 0 dB *	Very low
Outcome: Speech	recognition threshold	d for spondees in noi	se					
2(17)	Serious	Not serious	Not serious	Not serious	Not detected	No	N.S.*	Very low
Outcome: Aided p	oure-tone detection th	nresholds in quiet (in	high frequencies)					
4 (15)	Serious	Not serious	Serious	Serious	Not detected	No	-55 to 0 dB *	Very low
Outcome: Benefit								
5(34)	Serious	Not serious	Serious	Not serious	Not detected	No	Improved **	Very low
Outcome: Satisfac	ction							
1 (11)	Very serious	Notrelevant	Serious	Serious	Not detected	No	-26 to +29% *	Very low
Outcome: Sound	quality							
3 (32)	Serious	Serious	Not serious	Serious	Not detected	No	N.S.	Very low

Note. * = Reports of deleterious effects on some individuals; ** = for some individuals. HA = Hearing aids; N.S. = Not significant; SNR = Signal-to-noise ratio.

Table 3. Quality Assessment and Summary of Findings: EAS Implant vs. Conventional HA

No. of studies (participants)	Limitations	Inconsistency	Indirectness	Imprecision	Publication bias	Particular strengths	Range of reported absolute effect across studies	Quality
Outcome: % senten	ce recognition in qu	liet						
13 (117)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	+39.4 to 92%	Low
Outcome: % senten	ce recognition in no	ise (+10 dB SNR)						
17 (216)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	+24 to 88%	Low
Outcome: % senten	ce recognition in no	vise (+5 dB SNR)						
4 (73)	Serious	Not serious	Not serious	Serious	Not detected	No	+37 to 64% *	Very low
Outcome: % word re	ecognition in quiet							
4 (89)	Serious	Not serious	Not serious	Serious	Not detected	No	+6 to 46%	Very low
Outcome: % word re	ecognition in noise (+10 dB SNR)						
1(30)	Serious	Notrelevant	Not serious	Serious	Not detected	No	+41 to 70%	Very low
Outcome: % monos	yllable recognition i	n quiet						
33 (574)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	+17 to 75% *	Low
Outcome: % monos	yllable recognition i	n noise (+10 dB SNR)						
10 (261)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	+32 to 90%	Low
Outcome: Speech re	ecognition threshol	d for sentences in quiet	-					
1 (22)	Serious	Notrelevant	Not serious	Not serious	Not detected	No	-12 dB	Very low
Outcome: Speech re	ecognition threshol	d for sentences in noise	9					
6 (147)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	-10.2 to -5.7 *	Low
Outcome: Benefit								
10 (183)	Serious	Not serious	Serious	Serious	Not detected	No	12 to 70%	Very low

Note. * = Reports of deleterious effects on some individuals. EAS = Electric acoustic stimulation; HA = Hearing aids; SNR = Signal-to-noise ratio.

Table 4. Quality Assessment and Summary of Findings: EAS Implant vs. Conventional CI

No. of studies (participants)	Limitations	Inconsistency	Indirectness	Imprecision	Publication bias	Particular strengths	Range of reported absolute effect across studies	Quality
Outcome: % senten	ice recognition in qu	liet						
13 (117)	Serious	Not serious	Not serious	Serious	Not detected	No	0 to +29% *	Very low
Outcome: % senten	ice recognition in no	vise (+10 dB SNR)						
17 (216)	Serious	Not serious	Not serious	Serious	Not detected	No	0 to +26.2% *	Very low
Outcome: % senten	ice recognition in no	vise (+5 dB SNR)						
4 (73)	Serious	Not serious	Not serious	Serious	Not detected	No	0 to +25%	Very low
Outcome: % senten	ice recognition in no	vise (0 dB SNR)						
1(7)	Very serious	Not relevant	Not serious	Not serious	Not detected	No	N.S.	Very low
Outcome: % senten	ice recognition in no	vise (-1 dB SNR)						
1 (1)	Serious	Not relevant	Not serious	Not serious	Not detected	No	+22%	Very low
Outcome: % word re	ecognition in quiet							
4 (89)	Serious	Not serious	Not serious	Serious	Not detected	No	0 to +15% *	Very low
Outcome: % word re	ecognition in noise (+10 dB SNR)						
1(30)	Serious	Notrelevant	Not serious	Serious	Not detected	No	+4 to 11%	Very low
Outcome: % monos	syllable recognition i	n quiet						
33 (574)	Serious	Not serious	Not serious	Serious	Not detected	No	0 to +67% *	Very low
Outcome: % monos	syllable recognition i	n noise (+10 dB SNR)						
10 (261)	Serious	Not serious	Not serious	Serious	Not detected	No	+3 to 16.3%	Very low
Outcome: % monos	syllable recognition i	n noise (0 dB SNR)						
2 (34)	Not serious	Not serious	Not serious	Not serious	Not detected	No	+14.2 to 15.3%	Low
Outcome: % phone	me recognition in qu	uiet						
1(9)	Serious	Not relevant	Not serious	Serious	Not detected	No	+7.5 to 27.8%	Very low
Outcome: Speech r	ecognition threshol	d for sentences in quie	t					
1(22)	Serious	Notrelevant	Not serious	Not serious	Not detected	No	-6.9 dB	Very low
Outcome: Speech r	ecognition threshol	d for sentences in nois	e					
6 (147)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	-7.2 to -3.7 dB	Low
Outcome: Speech r	ecognition threshol	d for spondees in noise	9					
5(80)	Serious	Not serious	Not serious	Serious	Not detected	No	-14.4 to 0 dB	Verylow
Outcome: Speech r	ecognition threshol	d for monosyllables in I	noise					
1 (11)	Very serious	Notrelevant	Not serious	Not serious	Not detected	No	-4.0 dB	Verylow
Outcome: Benefit								
10 (183)	Serious	Not serious	Serious	Serious	Not detected	No	0%	Very low

Note. * = Reports of deleterious effects on some individuals. CI = Cochlear implant; EAS = Electric acoustic stimulation; N.S. = Not significant; SNR = signal-to-noise ratio.

