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Bone-anchored hearing aids (BAHAs) for people who are bilaterally deaf: a systematic review and economic evaluation

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Abstract

Bone-anchored hearing aids (BAHAs) for people who are bilaterally deaf: a systematic review and economic evaluation

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Background: A bone-anchored hearing aid (BAHA) consists of a permanent titanium fixture, which is surgically implanted into the skull bone behind the ear, and a small detachable sound processor that clips onto the fixture. BAHAs are suitable for people with conductive or mixed hearing loss who cannot benefit fully from conventional hearing aids.
Objectives: To assess the clinical effectiveness and cost-effectiveness of BAHAs for people who are bilaterally deaf.
Data sources: Nineteen electronic resources, including MEDLINE, EMBASE and The Cochrane Library (inception to November 2009). Additional studies were sought from reference lists and clinical experts.
Review methods: Inclusion criteria were applied by two reviewers independently. Data extraction and quality assessment were undertaken by one reviewer and checked by a second. Prospective studies of adults or children with bilateral hearing loss were eligible. Comparisons were BAHAs versus conventional hearing aids [air conduction hearing aid (ACHA) or bone conduction hearing aid (BCHA)], unaided hearing and ear surgery; and unilateral versus bilateral BAHAs. Outcomes included hearing measures, validated measures of quality of life (QoL), adverse events and measures of cost-effectiveness. For the review of cost-effectiveness, full economic evaluations were eligible.
Results: Twelve studies were included (seven cohort pre–post studies and five cross-sectional 'audiological comparison' studies). No prospective studies comparing BAHAs with ear surgery were identified. Overall quality was rated as weak for all included studies and meta-analysis was not possible due to differences in outcome measures and patient populations.
There appeared to be some audiological benefits of BAHAs compared with BCHAs and improvements in speech understanding in noise compared with ACHAs; however, ACHAs may produce better audiological results for other outcomes. The limited evidence reduces certainty. Hearing is improved with BAHAs compared with unaided hearing. Improvements in QoL with BAHAs were identified by a hearing-specific instrument but not generic QoL measures. Studies comparing unilateral with bilateral BAHAs suggested benefits of bilateral

BAHAs in many, but not all, situations. Prospective case series reported between 6.1% and 19.4% loss of implants. Most participants experienced no or minor skin reactions.
A decision analytic model was developed. Costs and benefits of unilateral BAHAs were estimated over a 10-year time horizon, applying discount rates of 3.5%. The incremental cost per user receiving BAHA, compared with BCHA, was £16,409 for children and £13,449 for adults. In an exploratory analysis the incremental cost per quality-adjusted life-year (QALY) gained was between £55,642 and £119,367 for children and between £46,628 and £100,029 for adults for BAHAs compared with BCHA, depending on the assumed QoL gain and proportion of each modelled cohort using their hearing aid for ≥8 or more hours per day. Deterministic sensitivity analysis suggested that the results were highly sensitive to the assumed proportion of people using BCHA for ≥8 hours per day, with very high incremental cost-effectiveness ratio values (£500,000–1,200,000 per QALY gained) associated with a high proportion of people using BCHA. More acceptable values (£15,000–37,000 per QALY gained) were associated with a low proportion of people using BCHA for ≥8 hours per day (compared with BAHA).
Limitations: The economic evaluation presented in this report is severely limited by a lack of robust evidence on the outcome of hearing aid provision. This has lead to a more restricted analysis than was originally anticipated (limited to a comparison of BAHA and BCHA). In the absence of useable QoL data, the cost-effectiveness analysis is based on potential utility gains from hearing, that been inferred using a QoL instrument rather than measures reported by hearing aid users themselves. As a result the analysis is regarded as exploratory and the reported results should be interpreted with caution.
Conclusions: Exploratory cost-effectiveness analysis suggests that BAHAs are unlikely to be a cost-effective option where the benefits (in terms of hearing gain and probability of using of alternative aids) are similar for BAHAs and their comparators. The greater the benefit from aided hearing and the greater the difference in the proportion of people using the hearing aid for ≥8 hours per day, the more likely BAHAs are to be a cost-effective option. The inclusion of other dimensions of QoL may also increase the likelihood of BAHAs being a cost-effective option. A national audit of BAHAs is needed to provide clarity on the many areas of uncertainty surrounding BAHAs. Further research into the non-audiological benefits of BAHAs, including QoL, is required.
Funding: This report was commissioned by the National Institute for Health Research Health Technology Assessment programme.

