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Association of Bone Conduction Devices for Single-Sided Sensorineural Deafness With Quality of Life
A Systematic Review and Meta-analysis

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Supplemental content

IMPORTANCE Although bone conduction devices (BCDs) have been shown to improve audiological outcomes of patients with single-sided sensorineural deafness (SSD), their effects on the patients' quality of life (QOL) are unclear.

OBJECTIVE To investigate the association of BCDs on QOL in patients with SSD.

DATA SOURCES Literature search of databases (Medline, Embase, Cochrane Library, and ClinicalTrials.gov) from January 1, 1978, to June 24, 2021, was performed.

STUDY SELECTION Prospective interventional studies with 10 or more participants with SSD (defined as pure tone average >70 dB hearing loss in the worse hearing ear and ≤30 dB in the better hearing ear) who underwent unilateral BCD implantation and assessment of QOL before and after the intervention using a validated tool were eligible for inclusion. Studies on adults and children were eligible for inclusion. Patients with only conductive, mixed, or bilateral hearing loss were excluded.

DATA EXTRACTION AND SYNTHESIS Data were extracted by 2 independent reviewers. Study clinical and demographic characteristics were obtained. Meta-analysis of mean differences in QOL scores before and after the intervention was performed. Study bias was assessed using Joanna Briggs Institute risk of bias tool.

MAIN OUTCOMES AND MEASURES The main study outcome was mean change in QOL scores at 6 months after insertion of BCDs. The 3 QOL instruments used in the studies included the Abbreviated Profile of Hearing Aid Benefit (APHAB), the Health Utilities Index–3 (HUI-3), and the Speech, Spatial and Qualities of Hearing Scale (SSQ). The APHAB and the SSQ are the hearing-related QOL measures, whereas the HUI-3 is a generic QOL measure.

RESULTS A total of 486 articles were identified, and 11 studies with 203 patients met the inclusion criteria. Only adult studies met inclusion criteria. Ten of 11 studies were nonrandomized cohort studies. The BCDs assessed were heterogeneous. There was a significant statistical and clinically meaningful improvement in the global APHAB scores (mean change, 15.50; 95% CI, 12.63-18.36; I^2 = 0) and the SSQ hearing qualities (mean change, 1.19; 95% CI, 0.46-1.92; I^2 = 78.4%), speech (mean change, 2.03; 95% CI, 1.68-2.37; I^2 = 0), and spatial hearing (mean change, 1.51; 95% CI, 0.57-2.44; I^2 = 81.1%) subscales. There was no significant change detected in the mean HUI-3 scores (mean change, 0.03; 95% CI, −0.04 to 0.10; I^2 = 0). The risk of bias was assessed to be low to moderate.

CONCLUSIONS AND RELEVANCE These findings suggest that adult patients who receive BCDs may experience improvements in hearing-specific QOL measures but not in generic QOL measures. Prospective QOL studies should be considered in this cohort, particularly for children with SSD.

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Bone conduction devices (BCDs) are an established form of treatment for conductive hearing loss or single-sided sensorineural deafness (SSD). The BCDs work by converting sound energy to vibration of the skull bones, which results in a wave in the basilar membrane of the cochlea similar to that produced through air conduction of sound.[1] It is well known that BCDs improve hearing outcomes in patients with SSD, via the principle of rerouting sound from the affected side of the head to the contralateral normally hearing ear.[2-4] To our knowledge, the effects of BCDs on quality of life (QOL) have been less well evaluated. Studies have explored individual nonhearing benefits of BCDs in SSD without demonstrating improvement.[5] In literature reviews evaluating the association of BCDs with overall QOL in patients with unilateral and bilateral hearing loss,[2,6] many studies are underpowered or show only modest benefits (reporting safety and "satisfaction"), meaning it remains unclear from these publications whether patients with SSD experience a meaningful QOL benefit.

Hearing is integral to many aspects of human life and contributes to speech, cognition, communication, work, socializing, and entertainment. It therefore appears logical that an improvement in hearing should result in better QOL. Single-sided sensorineural deafness has been shown to be associated with decreased QOL compared with general population mean QOL scores.[7] Bone conduction devices have previously been used in patients with SSD with good audiological outcomes[3]; therefore, one would assume that improving hearing with BCDs would result in an improvement in QOL. However, improvements in physical health do not universally result in an improvement in QOL, as prior studies have suggested.[8] To interrogate this hypothesis further, we conducted a meta-analysis of prospective interventional studies that included a QOL measure for patients with SSD treated with a unilateral BCD implantation.

Key Points

Question What measures are used to assess quality of life (QOL) after bone conduction device implantation in patients with single-sided sensorineural deafness, and how did QOL change in these patients?

Findings This systematic review and meta-analysis of 11 studies with 203 adult patients searched studies that assessed QOL and found hearing-specific QOL assessed with the Abbreviated Profile of Hearing Aid Benefit (APHAB) and the Speech, Spatial and Qualities of Hearing Scale (SSQ) and generic QOL with the Health Utilities Index–3 (HUI-3). Significant improvements were found in the global APHAB and SSQ but not the HUI-3.

Meaning These findings suggest that the APHAB and the SSQ may be more sensitive to detecting the changes produced by hearing interventions compared with the HUI-3.