Table 5. Quality Assessment and Summary of Findings: Conventional Clvs. Conventional HA

No. of studies (participants)	Limitations	Inconsistency	Indirectness	Imprecision	Publication bias	Particular strengths	Range of reported absolute effect across studies	Quality
Outcome: % speech	recognition for ser	ntences and words in qu	liet and in noise					
1(27)	Very serious	Not relevant	Not serious	Not serious	Not detected	No	+44%	Very low
Outcome: % senten	ce recognition in qu	liet						
7 (73)	Serious	Not serious	Not serious	Not serious	Not detected	Large effect	+35 to 51%	Low
Outcome: % senten	ce recognition in nc	vise (+10 dB SNR)						
10 (147)	Serious	Not serious	Not serious	Serious	Not detected	No	+6 to 80%	Very low
Outcome: % senten	ce recognition in nc	oise (+5 dB SNR)						
1(7)	Serious	Not relevant	Not serious	Not serious	Not detected	No	+50%	Very low
Outcome: % word re	ecognition in quiet							
3 (70)	Serious	Not serious	Not serious	Serious	Not detected	No	+6 to 34%	Very low
Outcome: % word re	ecognition in noise (+10 dB SNR)						
1(30)	Serious	Not relevant	Not serious	Serious	Not detected	No	+30 to 66%	Very low
Outcome: % monos	yllable recognition i	n quiet						
13 (222)	Serious	Not serious	Not serious	Serious	Not detected	Large effect	+21 to 54%	Low
Outcome: % monos	yllable recognition i	n noise (+10 dB SNR)						
2 (114)	Serious	Not serious	Not serious	Not serious	Not detected	No	+25 to 48%	Very low
Outcome: Speech re	ecognition threshol	d for sentences in quie	t					
1(22)	Serious	Not relevant	Not serious	Not serious	Not detected	No	-5 dB	Very low
Outcome: Speech re	ecognition threshol	d for sentences in noise	5					
2 (39)	Serious	Not serious	Not serious	Not serious	Not detected	No	-2 to -1.7 dB	Very low

Note. CI = Cochlear implant; HA = Hearing aids; SNR = Signal-to-noise ratio.

For the majority of outcomes, the risk of bias– labelled as "study limitations" in the tables–was serious. The five most frequently observed risks of biases were a small number of participants; the use of non-optimal, incomplete, or flawed statistical analyses; the non-reporting of funding sources; a too short follow-up; and the funding of studies by the manufacturer. Outcomes were not downgraded because of the absence of blinding, randomization, allocation concealment, or control group; those risks of biases were not taken into account, considering the predominant type of research design used in included studies. A summary of the observed risks of biases and their occurrence is presented in Table 6.

The quality score of many outcomes had to be downgraded, not only because of study limitations, but also for serious inconsistency or imprecision, due to an important variability in the estimate of the effect across studies. The quality scores of a few outcomes were upgraded. This was done when the observed effect on a particular outcome was larger than the minimal detectable change of the test, in the absence of inconsistency and across a significant number of studies. An effect was considered larger than the minimal detectable change when the score change with the assessed hearing technology was greater than 15% to 20% (Raffin & Thornton, 1980; Thornton & Raffin, 1978), or 3 dB SNR (Brand & Kollmeier, 2002).

Outcomes and Effects

EAS implant versus frequency-lowering and conventional HAs. Despite the wide variety of outcomes used in the literature to assess the effectiveness of frequency-transposition and frequency-compression HAs, EAS implants, and conventional CIs, those technologies have been tested on only two shared speech recognition outcomes: monosyllable recognition in quiet (in %) and speech recognition threshold for sentences in noise (in dB SNR). Therefore, those outcomes were selected to compare the performance between the assessed technologies. A summary of this comparison is presented in Table 7. Results show that few data are available about the effectiveness of frequencylowering HAs on the two comparison critical outcomes, and this makes comparisons between HA and CI technologies quite uncertain. Nevertheless, from available data, there is a tendency suggesting that the EAS implant may be the alternative able to provide

the greatest improvement in speech perception over other assessed technologies, when compared to conventional HAs. The conventional CI might also be superior to frequency-lowering HAs for monosyllable recognition in quiet, but not for speech recognition threshold for sentences in noise, for which the frequency-compression HA may be more beneficial. A great variability was observed in the reported effects across studies, especially for implant technologies; on an individual basis, the reported scores were ranging from 0% to 100% in some studies.

Table 8 shows pre-intervention unaided puretone thresholds and speech scores for monosyllable recognition in quiet and speech recognition threshold for sentences in noise for participants of included studies, by technology. Despite the fact that present clinical indications for frequency-lowering and EAS implants are quite comparable, in past research, those technologies seem to have been assessed on patients with dissimilar degrees of hearing loss. From preintervention unaided pure-tone thresholds, it appears that implant technologies were tested on participants with a more severe hearing loss, in comparison with frequency-lowering HAs (with a difference in mean thresholds at 1,000 to 4,000 Hz ranging from 9.8 to 16.2 dB), while frequency-compression and frequency-transposition HAs were assessed on more comparable samples. Few data are available on pre-intervention monosyllable recognition in quiet and speech recognition threshold for sentences in noise outcomes for frequency-lowering HAs, but from available data, speech recognition abilities of participants included in frequency compression studies might have been better than those included in studies that assessed implant technologies.