Contents

Glossary

Aided thresholds The softest sounds that a person can hear while wearing hearing aid(s).

Baffle side The side on which the hearing aid is worn.

Deaf people The Royal National Institute for Deaf People uses the term 'deaf people' in a general way when talking about people with all degrees of deafness. Similarly, throughout this report we use the term 'people who are bilaterally deaf' to describe people with all degrees of deafness and hearing loss but affecting both ears to the same or differing degrees.

Decibel (dB) Logarithmic unit of sound intensity. Letters A, B or C following dB in parenthesis [e.g. dB(A)] indicate the frequency weightings on a sound level meter.

Decibels hearing level (dB HL) Decibel scale referenced to accepted standards for normal hearing (0 dB is average normal hearing for each audiometric test frequency).

Decibels sound pressure level (dB SPL) Decibel scale referenced to a physical standard for pressure intensity.

Directional hearing (auditory localisation) The ability to locate the direction from which a sound is coming, making use of differences in intensity and/or phase between the two ears.

Free field A sound field whose boundaries exert a negligible effect on the sound waves, but often used synonymously with sound field. Note that the term 'sound field' testing is used throughout the report, despite some included studies using the term 'free field' testing, as this is unlikely for most studies.

Masking The process by which the threshold of audibility for one sound is raised by the presence of another (masking) sound.

Masking level difference (binaural masking level difference) The difference in threshold of the signal when the signal and a masker have the same phase and level relationships at the two ears and when the interaural phase and/or level relationships of the signal and masker differ.

Maximum phoneme score The highest score obtained in a phoneme test irrespective of presentation level.

Phoneme The minimal unit of sound in a language that is distinct from other sounds. Phoneme identification is used in speech perception and auditory language comprehension, testing phonological awareness or vowel identification abilities.

Plomp test A test measuring speech reception threshold in quiet or noise.

Pure tone A sound whose instantaneous sound pressure follows a sinusoidal function of time. Such a sound has only a single frequency component. Pure-tone stimuli are used to measure hearing sensitivity in audiometry.

Pure-tone audiometry The procedure most commonly used for the measurement of hearing impairment. Pure tones are presented via air conduction and bone conduction and the individual's sensitivity to discrete frequencies is measured.

Shadow side The side opposite to the hearing aid (head shadow refers to the reduction of sounds as they travel from one side of the head to the other).

Signal-to-noise ratio, speech-to-noise ratio Relationship between the sound levels of the signal and the noise of the listener's ear, commonly reported as the difference in decibels between the intensity of the signal and the intensity of the background noise (e.g. if the speech signal is measured at 70 dB and the noise is 64 dB, the signal-to-noise ratio is + 6 dB). The higher the signal-to-noise ratio, the more difficult an individual finds it to hear in noise.

Sound field A space where sound is propagated. In sound field testing, calibrated auditory signals are presented through loudspeakers into a sound-isolated room rather than through headphones to test hearing, often used when testing children who will not tolerate headphones and in evaluating hearing performance. Often used synonymously with free field. Note that the term 'sound field' testing is used throughout the report despite some included studies using the term 'free field' testing, as this is unlikely for most studies.

Speech audiometry Measurement of speech perception skills including speech awareness and speech recognition, one component of an audiometric test battery.

Speech detection threshold (speech awareness threshold) The lowest intensity level at which a person can detect the presence of a speech signal, it approximates the best hearing level in the 250–800 hertz (Hz) audiometric frequency region. Used clinically with children or others who have such a poor speech understanding that a speech recognition threshold cannot be obtained.

Speech-in-noise A functional hearing test assessing how well an individual can understand speech in a noisy environment.

Speech-in-quiet A functional hearing test assessing how well an individual can understand speech in a quiet environment.

Speech recognition threshold (speech reception threshold) The lowest intensity level at which a person can detect the presence of a speech signal, it approximates the best hearing level in the 250–800 Hz audiometric frequency region.

Threshold The intensity at which an individual can just barely hear a sound 50% of the time; all sounds louder than threshold can be heard, but sounds below threshold cannot be detected.