Methods

Before commencement of this review, we were not aware of a single tool that was accepted as the universal standard for measurement of QOL in patients with hearing loss. We included studies of both adult and pediatric populations and reviewed the results of generic and disease-specific measures of QOL. Generic measures consisted of tools that can be used for any population, whereas disease-specific measures have been validated for patients with a particular disease or condition.[9]

Search Strategy

A literature search of databases including Medline, Embase, Cochrane Library, and ClinicalTrials.gov from January 1, 1978, to June 24, 2021, was performed via the Healthcare Databases Advanced Search tool. The references were screened by 2 independent reviewers (T.H. and K.M.), and any disagreements were adjudicated by a reviewer (S.S.). The full strategy is provided in the eMethods in the Supplement. The reference lists of the selected articles were manually reviewed to locate additional studies. The systematic review and meta-analysis were conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) and Meta-analysis of Observational Studies in Epidemiology (MOOSE) guidelines. Systematic searches of gray literature (outside commercial publishing) were assessed using bibliographic references of our included studies. No additional gray sources were included.[10]

Study Eligibility

Prospective interventional studies with 10 or more participants with SSD who underwent implantation of a unilateral BCD and preintervention and postintervention assessment of QOL using a validated tool were eligible for inclusion. Single-sided sensorineural deafness was defined as pure tone average of more than 70 dB hearing loss in the worse hearing ear and 30 dB or less in the better hearing ear.[11] Studies on adults and children were eligible for inclusion. To our knowledge, no validated QOL measures are specifically designed to measure QOL after interventions for patients with SSD. Therefore, we included studies that reported general and hearing-specific QOL instruments previously validated in patients with hearing loss. Eligible BCDs were considered to be those worn temporarily (eg, soft-band) and intraoral and surgically implantable devices (eg, bone-anchored hearing aids).

Exclusion

Patients with mixed, conductive, or bilateral hearing loss were excluded. Studies that used patient-reported outcome measures not designed to measure QOL, such as patient satisfaction, were excluded. Studies that failed to report the preintervention and postintervention QOL scores or the change in mean scores were excluded. Meaningful changes in instrument scores are discussed in the Results section. Articles published in a language other than English were excluded. The main study outcome was the mean change in QOL score after BCD implantation at 6 months.

Data Extraction

Two authors (T.H. and K.M.) independently extracted the study data. The following information was collected: name of the first author, publication year, sample size, country, device type, QOL instrument, preintervention and postintervention QOL mean scores with SD or mean difference, follow-up period (months), patient population, sex, ethnicity, QOL tool in original language (ie, if a translated questionnaire, whether this was a validated translation of the QOL instrument in the language of the population being assessed), socioeconomic status (any record of deprivation, occupation, poverty, income, family size, or other measure), educational outcomes (any record of assessment of educational attainments or performance after the intervention), speech outcomes (any record of formal assessment of speech perception or language assessment after the intervention), and ethical approval. Where data were not readily available in the report, study authors were contacted via email and were given 2 weeks to respond. Two of the 9 authors who were contacted responded in the allocated time frame, and 1 of these authors was able to provide raw data that were included in our analysis.

Data Synthesis and Meta-analysis

Mean differences and SDs were extracted from individual studies, and a meta-analysis of the mean change in QOL scores was performed using the OpenMeta(Analyst) software, version 10.10 (Brown University). Where mean differences were not reported, a change-from-baseline SD was imputed using preintervention and postintervention means with SDs and a correlation coefficient of 0.59 to calculate mean differences.

This correlation coefficient is based on the experimental study by Balk et al,[12] validating the method of handling missing mean differences when performing meta-analyses. As per the Cochrane handbook, experimental analyses were performed with the correlation coefficients of 0.30 and 0.80 to confirm that no significant differences were found.[13]

Separate meta-analyses were performed for individual QOL measures. Statistical heterogeneity was determined by Freeman-Tukey transformation and assuming random effects as described by DerSimonian and Laird.[14] Random-effects model meta-analysis was performed if high heterogeneity defined as $I^2 < 50.0\%$ was found. Alternatively, fixed-effects model meta-analysis was used.

Assessment of Bias

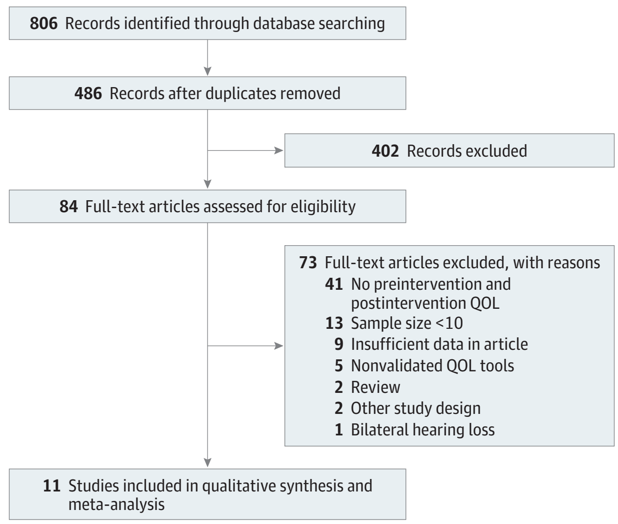
The risk of bias was assessed using the Joanna Briggs Institute (JBI) risk of bias tool for quasi-experimental studies.[15] The JBI tool does not state specific scores at which studies should no longer be included, and we believe that without all but the most significant concerns, it is appropriate to include all the studies while accounting for potential risk of bias in those with lower scores. The assessment of bias was performed independently by 2 reviewers (T.H. and E.W.). Any disagreements were adjudicated by the senior reviewer (S.S.).