EAS implants versus conventional CIs, bimodal stimulation and EAS implant plus contralateral HA. EAS and conventional CIs have been compared directly with each other on 16 speech perception outcomes, including 15 critical outcomes and one important outcome. Data comparing EAS implants with bimodal stimulation (i.e., a conventional CI in one ear, with an HA in the contralateral ear) versus EAS implant plus contralateral HA on six of those critical outcomes have also been identified. Details are presented in Table 9.

Table 6. Observed Risks of Biases and Their Occurrence

Code and risk of bias description	Total	Frequency compression	Frequency transposition	EAS implant	Conventional CI
Small sample size	197	42	23	127	5
Non-optimal, incomplete, or flawed statistical analyses	187	40	17	126	4
Funding sources not reported	123	21	13	82	7
Stopping early for benefit	107	28	13	65	1
Study funded by the manufacturer	106	31	10	65	0
One or more of the authors works for the manufacturer	88	9	13	61	5
Limited generalizability of results	64	17	11	36	0
HAs or CI processor fittings not well described	62	10	6	45	1
Incomplete accounting of patients and outcome events (including loss to follow-up)	61	9	1	49	2
Overgeneralized or unsupported conclusions	39	7	6	26	0
Important interindividual variability	38	7	0	30	1
Equivalency between groups not well established	31	0	0	30	1
Issues with research design	30	20	8	2	0
Carryover effects (including learning and training effects)	29	13	12	4	0
Sample heterogeneity	23	12	0	11	0
Unvalidated outcome measures	20	4	3	13	0
Hearing preservation not well defined	14	0	0	14	0
Inadequate outcome measure vs. population	12	1	0	11	0
Selective outcome reporting	7	0	2	5	0
Ceiling effect	7	1	1	5	0
Data collected with heterogeneous outcome measures	5	0	0	4	1
No randomization or counterbalancing (conditions, tasks, or items)	4	0	0	4	0

Note. CI = Cochlear implant; EAS = Electric acoustic stimulation; HA = Hearing aids; SNR = signal-to-noise ratio.

Table 7. Summary of Assessed Technologies' Effects in Comparison with Conventional HAs

Code and risk of bias description		Range of reported absolute effect across studies by technology						
Outcome	Frequency transposition (<i>N</i> studies, <i>N</i> participants)	Frequency compression (<i>N</i> studies, <i>N</i> participants)	EAS implant (N studies, N participants)	Conventional CI (<i>N</i> studies, <i>N</i> participants)				
% monosyllable recognition in quiet	0 to +9.1%(4, 17)	0 to +16% (1, 6)	+17 to 75% * (33, 574)	+21.3 to 54.3% (13, 222)				
Speech recognition threshold for sentences in noise	N.S. (2, 14)	-7.4 to 0 dB * (4, 36)	-10.2 to -5.7 dB * (6, 147)	-2.0 to -1.7 dB (2, 39)				

Note. * = Reports of deleterious effects on some individuals. CI = Cochlear implant; EAS = Electric acoustic stimulation; HA = Hearing aids.

Table 8. Mean Pure-Tone Thresholds and Speech Scores Across Studies, Before Intervention, by Hearing Technology

Mean pure-tone thresholds in hertz (<i>n</i>)										Mean srt in noise, in db SNR (<i>n</i>)
Technology	250	500	750	1000	1500	2000	3000	4000	quiet (n)	
Frequency transposition	33.6 (63)	45.5 (63)	58.4 (63)	69.3 (63)	81.9 (63)	92.5 (63)	95.6 (63)	98.8 (63)	23.1 (11)	16.8 (14)
Frequency compression	46.4 (209)	54.2 (209)	63.7 (209)	69.8 (209)	81.0 (209)	86.8 (209)	91.9 (209)	96.6 (209)	41.0 (6)	4.5 (40)
EAS implant	36.1 (804)	52.5 (804)	69.7 (804)	83.2 (804)	96.1 (764)	102.8 (763)	105.2 (714)	106.5 (695)	29.3 (677)	9.5 (127)
Conventional implant	42.5 (36)	53.8 (36)	74.4 (36)	91.3 (36)	101.9 (36)	112.5 (36)	115.6 (36)	118.8 (36)	30 (27)	n/a

Note. EAS = Electric acoustic stimulation; *N* = Number of observations; n/a = Not available; SRT = Sentence recognition threshold; SNR = Signal-to-noise ratio.