Warble-tone thresholds An acoustic signal produced by modifying a pure tone with small and rapid changes in frequency; used in sound field audiometry to minimise the likelihood of standing waves from reflective surfaces.

Word recognition test (speech discrimination test) A speech audiometry measure that typically uses monosyllabic words presented at a suprathreshold level in an open-set format; provides an assessment of a person's speech understanding as a per cent correct score.

List of abbreviations

AC	air conduction
ACHA	air conduction hearing aid
BAHA	bone-anchored hearing aid
BC	bone conduction
BCHA	bone conduction hearing aid
BNF	*British National Formulary*
CHL	conductive hearing loss
CI	confidence interval
DEALE	declining exponential approximation to life expectancy
dB	decibel(s)
dB(A)	decibels A-weighted (decibels measured using an A scale sound filter)
dB HL	decibels hearing level
dB SPL	decibels sound pressure level
DSA	deterministic sensitivity analysis
ENT	ear, nose and throat
EQ-5D	European Quality of Life-5 Dimensions
FDA	Food and Drug Admistration
HHDI	Hearing Handicap and Disability Index
HUI	Health Utilities Index
Hz	hertz (unit of frequency)
ICER	incremental cost-effectiveness ratio
IOI-HA	International Outcomes Inventory for Hearing Aids
kHz	kilohertz
MAIS	Meaningful Auditory Integration Scale
MUSS	Meaningful Use of Speech Scale
NIHR	National Institute for Health Research
OR	odds ratio
PCT	primary care trust
PSA	probabilistic sensitivity analysis
PSS	Personal Social Services
PTA	pure-tone average
QALY	quality-adjusted life-year
QoL	quality of life
RCT	randomised controlled trial
SD	standard deviation
SF-36	Short Form questionnaire-36 items
SNHL	sensorineural hearing loss
SNR	signal-to-noise ratio
SRT	speech reception threshold
SUHT	Southampton University Hospitals NHS Trust
UHB	University Hospitals Birmingham NHS Foundation Trust
WTP	willingness to pay

All abbreviations that have been used in this report are listed here unless the abbreviation is well known (e.g. NHS), or it has been used only once, or it is a non-standard abbreviation used only in figures/tables/appendices, in which case the abbreviation is defined in the figure legend or in the notes at the end of the table.

Executive summary

Background

A bone-anchored hearing aid (BAHA) consists of a permanent titanium fixture, which is surgically implanted into the skull bone behind the ear, and a small detachable sound processor that clips onto the fixture. Sound is transmitted to the cochlea via bone conduction. BAHAs are suitable for people with conductive or mixed hearing loss who cannot benefit fully from conventional hearing aids. They can be used unilaterally or bilaterally for people with bilateral hearing loss.

Objectives

- To assess the clinical effectiveness and cost-effectiveness of BAHAs for people who are bilaterally deaf. The evaluation will consider BAHAs compared with conventional hearing aids, ear surgery and the unaided condition, and the use of unilateral or bilateral BAHAs.
- To adapt an existing economic model or develop a new economic model relevant to the UK setting.
- To identify areas where further research is required.

Methods

Data sources

Nineteen electronic databases, including MEDLINE, EMBASE and The Cochrane Library, were searched from inception to November 2009. Bibliographies of relevant papers were checked and experts were contacted to identify additional studies.

Study selection

Titles and abstracts were screened for eligibility and inclusion criteria defined a priori were applied to the full text of selected papers by two reviewers independently. The inclusion criteria were as follows:

- Participants: adults or children with bilateral hearing loss.
- Interventions: BAHAs attached to a surgically implanted titanium fixture.
- Comparisons: unilateral versus bilateral BAHAs, conventional hearing aids [air conduction hearing aid (ACHA) or bone conduction hearing aid (BCHA)], unaided hearing, ear surgery (tympanoplasty, myringoplasty, ossiculoplasty, stapedectomy and stapedotomy).
- Outcomes: hearing measures, aided hearing thresholds, speech recognition scores, validated measures of quality of life (QoL) and patient satisfaction, adverse events, measures of cost-effectiveness [cost per quality-adjusted life-year (QALY); cost per life-year saved] and consequences for health-service resources.
- Types of studies:
  - Systematic review of clinical effectiveness – randomised controlled trials, controlled clinical trials, prospective cohort analytic studies (with control group), prospective cohort pre and post studies (one group, before and after BAHA surgery), cross-sectional 'audiological comparison studies' (one time point) and prospective case series. Only

studies with the most rigorous designs were included for each comparator. Where higher level evidence was limited to BAHA models no longer in current use, lower level evidence for models in current use was included. Abstracts were considered if sufficient information was presented.