Results

Literature Search

The results of the search strategy are summarized in the PRISMA flowchart (**Figure 1**). A total of 486 articles were identified. After the screening of titles and abstracts, 84 articles were retrieved, and full manuscript review was performed. After applying inclusion and exclusion criteria, the final 11 studies were selected to be included in the meta-analysis.[16-26] Ten studies[16-19,21-26] were prospective, nonrandomized, preintervention and postintervention studies. One study was a randomized clinical trial[20] that included a treatment arm with patients undergoing QOL assessment before and after insertion of BCDs.

Figure 1. PRISMA Flow Diagram



QOL indicates quality of life.

Study Characteristics

The summary of the 11 included studies is provided in the **Table**. All studies were performed in adult populations in North America, Europe, Australia, and Korea. A total of 203 adult patients underwent preintervention and postintervention QOL assessments. In 10 studies,[16-23,25,26] surgically implanted BCDs were inserted (3 transcutaneous and 6 percutaneous), whereas in 1 additional study,[24] a dental conduction device was used.

Sociodemographic Characteristics of the Studies

Additional sociodemographic and secondary variables were prospectively chosen for analysis. Six of 9 studies[16-18,23,25,26] that document sex differentiate between hearing etiology sufficiently so that we know the sex of those patients with SSD. None of the studies recorded patient ethnicity. None of the studies reported background educational or long-term speech outcomes outside the QOL instruments. Only 4 studies[18,23,25,26] reported using QOL instruments originally produced in the language of the patient, but 1 additional study[16] described the use of a validated translation. Only 5 studies[17,19-21,25] reported ethical approval measures in the body of the paper.

Risk of Bias

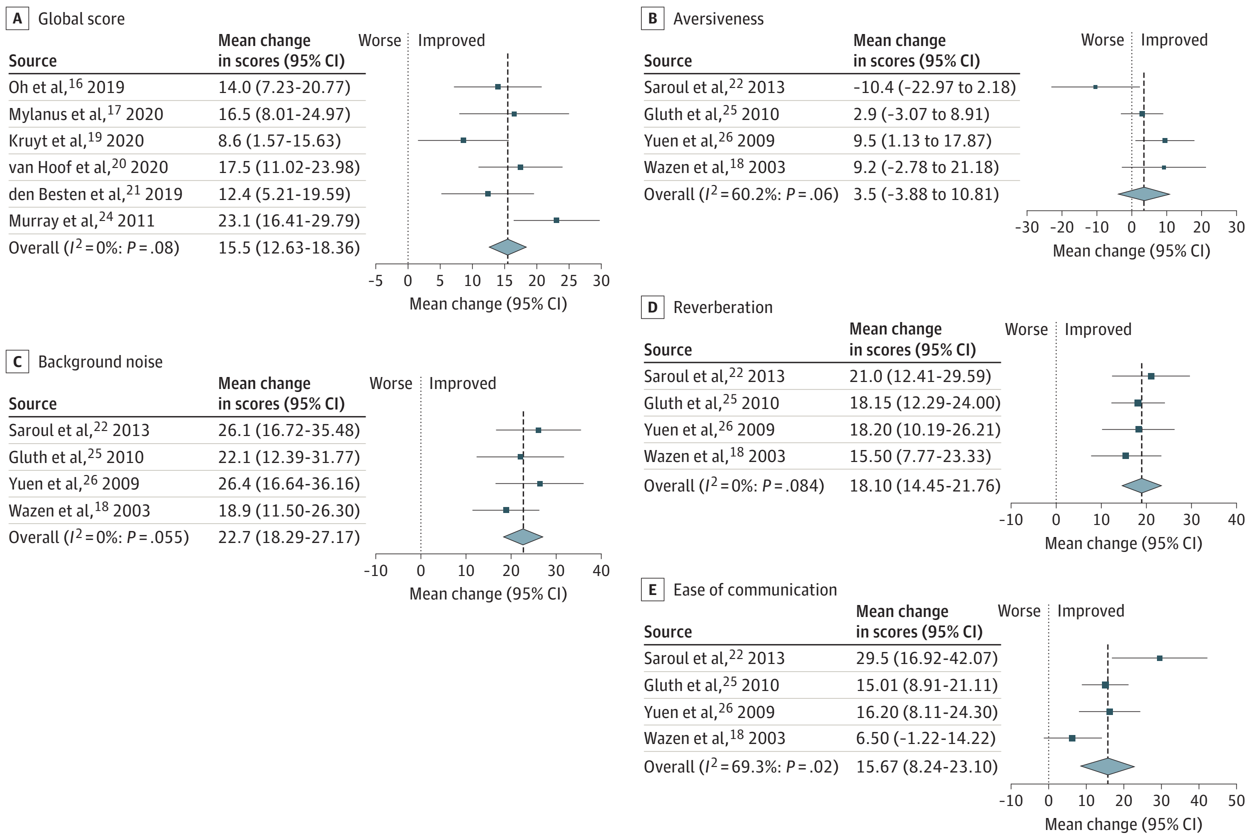
Risk of bias scores using the JBI tool ranged from 5 to 8, with higher scores indicating lower risk of bias (the highest possible score is 9). The median score was 6. We interpret this as a mild to moderate risk of bias for the studies included herein.