Table 9. Summary of Effects with EAS Implant in Comparison with Conventional CI, EAS Implant + Contralateral HA, and Bimodal Stimulation

	Range of reported absolute effect across studies by technology									
Outcome	EAS implant vs. conventional CI (<i>N</i> studies, <i>N</i> participants)	EAS implant vs. bimodal stimulation (N studies, N participants)	EAS implant + contralateral HA vs. EAS implant (<i>N</i> studies, <i>N</i> participants)	EAS implant + contralateral HA vs. bimodal stimulation (<i>N</i> studies, <i>N</i> participants)						
% sentence recognition in quiet	0 to +29% * (13, 117)	No data collected	No data collected	No data collected						
% sentence recognition in noise (+10 dB SNR)	0 to +26.2% * (17, 216)	Not significant (1, 24)	0 to +9% (3, 59)	Not significant (1, 24)						
% sentence recognition in noise (+5 dB SNR)	0 to +25% (4, 73)	No data collected	No data collected	No data collected						
% sentence recognition in noise (0 dB SNR)	Not significant (1, 7)	No data collected	No data collected	No data collected						
% sentence recognition in noise (-1 dB SNR)	+22% (1, 1)	No data collected	No data collected	No data collected						
% word recognition in quiet	0 to +15% * (4, 89)	Not significant (1, 54)	+9 to 13% (2, 73)	Not significant (1, 54)						
% word recognition in noise (+10 dB SNR)	+4 to 11% (1, 30)	No data collected	No data collected	No data collected						
% monosyllable recognition in quiet	0 to +67% * (33, 547)	No data collected	0 to +19% (6, 111)	0 to +8% (2, 56)						
% monosyllable recognition in noise (+10 dB SNR)	+3 to 16.3% (10, 261)	No data collected	+4.5 to 5.6% (2, 35)	+5% (1, 34)						
% monosyllable recognition in noise (0 dB SNR)	+14.2 to 15.3% (2, 34)	No data collected	No data collected	No data collected						
% phoneme recognition in quiet	+7.5 to 27.8% *(1, 9)	No data collected	No data collected	No data collected						
Speech recognition threshold for sentences in quiet	-6.9 dB (1, 22)	No data collected	No data collected	No data collected						
Speech recognition threshold for sentences in noise	-7.2 to -3.7 dB (6, 147)	Not significant (1, 34)	-1.8 to 0 dB (5, 79)	-4.4 to 0 dB (2, 59)						
Speech recognition threshold for spondees in noise	-14.4 to 0 dB (5, 80)	No data collected	No data collected	No data collected						
Speech recognition threshold for monosyllables in noise	-4.0 dB (1, 11)	-2.2 dB (1, 9)	-2.6 to -1.1 dB (2, 20)	Not significant (1, 9)						
Subjective benefit	Not significant (10, 183)	No data collected	No data collected	No data collected						

Note. CI = Cochlear implant; EAS = Electric acoustic stimulation; HA = Hearing aids; SNR = Signal-to-noise ratio.

Results show that EAS implants might be superior to conventional CIs, but there is a great variability on reported effects across studies, including reports of deleterious effects on some individuals. Many studies have reported no effect of EAS in quiet testing conditions, but also for sentence recognition in noise tasks. However, speech recognition threshold testing in noise appears to favour EAS. Then, the EAS effect may be greater in noisy conditions. Unfortunately, this potential advantage of the EAS implant over conventional CIs is not supported by subjective benefit data.

Few studies have compared the EAS implant with bimodal stimulation versus EAS implants plus a contralateral HA for the targeted population. According to the sparse available data, the addition of a contralateral HA to the unilateral EAS could improve speech perception in quiet and in noise. However, the extent of the improvement appears smaller than the gain obtained with the EAS in comparison to conventional CIs. Also, no sufficient data was found to support the superiority of EAS or EAS plus a contralateral HA over bimodal stimulation.

Discussion

In this systematic review, available data on the efficacy of frequency-transposition HAs, frequencycompression HAs, EAS implants, and conventional Cls were analyzed to determine which of these technologies is the most effective to improve speech perception for people with a severe-to-profound sensorineural HFHL. The reviewed evidence, representing the current state of knowledge on this topic from which clinical fitting decisions should be taken, was generally quoted as being of low or very low scientific quality; therefore, any decision implies some substantial uncertainty.

Results suggest that the EAS implant might represent the technological alternative that can provide the greatest benefit to this clinical population, when compared to conventional HAs, on two critical outcomes selected for the main comparison (monosyllable recognition in quiet and speech recognition threshold for sentences in noise). This benefit may be greater with the use of a contralateral HA in addition to the EAS implant. This would need to be confirmed with further higher quality research. The extent of the EAS implant benefit found in this review is consistent with the results of Incerti et al. (2013).

However, a great variability in the reported EAS implant's effects inter- and intra-studies was observed, including reports of deleterious effects for some individuals. This makes the real effect of EAS implants difficult to specify, and illustrates that the potential benefit of the EAS implant may not be warranted to all individuals, as suggested by Talbot and Hartley (2008). This variability may result from different sources. It may be attributed to interindividual differences between participants across studies, such as hearing level, duration of deafness, and experience with HAs before implantation. Those factors have been correlated with EAS and CI benefit in the past, and depending on their extent, might reflect the possible presence of a neural reorganization of the auditory system, which is an important factor that was not discussed directly in most reviewed studies. The observed variability could also have been caused by technological differences in implant and processor types used in different studies, or in processor and HA fittings, which were not always well described by authors. Another factor to consider is that speech perception outcomes were assessed in different countries, using measurement tools in different languages and possibly with different psychometric properties, which could also explain the discrepancies in reported results across studies. The use of more uniform methods, participants, technologies, or measurement tools across studies may have led to a better estimate of the EAS implant's effect. In this context, making the decision to implant a patient with a HFHL requires a cautious approach.