- Systematic review of cost-effectiveness – full economic evaluations reporting both costs and outcomes were eligible. Conference abstracts were not eligible for inclusion in the cost-effectiveness section.

Data extraction and quality assessment

Data extraction and quality assessment were undertaken by one reviewer and checked by a second reviewer, with differences resolved through discussion.

Data synthesis

Clinical effectiveness data were synthesised through a narrative review with full tabulation of results. Audiological outcome measures were discussed throughout the review of clinical effectiveness as reported by the included studies, including the use of descriptions such as 'improvement' or 'deterioration'. To aid interpretation of the data, lower hearing thresholds were considered to be 'better' than higher thresholds, but it is acknowledged that this is a simplistic approach and, although true in many cases, it is not necessarily so.

Results

Quantity and quality of studies

Searching identified 665 references; 41 of these met the inclusion criteria. After selecting the highest level of evidence available for each comparator and identifying additional studies with BAHA models in current use, 12 studies (reported in 15 publications) were included in the review of clinical effectiveness (seven cohort pre–post studies and five cross-sectional audiological comparison studies). No studies with a control group were identified. Seven studies compared BAHAs with conventional hearing aids, three of these and one additional study compared BAHAs with unaided hearing, and four studies compared unilateral and bilateral BAHAs. No prospective studies comparing BAHAs with ear surgery were identified. The overall quality was rated as weak for all included studies and meta-analysis was not possible due to differences in outcome measures and patient populations.

Summary of clinical effectiveness

BAHAs versus BCHA

Two studies found an improvement in sound field pure-tone average and warble-tone thresholds with BAHAs, but statistical analysis was reported by only one study ($p < 0.01$). One study found hearing was better with the BCHA at 0.25 and 0.50 kilohertz (kHz) [p-value not reported (NR)]. Studies reported improvements in 100% speech audiometry discrimination [62 decibels hearing level (dB HL) vs 48 dB HL], location of a sound (0% vs 80% of cases) and maximum phoneme score [mean standard deviation (SD) 36.1% (28.9%) vs 48.7% (31.7%)], but statistical significance was not reported. An improvement in speech reception threshold in quiet {mean difference 2.7 decibels (dB) (SD 4.4 dB), $p < 0.05$} and speech-to-noise ratio [2.5 dB (SD 2.2 dB), $p < 0.05$] was found in one study, but another study found no difference in speech recognition threshold {mean decibels A-weighted [dB(A)] (SD): 40 (7.1) vs 38.8 (11.1), $p =$ NR}. No statistically significant difference in mean sound field speech discrimination score at 63 dB was found by one study. Statistically significant improvements in QoL were found with a disease-specific instrument but not with generic QoL measures in one study.

BAHAs versus ACHA

Results for sound field pure-tone or warble-tone thresholds were inconsistent between the studies; for example, one study found the ACHA produced better results between 1 and 4 kHz (p = NR), another found an improvement in mean thresholds (0.5–4.0 kHz, $p < 0.0.1$) with the BAHA. The direction of the effect was also unclear for speech audiometry. Three studies reported better outcomes with the ACHA for speech discrimination scores [mean (SD) 91.6% (14.7%) vs 84% (22.3%), p = NR], maximum phoneme score [mean (SD) 81.6% (8.7%) vs 67.6% (22.2%), p = NR] or speech recognition threshold [mean (SD) 39 dB(A) (10.8) vs 45 dB(A) (5), p = NR; mean deterioration with BAHA –6.4 dB (SD 3.7), $p < 0.05$]. One study found no difference in maximum phoneme score [difference 1.0% (SD 5.4%), p = not significant]. However, three studies found an improvement in speech-to-noise ratio with BAHA (difference range 1.1–2.5 dB). Speech discrimination score was statistically significantly better with the BAHA in the congenital group but not in the chronic suppurative otitis media group in one study. Statistically significant improvements in QoL were found with a disease-specific instrument but not with generic QoL measures in one study.