Table. Summary of Included Studies

Source (location)	Sample size	Device	QOL measures	Follow-up, mo	Sex reported	Ethnicity (self-reported)	PROM/QALY in original language	Country-specific language used	SES reported	Educational outcomes reported	Speech outcomes reported	Ethics permission locally
Oh et al,[16] 2019 (Korea [single center])	22	Bonebridge (Med-El Corporation) (percutaneous)	APHAB	6	Yes	No	No	Yes	No	No	No	No
Mylanus et al,[17] 2020 (Europe, Australia, and US [multicenter])	14	Cochlear Osia (Cochlear Bone Anchored Solutions AB) (transcutaneous)	HUI-3, APHAB, and SSQ	12	Yes	No	NR	NR	No	No	No	Yes
Kruyt et al,[19] 2020 (Europe and US [multicenter])	15	Cochlear Baha Attract (Cochlear Bone Anchored Solutions AB) (transcutaneous)	HUI-3, APHAB, and SSQ	24	Yes	No	NR	NR	No	No	No	Yes
van Hoof et al,[20] 2020 (Europe [multicenter])	15	Cochlear Baha (Cochlear Bone Anchored Solutions AB) (percutaneous)	AHPAB and HUI-3	12	Yes	No	NR	NR	No	No	No	Yes
den Besten et al,[21] 2019 (Europe [multicenter])	15	Baha Attract (Cochlear Bone Anchored Solutions AB) (transcutaneous)	HUI-3, APHAB, and SSQ	6	Yes	No	NR	NR	No	No	No	Yes
Saroul et al,[22] 2013 (France [single center])	20	Cochlear Baha Divino (Cochlear Bone Anchored Solutions AB) (percutaneous)	APHAB	24	No	No	NR	NR	No	No	No	No
Pai et al,[23] 2012 (UK [single center])	25	NR	SSQ	6	Yes	No	Yes	NA	No	No	No	NR
Murray et al,[24] 2011 (US [multicenter])	22	SoundBite (Sonitus Medical, Inc) (dental)	APHAB	6	No	No	NR	NR	No	No	No	No
Gluth et al,[25] 2010 (Australia [single center])	21	Compact, Divino (percutaneous)	APHAB	3	Yes	No	Yes	NA	No	No	No	Yes
Yuen et al,[26] 2009 (Canada [single center]	16	Divino (percutaneous)	APHAB	3	Yes	No	Yes	NR	NR	NR	No	No
Wazen et al,[18] 2003 (US [multicenter])	18	Divino, Intenso (percutaneous)	APHAB	12	Yes	No	Yes	NA	No	No	No	No

Abbreviations: APHAB, Abbreviated Profile of Hearing Aid Benefit; HUI-3, Health Utilities Index–3; NA, not applicable; NR, not reported; PROM, patient-reported outcome measure; QALY, quality-adjusted life-year; QOL, quality of life; SES, socioeconomic status; SSQ, Speech, Spatial and Qualities of Hearing Scale.

Figure 2. Forest Plot of Meta-analyses for Studies Assessing Abbreviated Profile of Hearing Aid Benefit



Mean change is given for the global score and the 4 subscales. Different marker sizes indicate the different relative sizes of 95% CIs. Point estimates reflect the following: A, level of overall disability associated with hearing impairment; B, unpleasantness/tolerability of environmental sounds; C, communication with high background noise; D, communication in reverberant rooms (eg, a classroom); E, communication under favorable conditions.

QOL Instruments

Only 3 QOL instruments were used in the analyzed studies: the Abbreviated Profile of Hearing Aid Benefit (APHAB),[27] the Health Utilities Index-3 (HUI-3),[28] and the Speech, Spatial and Qualities of Hearing Scale (SSQ).[29] In brief, the APHAB is a 24-item inventory that assesses the amount of difficulty that a person experiences when communicating in a variety of conditions. The APHAB provides a global score as well as subscale scores for the ease of communication, reverberation, background noise, and aversiveness domains. The HUI-3 is a health status measure based on 8 attributes: vision, hearing, speech, walking, dexterity, emotion, cognition, and pain. A comprehensive health state score is calculated using the scores from all these attributes. The SSQ is a 49-item questionnaire that measures disability associated with hearing and provides scores across 3 subscales: speech recognition (in a variety of contexts), spatial hearing (segregation, direction, distance, and movement of sound), and hearing qualities (ease of listening, naturalness, and clarity).

Clinically Meaningful Difference

Changes in scores that are statistically significant may not correlate with significant changes in patient experience. Although studies rarely agree on these measures, the following guidance is suggested for the QOL instruments included in the analysis:

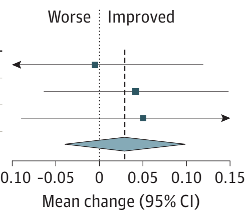
- APHAB: for individual subscales, a difference of 10% between unaided and aided scores allows reasonable certainty that the change in scores represents a real difference between conditions[30];
- HUI-3: the smallest difference in utility scores between levels of an HUI attribute is 0.05[31]; and
- SSQ: research suggests variable response rates for age, sex, educational attainment, and general health, but self-reporting of hearing difficulties was the single most influential factor in score changes leading to decreases of 0.7 to 1.3 points; this was most pronounced in the speech recognition subscale, with 0.5 points correlating with approximately 10 dB of hearing loss on pure tone average.[32]

Meta-analysis of APHAB Global and Subscale Scores

There was a significant improvement in the global APHAB scores (mean change, 15.50; 95% CI, 12.63-18.36; I^2 = 0; P = .08) found based on the data from 6 studies[16,17,19,20,22,24] with 98 patients (**Figure 2**A). Similarly, significant improvements were observed in the background noise (mean change, 22.73; 95% CI, 18.29-27.17; I^2 = 0), reverberation (mean change, 18.10; 95% CI, 14.45-21.76; I^2 = 0), and ease of communication (mean