Reported effects for frequency-transposition and frequency-compression HAs were smaller than for the EAS implant. This result may have been influenced by the fact that participants in EAS implant studies had a greater hearing loss than those included in frequency-lowering studies. The amount of possible gain in studies assessing frequency-lowering effectiveness could have been more limited because of participants' better preoperative hearing abilities, which may have given an advantage to the EAS implant over frequency-lowering HAs. If preoperative hearing levels and speech perception abilities of participants in EAS studies have been better, the superiority of the EAS implant might be reduced. We further explored this possibility by extracting the reported effects for monosyllable recognition in quiet for two subgroups of EAS studies, a first group with better preoperative hearing levels (mean threshold at 2000 Hz ≤ 100

dB HL; Adunka, Pillsbury, Adunka, & Buchman, 2010; Arnoldner et al., 2010; Skarzynski & Lorens, 2010; Skarzynski et al., 2012), and a second group with worse preoperative hearing levels (mean threshold at 2000 Hz ≥ 110 dB HL; Gantz, Turner, Gfeller, & Lowder, 2005; Gstoettner et al., 2009; Gstoettner et al., 2006; Kiefer et al., 2005; Lee et al., 2010; Punte, Vermeire, & Van de Heyning, 2010; Simpson, McDermott, Dowell, Sucher, & Briggs, 2009; Usami et al., 2014). Results of this analysis are presented in Table 10. It appears that when preoperative hearing levels are better, the gain obtained from an EAS implant may indeed be smaller. However, with comparable pre-intervention hearing levels, the gain is still larger for this EAS sub-group than for the frequency lowering group. This suggests that the potential advantage of the EAS implant over frequency-lowering HAs would still hold if preoperative hearing levels between groups were the same. As clinical indications for those hearing technologies are merging, more research is needed to compare the effectiveness of the EAS implant and frequency-lowering HAs with participants having a similar audiometric profile, and on a greater number of outcomes.

Like the EAS implant, effects of frequency-lowering HAs were also highly variable, including reports of deleterious effects in some individuals. This may be related to the fact that stimuli and fitting methods for this technology have been lacking in the past. Recent studies demonstrated that the quality of frequency lowering fitting can be a factor both in benefit and in deleterious effects, at least with frequency compression (Souza, Arehart, Kates, Croghan, & Gehani, 2013). Nevertheless, a significant positive effect was found in some studies and individuals using those technologies. This suggests that frequencylowering HAs may be a valuable treatment alternative to conventional HAs for some patients, which is consistent with the findings of McCreery et al. (2012). Thus, exploring the benefit of frequency-lowering HAs with individual patients before implantation would be a valuable step in the candidacy assessment process. Considering the risks and costs related to cochlear implantation, trials with frequency-lowering HAs prior to implantation may help to determine if a given patient could benefit from this technology to a greater extent than with conventional HAs. While the benefit might eventually be smaller with frequency-lowering HAs than with an EAS implant, it could be mostly significant for some patients, leading them or the clinician to reconsider implantation.

Conversely, results of the trial could not be significant for the patient; then, the decision to proceed with implantation would be reinforced. In either case, the CI candidacy assessment process would be better informed and evidence-based. This would lead to better informed consents and clinical decisions, and might be especially useful with borderline candidates such as those with more residual hearing or speech perception abilities (i.e., candidates who may lose hearing abilities following implantation), or those who are poorly motivated.

As reported by Talbot and Hartley (2008) and Incerti et al. (2013), the risk of losing residual hearing following surgery for a CI is real. But data collected in this review suggest that in cases where the patient cannot benefit from an EAS processor after implantation, the benefit of a conventional CI processor on speech perception would probably still be greater than the benefit that would have been gained from frequency-lowering or conventional HAs. Moreover, if the contralateral ear of the patient can be fitted with an HA to give access to bimodal stimulation, then the benefit of speech perception may reach the same level as with an EAS processor. Those results are reassuring from ethical and clinical perspectives, meaning that a patient who loses residual hearing following implantation would still have access to valuable technological options.

Scientific Quality of Reviewed Evidence

According to the GRADE framework, the quality of the evidence reviewed in this study was globally quoted as low or very low for all assessed outcomes. This is not really surprising, since the GRADE quality scale includes only four steps (high, moderate, low, and very low) and because outcomes issued from non-randomized trials or observational studies, which was the case for all outcomes in this review, are systematically quoted as being low-quality evidence (Balshem et al., 2011). This quoting may appear unfair to many included studies. Indeed, the use of the idealized randomized controlled trial design is not always feasible, nor desirable, in CI and HA research, and a repeated-measure within-subject design is often seen as better suited to the field. However, GRADE-like most scientific evidence quality scalesdoes not consider repeated-measure within-subject designs to be equal to randomized controlled trials or better than good quality observational designs.

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Table 10. Mean Pure-Tone Thresholds and Speech Scores Across EAS Studies for Participants with Better and Worse Preoperative Hearing Levels

Mean pure-tone thresholds in Hertz (number of observations)									% Mean preoperative monosyllable recognition in quiet (number of observations)	% Mean monosyllable recognition in quiet; Range of reported absolute effect across studies
Technology	250	500	750	1000	1500	2000	3000	4000		
EAS implant with better preoperative hearing	37.5 (221)	52.4 (221)	67.8 (221)	80.3 (221)	92.1 (221)	97.4 (221)	102.6 (200)	105.2 (200)	31.3 (129)	21 to 56
EAS implant with worse preoperative hearing	34.2 (76)	55.0 (76)	76.4 (76)	93.2 (76)	106.6 (76)	112.4 (76)	113.1 (59)	113.4 (49)	20.2 (70)	32 to 61.9

Note. EAS = Electric acoustic stimulation.