BAHAs versus unaided hearing

Of the four included studies, all found improvements in sound field thresholds with BAHA, which were statistically significant in the two studies reporting analysis. Three studies reported speech audiometry and found improvements with BAHAs compared with unaided hearing.

Unilateral versus bilateral BAHAs

An improvement in sound field average tone thresholds with bilateral BAHAs compared with unilateral BAHAs was found in adults (2–15 dB) and a small group (n = 3) of children [30 (SD 5) dB HL vs 25 (SD 5) dB HL].

Speech recognition thresholds in quiet were statistically significantly lower with bilateral BAHAs in two studies [41.5 dB(A) vs 37.5 dB(A); 38.7 dB HL vs 33.3 dB HL], although one study found similar results between unilateral and bilateral BAHAs. Three studies demonstrated that bilateral BAHAs produced better results than unilateral BAHAs when noise was presented from the baffle/best side (the side with the BAHA in the unilateral condition), but not when noise was presented from the shadow side (the side opposite to the BAHA in the unilateral condition); this is due to the increased noise transmitted to the ears with an extra BAHA on the shadow (noise) side. Three studies found that localisation of sound was improved with bilateral BAHAs. Two studies suggested that BAHAs enable binaural hearing. Similar results were found for unilateral and bilateral BAHAs on the Meaningful Auditory Integration Scale and Meaningful Use of Speech Scale and the International Outcomes Inventory for Hearing Aids for most items.

Adverse events

The included studies reported very limited data on adverse events. Five prospective case series reported rates of loss of implants ranging between 6.1% of implants (9–25 months' follow-up) and 19.4% of implants (median 6 years' follow-up). The vast majority of participants experienced no, or minor, skin reactions.

Summary of cost-effectiveness studies

Systematic searches identified no relevant, published full economic evaluations of BAHAs. One unpublished economic evaluation, with a minority of participants having bilateral hearing loss, was identified. Two cost studies were identified, one of which was used to help inform the cost analysis for the economic model. One QoL study was also identified, but on further inspection data were of limited value.

Summary of economic model

A decision-analytic model was developed to estimate the cost-effectiveness of unilateral BAHAs compared with BCHAs for a cohort of adults and children with hearing loss and who were ineligible for conventional ACHAs. The model was informed by a systematic search of the literature to identify parameters on the natural history and epidemiology for people with profound hearing loss, health-related QoL and costs. The intervention effects in terms of improvement in hearing and adverse events were derived from the systematic review of clinical effectiveness. The perspective of the analysis was that of the NHS and Personal Social Services. The model estimated the costs and benefits of unilateral BAHAs over a 10-year time horizon, applying discount rates of 3.5%. The outcome of the economic evaluation is reported as cost per case and cost per successful implantation.

The incremental cost per user receiving a BAHA, compared with BCHA, was £16,409 for children and £13,449 for adults. The cost per case successfully treated with a BAHA was estimated at £18,681 for children and £15,785 for adults, over a 10-year time horizon. In an augmented, exploratory analysis (inferring QoL gains using the hearing dimension of the Health Utilities Index-3) the incremental cost per QALY gained was between £55,642 and £119,367 for children and between £46,628 and £100,029 for adults for BAHAs compared with BCHA, depending on the assumed QoL gain and proportion of each modelled cohort using their hearing aid for ≥ 8 hours per day.

Caution should be taken with the interpretation of the results from the economic evaluation owing to the paucity of evidence on the benefits of the BAHAs, particularly the absence of any robust mapping between audiological benefits (reported in studies included in the review of clinical effectiveness) and overall impact on QoL. As a consequence, the results of the economic evaluation should be regarded as exploratory.

Sensitivity analyses

Deterministic sensitivity analyses suggested that the results of our cost analysis were generally robust to variation in the value of input parameters. The results were most sensitive to variation in the probability of re-operation (when implants lose bone integration), the cost of surgical implantation and, to a lesser extent, the probability of intolerable pain requiring removal of the BAHA fixture.