Figure 3. Forest Plot of Meta-analyses for Studies Assessing Health Utilities Index–3

Source	Mean change in scores (95% CI)
Kruyt et al,[19] 2020	-0.005 (-0.13 to 0.12)
van Hoof et al,[20] 2020	0.042 (-0.06 to 0.15)
den Besten et al,[21] 2019	0.050 (-0.09 to 0.19)
Overall (I^2 = 0%: P = .81)	0.029 (-0.04 to 0.10)



Different marker sizes indicate the different relative sizes of 95% CIs. Point estimates reflect the degree of impairment or disability.

change, 15.67; 95% CI, 8.24-23.10; I^2 = 0) subscales but not the aversiveness subscale (mean change, 3.46; 95% CI, −3.88 to 10.81; I^2 = 60.2%) (Figure 2B-E). Heterogeneity was low in all analyses except for aversiveness subscale.

Meta-analysis of HUI-3 Comprehensive Scores

Data on the mean changes measured by HUI-3 comprehensive status were available from 3 studies[19-21] with 45 patients. No significant change was detected in mean scores (overall mean change, 0.03; 95% CI, −0.04 to 0.10) (**Figure 3**). Statistical heterogeneity was found to be low (I^2 = 0; P = .81).

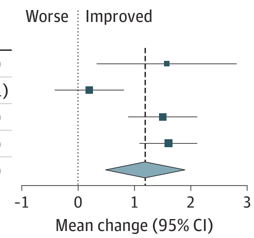
Meta-analysis of SSQ Subscales

Four studies[17,19,21,23] including 69 patients evaluated QOL using the SSQ. Significant improvements in the mean difference were observed across hearing qualities (mean change, 1.19; 95% CI, 0.46-1.92), speech recognition (overall mean change, 2.03; 95% CI, 1.68-2.37), and spatial hearing (overall mean change, 1.51; 95% CI, 0.57-2.44) subscales (**Figure 4**). Statistical heterogeneity was high for the hearing qualities (I^2 = 78.4%; P = .003) and spatial hearing subscales (I^2 = 81.1%; P = .001), but low for the speech recognition subscale (I^2 = 0; P = .054).

Figure 4. Forest Plot of Meta-analyses for Studies Assessing Speech, Spatial and Qualities of Hearing Scale

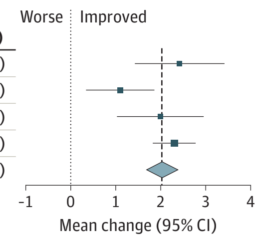
A Other qualities

Source	Mean change in scores (95% CI)
Mylanus et al,[17] 2020	1.6 (0.33 to 2.81)
Kruyt et al,[19] 2020	0.2 (-0.41 to 0.81)
den Besten et al,[21] 2019	1.5 (0.89 to 2.11)
Pai et al,[23] 2012	1.6 (1.09 to 2.11)
Overall (I^2 = 78.4%: P = .003)	1.2 (1.46 to 1.92)



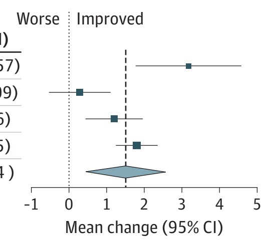
B Speech

Source	Mean change in scores (95% CI)
Mylanus et al,[17] 2020	2.4 (1.43 to 3.41)
Kruyt et al,[19] 2020	1.1 (0.34 to 1.86)
den Besten et al,[21] 2019	2.0 (1.03 to 2.95)
Pai et al,[23] 2012	2.3 (1.83 to 2.77)
Overall (I^2 = 0%: P = .054)	2.0 (1.68 to 2.37)



C Spatial

Source	Mean change in scores (95% CI)
Mylanus et al,[17] 2020	3.2 (1.793 to 4.57)
Kruyt et al,[19] 2020	0.3 (-0.53 to 1.09)
den Besten et al,[21] 2019	1.2 (0.44 to 1.96)
Pai et al,[23] 2012	1.8 (1.25 to 2.35)
Overall (I^2 = 81.1%: P = .001)	1.5 (0.57 to 2.44)



Different marker sizes indicate the different relative sizes of 95% CIs. Point estimates reflect the following: A, auditory disability; B, speech recognition disability; C, spatial hearing disability.

Discussion

This review found that across 203 patients and 11 studies, generic QOL scores did not improve but hearing-specific QOL measures improved for patients treated with BCDs for SSD. This finding adds to current developments in our understanding of the benefit of treatment for asymmetrical hearing loss and greater understanding that even a single hearing ear may still leave patients with significant morbidity.[33]

One advantage of generic vs disease-specific QOL questionnaires and instruments is the improved ability to compare QOL across populations and different disease or health states when making decisions concerning health economics or health care delivery.[34] Our review suggests that the disease-specific QOL instruments were generally associated with greater sensitivity for detecting and demonstrating small changes in our patients' day-to-day lives, and some specific tools are explicitly designed to measure changes in QOL over time or with treatment, but this leaves us with 2 possible conclusions. Either hearing-specific instruments are overly sensitive to changes and hearing does not have the significant impact on overall QOL that we appreciated, or the generic instruments are too blunt to appreciate the genuine burden of SSD.