Therefore, the use of another scientific quality scale would probably not have led to better quality ratings.

To obtain a higher degree of quality using the GRADE scale, studies should not present serious or very serious limitations (risks of biases); the evidence should be consistent and precise across studies; and they should present specific strengths, such as a large or very large effect. That was not the case for most assessed outcomes. A downgrade to a very low level of quality may have been avoided in many studies with better research designs. For example, justifying the sample size with the help of an a priori sample size and a posteriori power calculations, using better statistical analyses, correctly reporting funding sources, using a longer follow-up, seeking better independence from the manufacturer, or describing more precisely the fitting techniques used, may have led some outcomes to reach a moderate level of scientific quality. Authors should consider seeking better ways to control for risks of biases in their future research, which could raise the level of quality and improve stakeholders' confidence in the decision process.

Study Limitations

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In this systematic review, the effectiveness of hearing technologies for people with an HFHL was assessed by considering speech perception outcomes. However, the benefit from hearing technologies is known to be multidimensional-not only related to speech perception, but also day-today experienced hearing disabilities, limitations in social participation, quality of life, and other personal factors (Gatehouse, 1994). Those outcomes-such as other potential important outcomes for daily living, like music perception and sound localization-were not included in this review. The benefit of frequencytransposition HAs, frequency-compression HAs, EAS implants, and conventional CIs on those outcomes should also be considered when making clinical decisions for the targeted population. Another limitation of this review is that comparisons between the effectiveness of frequency-lowering HAs and CI technologies were done indirectly, by comparing them to conventional HAs. No study comparing frequency compression and frequency transposition directly with each other or directly to CI technologies was found for people with a severe-to-profound HFHL. Future research should address this lack of evidence.

Conclusions

From the available data, which are of poor scientific quality, the EAS implant might appear to be the first indication for treating people with an HFHL. However, frequency-transposition and frequency-compression HAs can provide some benefit for individuals. In this context, and considering the potential risks and high costs related to cochlear implantation, trials with frequency-lowering HAs should be considered on an individual basis prior to implantation. More research of higher scientific quality, based on repeated-measures or cross-over designs with a better control of risks of biases, is needed to circumscribe more precisely and with more confidence the benefit of the EAS implant, and to compare it directly with frequency-lowering HAs, on a greater number of shared outcomes and on patients with a similar audiometric profile. This may help to define clearer clinical indications for each technology and to better guide clinical decisions made with patients with an HFHL.

References

- Adunka, O. F., Buss, E., Clark, M. S., Pillsbury, H. C., & Buchman, C. A. (2008). Effect of preoperative residual hearing on speech perception after cochlear implantation. *The Laryngoscope, 118,* 2044–2049. doi: 10.1097/ MLG.0b013e3181820900
- Adunka, O. F., Pillsbury, H. C., Adunka, M. C., & Buchman, C. A. (2010). Is electric acoustic stimulation better than conventional cochlear implantation for speech perception in quiet? *Otology and Neurotology*, *31*, 1049–1054. doi: 10.1097/MAO.0b013e3181d8d6fe
- Alexander, J. (2013). Individual variability in recognition of frequency-lowered speech. Seminars in Hearing, 34, 86–109. doi: 10.1055/s-0033-1341346
- Arnoldner, C., Helbig, S., Wagenblast, J., Baumgartner, W. D., Hamzavi, J. S., Riss, D., & Gstoettner, W. (2010). Electric acoustic stimulation in patients with postlingual severe high-frequency hearing loss: Clinical experience. Advances in Oto-Rhino-Laryngology, 67, 116–124. doi: 10.1159/000262603
- Auriemmo, J., Kuk, F., Lau, C., Marshall, S., Thiele, N., Pikora, M., ... Stenger, P. (2009). Effect of linear frequency transposition on speech recognition and production of school-age children. *Journal of the American Academy of Audiology, 20,* 289–305. doi: 10.3766/jaaa.20.5.2
- Balshem, H., Helfand, M., Schünemann, H. J., Oxman, A. D., Kunz, R., Brozek, J., . . . Guyatt, G. H. (2011). GRADE guidelines: 3. Rating the quality of evidence. *Journal of Clinical Epidemiology*, 64, 401–406. doi: 10.1016/j. jclinepi.2010.07.015
- Blamey, P., Artieres, F., Baskent, D., Bergeron, F., Beynon, A., Burke, E., . . . Lazard, D. S. (2013). Factors affecting auditory performance of postlinguistically deaf adults using cochlear implants: An update with 2251 patients. *Audiology & Neurotology*, *18*, 36–47. doi: 10.1159/000343189
- Bohnert, A., Nyffeler, M., & Keilmann, A. (2010). Advantages of a non-linear frequency compression algorithm in noise. *European Archives of Otorhinolaryngology*, 267, 1045–1053. doi: 10.1007/s00405-009-1170-x

- Brand, T., & Kollmeier, B. (2002). Efficient adaptive procedures for threshold and concurrent slope estimates for psychophysics and speech intelligibility tests. *The Journal of the Acoustical Society of America*, *111*, 2801–2810. doi: 10.1121/1.1479152
- Ching, T. Y. C., Dillon, H., & Byrne, D. (1998). Speech recognition of hearing-impaired listeners: Predictions from audibility and the limited role of high-frequency amplification. *The Journal of the Acoustical Society of America*, *103*, 1128–1140. doi: 10.1121/1.421224
- Cullen, R. D., Higgins, C., Buss, E., Clark, M., Pillsbury, H. C., & Buchman, C. A. (2004). Cochlear implantation in patients with substantial residual hearing. *The Laryngoscope*, 114, 2218–2223. doi: 10.1097/01.mlg.0000149462.88327.7f
- Dollaghan, C. A. (2007). The handbook for evidence-based practice in communication disorders. Baltimore, MD: Brookes.
- Dowell, R. C., Hollow, R., & Winton, E. (2004). Outcomes for cochlear implant users with significant residual hearing: Implications for selection criteria in children. *Archives of Otolaryngology – Head & Neck Surgery, 130*, 575–581. doi: 10.1001/ archotol.130.5.575
- Ellis, R. J., & Munro, K. J. (2015). Benefit from, and acclimatization to, frequency compression hearing aids in experienced adult hearing-aid users. *International Journal of Audiology*, 54, 37–47. doi: 10.3109/14992027.2014.948217
- Fitzpatrick, E. M., Hamel, C., Stevens, A., Pratt, M., Moher, D., Doucet, S. P., ... Na, E. (2016). Sign language and spoken language for children with hearing loss: A systematic review. *Pediatrics*, 137. doi: 10.1542/peds.2015-1974
- Gantz, B. J., Turner, C., Gfeller, K. E., & Lowder, M. (2005). Preservation of hearing in cochlear implant surgery: Advantages of combined electrical and acoustical speech processing. *Laryngoscope*, *115*, 796–802. doi: 10.1097/01. MLG.0000157695.07536.D2
- Gatehouse, S. (1994). Components and determinants of hearing aid benefit. *Ear and Hearing*, 15, 30–49.
- Glasziou, P., Irwig, L., Bain, C., & Colditz, G. (2001). Systematic reviews in health care: A practical guide. Cambridge, United Kingdom: Cambridge University Press.
- Glista, D., Scollie, S., & Sulkers, J. (2012). Perceptual acclimatization post nonlinear frequency compression hearing aid fitting in older children. *Journal of Speech, Language, and Hearing Research*, 55, 1765–1787. doi: 10.1044/1092-4388(2012/11-0163)
- Gstoettner, W., Helbig, S., Settevendemie, C., Baumann, U., Wagenblast, J., & Arnoldner, C. (2009). A new electrode for residual hearing preservation in cochlear implantation: First clinical results. *Acta Oto-Laryngologica*, *129*, 372–379. doi: 10.1080/00016480802552568
- Gstoettner, W. K., Helbig, S., Maier, N., Kiefer, J., Radeloff, A., & Adunka, O. F. (2006). Ipsilateral electric acoustic stimulation of the auditory system: Results of longterm hearing preservation. *Audiology and Neurotology*, *11*(Suppl. 1), 49–56. doi: 10.1159/000095614
- Guyatt, G., Oxman, A. D., Akl, E. A., Kunz, R., Vist, G., Brozek, J., ... Schünemann, H. J. (2011). GRADE guidelines: 1. Introduction–GRADE evidence profiles and summary of findings tables. *Journal of Clinical Epidemiology*, 64, 383–394. doi: 10.1016/j.jclinepi.2010.04.026
- Guyatt, G. H., Oxman, A. D., Kunz, R., Atkins, D., Brozek, J., Vist, G., ... Schünemann, H. J. (2011). GRADE guidelines: 2. Framing the question and deciding on important outcomes. *Journal of Clinical Epidemiology*, 64, 395–400. doi: 10.1016/j. jclinepi.2010.09.012
- Guyatt, G. H., Oxman, A. D., Schünemann, H. J., Tugwell, P., & Knottnerus, A. (2011). GRADE guidelines: A new series of articles in the Journal of Clinical Epidemiology. *Journal of Clinical Epidemiology*, 64, 380–382. doi: 10.1016/j. jclinepi.2010.09.011
- Guyatt, G. H., Oxman, A. D., Vist, G., Kunz, R., Brozek, J., Alonso-Coello, P., . . . Schünemann, H. J. (2011). GRADE guidelines: 4. Rating the quality of evidence– Study limitations (risk of bias). *Journal of Clinical Epidemiology*, 64, 407–415. doi: 10.1016/j.jclinepi.2010.07.017
- Harbour, R., & Miller, J. (2001). A new system for grading recommendations in evidence based guidelines. *British Medical Journal, 323*(7308), 334–336. doi: 10.1136/bmj.323.7308.334
- Higgins, J. P. T., & Green, S. (2011). Cochrane handbook for systematic reviews of interventions version 5.1.0 [updated March 2011]. Retrieved from www. handbook.cochrane.org