The generic QOL tool included in this meta-analysis was the HUI-3, which includes domains focused on speech and hearing. This instrument nonetheless failed to show significant improvement. Whether other generic tools would show similar results remains to be seen. The UK National Institute for Health and Care Excellence (NICE) 2012 guidance[35] for assessment of cost-utility advises the use of estimated quality-adjusted life-years gained as a preferential measure of health effects when considering allocation and distribution of health care resources. If the benefits patients receive from BCDs are not evident on generic QOL measurement, then important implications for future health technology assessment and cost-effectiveness analysis follow for hearing health interventions.

The EuroQol 5 dimensions (EQ-5D-5L) is the world's most commonly used QOL instrument,[36] and in the UK, NICE prefers the use of this metric specifically for comparison between disease states.[34] The EQ-5D-5L has been shown not to produce statistically significant changes before and after hearing interventions, with very low effect sizes where reported, but it does not include any speech- or hearing-specific metrics.[37] The EQ-5D-5L includes a visual analog scale score that some studies have found better demonstrates improvements in patients with hearing impairments,[37] and where the EQ-5D-5L has suggested benefit in hearing health, it has shown improvements largely associated with the anxiety/depression dimension, which may mean that further research into the psychosocial dimensions of SSD could further improve our understanding of its true impact.[36-38]

The separate meta-analysis of each QOL tool is a strength of this review. Although guides exist describing the benefits and interpretational challenges of using standard mean difference to estimate pooled size effects from different QOL instruments,[39] our underlying assumption is that such methods introduce significant heterogeneity, given that different scales rarely measure the same constructs. Effects should only be pooled if individual questions can be determined to measure similar constructs in the same directions (ie, 2 questions about anxiety where a higher score indicates worse anxiety in both instruments[40]).

This review found that the use of QOL instruments in otolaryngology and audiology research in this area was heterogenous. Not all instruments were used before and after the intervention, and some instruments claim to be specifically designed for post hoc analysis. We advocate the dual use of generic and disease-specific QOL tools, both before and after intervention, whenever a thorough genuine assessment of QOL is the intention of the research team.[41]

Limitations

It is known from previous studies that a plethora of outcomes have been used in the field of SSD, particularly with QOL measures and instruments, with 1 study[42] finding 344 unique outcome instruments that reported 520 outcome domains. Work to achieve consensus on a unified core outcome set of measures derived from all relevant stakeholders is admirable.[43] Nonetheless, our review was limited not just by the heterogeneity of QOL measures in included studies but also by the nonrandomized and single-time point nature of many study designs. The interventions offered were also disparate, with a variety of implants, manufacturers, and operative sites (sometimes with multiple devices in a single study), not to mention potential differences in performance of sound processors. We pooled all devices for our analysis. However, the devices assessed were heterogeneous. Three studies[17,19,21] looked at transcutaneous (no external abutment visible) rather than percutaneous surgical devices, and these are likely to have different outcomes not just in sound quality but also in wound healing, aftercare, and final cosmetic appearance, which could have a significant effect on QOL. Our sociodemographic findings revealed inconsistencies in multiple other reporting domains. There were no pediatric studies that fulfilled our criteria, and given the potential for children to benefit through the life course, we advocate for more QOL assessments in children with SSD. Reporting of sex differences was not consistent. Race and ethnicity were not addressed. Absent speech outcomes may have been reported in other studies that did not assess QOL, but comparisons between QOL and other measures could reveal more about how BCDs improve QOL when they are effective. Finally, there are implications for the use of these instruments outside their intended populations. Just as the use of QOL measures that are largely developed originally in adult patients has implications for their interpretation and validity when used in children (we would advocate the use of Child Health Utility 9D questionnaire[44]), translation and validation are needed when tools are used in other countries, particularly the development of country-specific value sets to enable quality-adjusted life-years to be derived and health economic evaluation to be more meaningful.

Conclusions

This systematic review and meta-analysis found that BCDs are associated with significant improvements in hearing-related QOL as measured by APHAB and SSQ scores in adult patients, whereas no difference was found in the measures of generic QOL. An instrument derived from a core outcome set sensitive to hearing interventions of all types and indications would allow more meaningful comparisons of interventions. These findings have important implications for future trials and studies on health economic evaluation of BCDs in SSD. Finally, well-designed prospective studies of a range of devices in a pediatric population with SSD are urgently needed.

ARTICLE INFORMATION

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Author Contributions: Mr Hampton had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. Messrs Hampton and Milinis contributed equally as co–first authors.
Concept and design: Hampton, Milinis, Sharma.
Acquisition, analysis, or interpretation of data: All authors.
Drafting of the manuscript: All authors.
Critical revision of the manuscript for important intellectual content: All authors.
Statistical analysis: Hampton, Milinis, Sharma.
Obtained funding: Hampton.
Administrative, technical, or material support: Milinis, Sharma.
Supervision: Sharma.

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Additional Information: The level of evidence (modified from the Oxford Centre for Evidence-based Medicine for ratings of individual studies) for this meta-analysis is 4.