- Hogan, C. A., & Turner, C. W. (1998). High-frequency audibility: Benefits for hearingimpaired listeners. *Journal of the Acoustical Society of America*, 104, 432–441. doi: 10.1121/1.423247
- Incerti, P. V., Ching, T. Y. C., & Cowan, R. (2013). A systematic review of electricacoustic stimulation: Device fitting ranges, outcomes, and clinical fitting practices. *Trends in Amplification*, 17, 3–26. doi: 10.1177/1084713813480857
- Kiefer, J., Pok, M., Adunka, O., Stürzebecher, E., Baumgartner, W., Schmidt, M., ... Gstoettner, W. (2005). Combined electric and acoustic stimulation of the auditory system: Results of a clinical study. *Audiology and Neurotology*, 10, 134–144. doi: 10.1159/000084023
- Kuk, F., Keenan, D., Korhonen, P., & Lau, C. (2009). Efficacy of linear frequency transposition on consonant identification in quiet and in noise. *Journal of the American Academy of Audiology*, 20, 465–479. doi: 10.3766/jaaa.20.8.2
- Kuk, F., Keenan, D., Peeters, H., Korhonen, P., & Auriemmo, J. (2008). 12 lessons learned about linear frequency transposition. *The Hearing Review*, 15(12), 32–41.
- Lee, A., Jiang, D., McLaren, S., Nunn, T., Demler, J. M., Tysome, J. R., . . . Fitzgerald O'Connor, A. (2010). Electric acoustic stimulation of the auditory system: Experience and results of ten patients using MED-EL's M and FlexEAS electrodes. *Clinical Otolaryngology*, *35*, 190–197. doi: 10.1111/j.1749-4486.2010.02140.x
- Lorens, A., Polak, M., Piotrowska, A., & Skarzynski, H. (2008). Outcomes of treatment of partial deafness with cochlear implantation: A DUET study. *The Laryngoscope*, *118*, 288–294. doi: 10.1097/MLG.0b013e3181598887
- McCreery, R. W., Venediktov, R. A., Coleman, J. J., & Leech, H. M. (2012). An evidencebased systematic review of frequency lowering in hearing aids for school-age children with hearing loss. *American Journal of Audiology*, *21*, 313–328. doi: 10.1044/1059-0889(2012/12-0015)
- Punte, A. K., Vermeire, K., & Van de Heyning, P. (2010). Bilateral electric acoustic stimulation: A comparison of partial and deep cochlear electrode insertion. A longitudinal case study. Advances in Oto-Rhino-Laryngology, 67, 144–152. doi: 10.1159/000262606
- Raffin, M. J. M., & Thornton, A. R. (1980). Confidence levels for differences between speech-discrimination scores. A research note. *Journal of Speech and Hearing Research*, 23, 5–18. doi: 10.1044/jshr.2301.05
- Rosner, B. (2006). Fundamentals of biostatistics (6th ed.). Belmont, CA: Thomson-Brooks/Cole.
- Sampaio, A. L. L., Araujo, M. F. S., & Oliveira, C. A. C. P. (2011). New criteria of indication and selection of patients to cochlear implant. *International Journal of Otolaryngology*, 2011(573968), 1–13. doi: 10.1155/2011/573968
- Scollie, S., Glista, D., Seto, J., Dunn, A., Schuett, B., Hawkins, M., ... Parsa, V. (2016). Fitting frequency-lowering signal processing applying the American Academy of Audiology pediatric amplification guideline: Updates and protocols. *Journal* of the American Academy of Audiology, 27, 219–236. doi:10.3766/jaaa.15059
- Scottish Intercollegiate Guidelines Network. (2017). Critical appraisal notes and checklists. Retrieved from http://www.sign.ac.uk/checklists-and-notes.html
- Simpson, A. (2009). Frequency-lowering devices for managing highfrequency hearing loss: A review. *Trends in Amplification, 13*, 87–106. doi: 10.1177/1084713809336421
- Simpson, A., McDermott, H. J., Dowell, R. C., Sucher, C., & Briggs, R. J. S. (2009). Comparison of two frequency-to-electrode maps for acousticelectric stimulation. *International Journal of Audiology*, 48, 63–73. doi: 10.1080/14992020802452184
- Skarzynski, H., & Lorens, A. (2010). Partial deafness treatment. Cochlear Implants International, 11(Suppl 1), 29–41. doi: 10.1179/146701010X12671178390799
- Skarzynski, H., Lorens, A., Matusiak, M., Porowski, M., Skarzynski, P. H., & James, C. J. (2012). Partial deafness treatment with the nucleus straight research array cochlear implant. *Audiology and Neurotology*, 17, 82–91. doi: 10.1159/000329366
- Souza, P. E., Arehart, K. H., Kates, J. M., Croghan, N. B., & Gehani, N. (2013). Exploring the limits of frequency lowering. *Journal of Speech, Language, and Hearing Research*, 56, 1349–1363. doi:10.1044/1092-4388(2013/12-0151)

- Talbot, K. N., & Hartley, D. E. H. (2008). Combined electro-acoustic stimulation: A beneficial union? *Clinical Otolaryngology*, 33, 536–545. doi: 10.1111/j.1749-4486.2008.01822.x
- Thornton, A. R., & Raffin, M. J. M. (1978). Speech-discrimination scores modeled as a binomial variable. *Journal of Speech and Hearing Research*, *21*, 507–518. doi: 10.1044/jshr.2103.507
- Usami, S., Moteki, H., Tsukada, K., Miyagawa, M., Nishio, S., Takumi, Y., . . . Tono, T. (2014). Hearing preservation and clinical outcome of 32 consecutive electric acoustic stimulation (EAS) surgeries. *Acta Oto-Laryngologica*, *134*, 717–727. doi: 10.3109/00016489.2014.894254
- Vickers, D., De Raeve, L., & Graham, J. (2016). International survey of cochlear implant candidacy. *Cochlear Implants International*, 17(Suppl. 1), 36–41. doi:10.1080/14 670100.2016.1155809
- von Ilberg, C. A., Baumann, U., Kiefer, J., Tillein, J., & Adunka, O. F. (2011). Electricacoustic stimulation of the auditory system: A review of the first decade. *Audiology and Neurotology*, 16(Suppl. 2), 1–30. doi:10.1159/000327765

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Declaration of Interest

The authors have no source of potential, real, or perceived conflicts of interest to declare.