REFERENCES

1. Stenfelt S. Acoustic and physiologic aspects of bone conduction hearing. *Adv Otorhinolaryngol*. 2011;71:10-21. doi:10.1159/000323574

2. Magele A, Schoerg P, Stanek B, Gradl B, Sprinzl GM. Active transcutaneous bone conduction hearing implants: systematic review and meta-analysis. *PLoS One*. 2019;14(9):e0221484. doi:10.1371/journal.pone.0221484

3. Kitterick PT, Smith SN, Lucas L. Hearing instruments for unilateral severe-to-profound sensorineural hearing loss in adults: a systematic review and meta-analysis. *Ear Hear*. 2016;37(5): 495-507. doi:10.1097/AUD.0000000000000313

4. Liu CC, Livingstone D, Yunker WK. The role of bone conduction hearing aids in congenital unilateral hearing loss: a systematic review. *Int J*

Pediatr Otorhinolaryngol. 2017;94:45-51. doi:10.1016/j.ijporl.2017.01.003

5. Wazen JJ, Ghossaini SN, Spitzer JB, Kuller M. Localization by unilateral BAHA users. *Otolaryngol Head Neck Surg*. 2005;132(6):928-932. doi:10.1016/j.otohns.2005.03.014

6. Brodie A, Smith B, Ray J. The impact of rehabilitation on quality of life after hearing loss: a systematic review. *Eur Arch Otorhinolaryngol*. 2018;275(10):2435-2440. doi:10.1007/s00405-018-5100-7

7. Sano H, Okamoto M, Ohhashi K, Iwasaki S, Ogawa K. Quality of life reported by patients with idiopathic sudden sensorineural hearing loss. *Otol Neurotol*. 2013;34(1):36-40. doi:10.1097/MAO.0b013e318278540e

8. Sfeatcu R, Cernuşcă-Miţariu M, Ionescu C, et al. The concept of wellbeing in relation to health and quality of life. 2014;10(4):123-128. Accessed May 19, 2021. http://www.ejst.tuiasi.ro/Files/46/12_Sfeatcu%20et%20al.pdf

9. McRackan TR, Bauschard M, Hatch JL, et al. Meta-analysis of cochlear implantation outcomes evaluated with general health-related patient-reported outcome measures. *Otol Neurotol*. 2018;39(1):29-36. doi:10.1097/MAO.0000000000001620

10. Aromataris E, Munn Z, eds. *JBI Manual for Evidence Synthesis*. JBI; 2020. doi:10.46658/JBIMES-20-01

11. van Zon A, Peters JPM, Stegeman I, Smit AL, Grolman W. Cochlear implantation for patients with single-sided deafness or asymmetrical hearing loss: a systematic review of the evidence. *Otol Neurotol*. 2015;36(2):209-219. doi:10.1097/MAO.0000000000000681

12. Balk EM, Earley A, Patel K, et al. Empirical assessment of within-arm correlation imputation in trials of continuous outcomes [Internet]. 2012. Accessed June 13, 2021. https://www.ncbi.nlm.nih.gov/books/NBK115799/

13. Higgins J, Thomas J, eds. Cochrane handbook for systematic reviews of interventions. Cochrane Training. Version 6.2. Published 2021. Accessed June 13, 2021. https://training.cochrane.org/handbook/current

14. DerSimonian R, Laird N. Meta-analysis in clinical trials. *Control Clin Trials*. 1986;7(3):177-188. doi:10.1016/0197-2456(86)90046-2

15. Joanna Briggs Institute. Checklist for quasi-experimental studies (non-randomized experimental studies). 2020. Accessed June 13, 2021. https://jbi.global/critical-appraisal-tools

16. Oh SJ, Goh EK, Choi SW, et al. Audiologic, surgical and subjective outcomes of active transcutaneous bone conduction implant system (Bonebridge). *Int J Audiol*. 2019;58(12):956-963. doi:10.1080/14992027.2019.1657242

17. Mylanus EAM, Hua H, Wigren S, et al. Multicenter clinical investigation of a new active osseointegrated steady-state implant system. *Otol Neurotol*. 2020;41(9):1249-1257. doi:10.1097/MAO.0000000000002794

18. Wazen JJ, Spitzer JB, Ghossaini SN, et al. Transcranial contralateral cochlear stimulation in unilateral deafness. *Otolaryngol Head Neck Surg*. 2003;129(3):248-254. doi:10.1016/S0194-5998(03)00527-8

19. Kruyt IJ, Monksfield P, Skarzynski PH, et al. Results of a 2-year prospective multicenter study evaluating long-term audiological and clinical outcomes of a transcutaneous implant for bone conduction hearing. *Otol Neurotol*. 2020;41(7):901-911. doi:10.1097/MAO.0000000000002689

20. van Hoof M, Wigren S, Ivarsson Blechert J, et al. A multinational cost-consequence analysis of a bone conduction hearing implant system: a randomized trial of a conventional vs a less invasive treatment with new abutment technology. *Front Neurol*. 2020;11:106. doi:10.3389/fneur.2020.00106

21. den Besten CA, Monksfield P, Bosman A, et al. Audiological and clinical outcomes of a transcutaneous bone conduction hearing implant: six-month results from a multicentre study. *Clin Otolaryngol*. 2019;44(2):144-157. doi:10.1111/coa.13248

22. Saroul N, Akkari M, Pavier Y, Gilain L, Mom T. Long-term benefit and sound localization in patients with single-sided deafness rehabilitated with an osseointegrated bone-conduction device. *Otol Neurotol*. 2013;34(1):111-114. doi:10.1097/MAO.0b013e31827a2020

23. Pai I, Kelleher C, Nunn T, et al. Outcome of bone-anchored hearing aids for single-sided deafness: a prospective study. *Acta Otolaryngol*. 2012;132(7):751-755. doi:10.3109/00016489.2012.655862

24. Murray M, Miller R, Hujoel P, Popelka GR. Long-term safety and benefit of a new intraoral device for single-sided deafness. *Otol Neurotol*. 2011;32(8):1262-1269. doi:10.1097/MAO.0b013e31822a1cac

25. Gluth MB, Eager KM, Eikelboom RH, Atlas MD. Long-term benefit perception, complications, and device malfunction rate of bone-anchored hearing aid implantation for profound unilateral sensorineural hearing loss. *Otol Neurotol*. 2010;31(9):1427-1434. doi:10.1097/MAO.0b013e3181f0c53e

26. Yuen HW, Bodmer D, Smilsky K, Nedzelski JM, Chen JM. Management of single-sided deafness with the bone-anchored hearing aid. *Otolaryngol Head Neck Surg*. 2009;141(1):16-23. doi:10.1016/j.otohns.2009.02.029

27. Cox RM, Alexander GC. The abbreviated profile of hearing aid benefit: ear and hearing. *Ear Hearing*. 1995;16(2):176-186. Accessed June 13, 2021. https://journals.lww.com/ear-hearing/Abstract/1995/04000/The_Abbreviated_Profile_of_Hearing_Aid_Benefit.5.aspx

28. Horsman J, Furlong W, Feeny D, Torrance G. The Health Utilities Index (HUI): concepts, measurement properties and applications. *Health Qual Life Outcomes*. 2003;1(1):54. doi:10.1186/1477-7525-1-54

29. Gatehouse S, Noble W. The Speech, Spatial and Qualities of Hearing Scale (SSQ). *Int J Audiol*. 2004;43(2):85-99. doi:10.1080/14992020400050014

30. Cox RM. Administration and application of the APHAB. *Hearing J*. 1997;50(4):32, 35-36, 38, 40, 41, 44, 45, 48. Accessed September 15, 2021. https://journals.lww.com/thehearingjournal/citation/1997/04000/administration_and_application_of_the_aphab.2.aspx

31. Horsman J, Furlong W, Feeny D, Torrance G. The Health Utilities Index (HUI): concepts, measurement properties and applications. *Health Qual Life Outcomes*. 2003;1:54. doi:10.1186/1477-7525-1-54

32. von Gablenz P, Otto-Sobotka F, Holube I. Adjusting expectations: hearing abilities in a population-based sample using an SSQ short form. *Trends Hear*. 2018;22:2331216518784837. doi:10.1177/2331216518784837

33. Snapp HA, Ausili SA. Hearing with one ear: consequences and treatments for profound unilateral hearing loss. *J Clin Med*. 2020;9(4):1010. doi:10.3390/jcm9041010

34. National Institute for Health and Care Excellence. Guide to the methods of technology appraisal 2013. April 4, 2013. Accessed September 15, 2021. https://www.nice.org.uk/process/pmg9/resources/guide-to-the-methods-of-technology-appraisal-2013-pdf-2007975843781

35. National Institute for Health and Care Excellence. Methods for the development of NICE public health guidance (3rd edition). September 26, 2012. Accessed September 15, 2021. https://www.nice.org.uk/process/pmg4/resources/methods-for-the-development-of-nice-public-health-guidance-third-edition-pdf-2007967445701

36. McCaffrey N, Kaambwa B, Currow DC, Ratcliffe J. Health-related quality of life measured using the EQ-5D-5L: South Australian population norms. *Health Qual Life Outcomes*. 2016;14(1):133. doi:10.1186/s12955-016-0537-0

37. Yang Y, Longworth L, Brazier J. An assessment of validity and responsiveness of generic measures of health-related quality of life in hearing impairment. *Qual Life Res*. 2013;22(10):2813-2828. doi:10.1007/s11136-013-0417-6

38. Summerfield AQ, Barton GR; UK Cochlear Implant Study Group. Sensitivity of EQ-5D-3L, HUI2, HUI3, and SF-6D to changes in speech reception and tinnitus associated with cochlear implantation. *Qual Life Res*. 2019;28(5):1145-1154. doi:10.1007/s11136-018-2070-6

39. Thorlund K, Walter SD, Johnston BC, Furukawa TA, Guyatt GH. Pooling health-related quality of life outcomes in meta-analysis-a tutorial and review of methods for enhancing interpretability. *Res Synth Methods*. 2011;2(3):188-203. doi:10.1002/jrsm.46

40. Murad MH, Wang Z, Chu H, Lin L. When continuous outcomes are measured using different scales: guide for meta-analysis and interpretation. *BMJ*. 2019;364:k4817. doi:10.1136/bmj.k4817

41. Wells GA, Russell AS, Haraoui B, Bissonnette R, Ware CF. Validity of quality of life measurement tools—from generic to disease-specific. *J Rheumatol Suppl*. 2011;88(suppl 88):2-6. doi:10.3899/jrheum.110906

42. Katiri R, Hall DA, Killan CF, Smith S, Prayuenyong P, Kitterick PT. Systematic review of outcome domains and instruments used in designs of clinical trials for interventions that seek to restore bilateral and binaural hearing in adults with unilateral severe to profound sensorineural hearing loss ('single-sided deafness'). *Trials*. 2021;22:220. doi:10.1186/s13063-021-05160-5

43. Katiri R, Hall DA, Buggy N, et al. Core Rehabilitation Outcome Set for Single Sided Deafness (CROSSSD) study: protocol for an international consensus on outcome measures for single sided deafness interventions using a modified Delphi survey. *Trials*. 2020;21(1):238. doi:10.1186/s13063-020-4094-9

44. The University of Sheffield School of Health and Related Research. Measuring and valuing health: CHU9D (paediatric quality of life). Published 2021. Accessed June 13, 2021. https://www.sheffield.ac.uk/scharr/research/themes/valuing-health#CHU-9D