Deterministic sensitivity analysis of the exploratory cost-effectiveness model suggested that the results were generally robust to variation in input probabilities and cost. The greatest variation, in relation to these factors, was associated with initial failure of bone integration, failure of BAHA implantation due to intolerable pain, the probability of re-operation due to loss of bone integration, the cost of day surgery for implantation and the cost of components of the BAHA system. The results of the cost-effectiveness analysis were highly sensitive to the assumed proportion of people using their hearing aid for ≥ 8 hours per day, with very high incremental cost-effectiveness ratio values (in the range from £500,000 to £1,200,000 per QALY gained) associated with a high proportion of people using BCHA for ≥ 8 hours per day. More acceptable values (in the range from £15,000 to £37,000 per QALY gained) were associated with a low proportion of people using BCHA for ≥ 8 hours per day (compared with BAHA). In a threshold analysis, differences in the proportion of people using their hearing aid for ≥ 8 hours per day (for BAHA compared with BCHA) of between 30% and 40% for the lowest estimated utility gain from aided hearing, and between 15% and 18% for the greatest estimated utility gain from aided hearing, were required for BAHAs to be cost-effective at a willingness-to-pay threshold of £30,000 per QALY gained.

Conclusions

The available evidence is methodologically weak and the results have a high risk of bias. As such, there is a high degree of uncertainty about the conclusions of this systematic review.

The findings suggest that hearing is improved with BAHAs compared with no hearing aid, and although there are audiological benefits of BAHAs when compared with conventional BCHAs, the audiological benefits of BAHAs when compared with ACHAs are less clear. Limited data suggest an improvement in QoL with BAHAs when compared with conventional aids, but there is an absence of evidence regarding other potential benefits, such as length of time the aid is able to be worn and improvement of discharging ears. The evidence suggests that there are some benefits of bilateral BAHAs compared with unilateral BAHAs. The results of our cost analysis demonstrate that BAHAs are significantly more costly than conventional BCHAs. The additional costs continue while individuals remain using their BAHA and are not restricted to the initial processes of surgical implantation and fitting of the BAHA sound processor. Our exploratory cost-effectiveness analysis of BAHAs versus BCHAs suggests that BAHAs are unlikely to be a cost-effective option where the benefits (in terms of hearing gain and probability of using of alternative aids) are similar for BAHAs and their comparators. The greater the benefit from aided hearing and, in particular, the greater the difference in the proportion of people using the hearing aid for ≥ 8 hours per day, the more likely BAHAs are to be a cost-effective option. The inclusion of other dimensions of QoL may also increase the likelihood of BAHAs being a cost-effective option.

Recommendations for further research

A national audit of BAHAs should be implemented to provide clarity on the many areas of uncertainty surrounding BAHAs. Further research into the non-audiological benefits of BAHAs, including QoL, is required. Good-quality trials are needed to establish the benefits of bilateral BAHAs compared with unilateral BAHAs in people who are bilaterally deaf.

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Chapter 1

Aim and background

Aim

The aim of this report is to synthesise the evidence assessing the clinical effectiveness and cost-effectiveness of bone-anchored hearing aids (BAHAs) for people who are bilaterally deaf.

The evaluation will consider BAHAs compared with conventional hearing aids, ear surgery and the unaided condition, and the use of unilateral or bilateral BAHAs. If the systematic review of economic evaluations shows that there are no appropriate good-quality economic evaluations, an economic model relevant to the UK setting is to be developed. The study aims to identify areas where further research is required.

Description of underlying health problem

Deafness and hearing loss can be described as mild, moderate, severe or profound (*Table 1*), and are defined according to the quietest sound a person can hear across a range of frequencies. The greater this threshold is, measured in units of decibels hearing level (dB HL), the worse the hearing loss is. Hearing loss that occurs in both ears is described as bilateral, and may be the same or different in each ear. Single-sided (unilateral) deafness is excluded from this evaluation.

Normal hearing occurs when sound waves travel through the external, middle and inner ear and are translated into nerve impulses, which are interpreted by the brain. The external ear acts as a sound-collecting funnel, with the passing sound waves causing the eardrum to vibrate. These vibrations (sound waves) are then passed on by the eardrum to three small bones (ossicles) in the middle ear, which amplify the vibrations and pass them on to the cochlea (inner ear). Movement of tiny hair-like cells in the fluid-filled cochlea convert the sound waves into nerve impulses, which are transmitted to the brain by the auditory nerve. Disturbances at any point in this pathway can cause hearing loss.

The main types of hearing loss are sensorineural loss and conductive loss,[2] and the presence of both types is referred to as mixed hearing loss. Sensorineural hearing loss (SNHL) is caused by damage to the outer or inner hair cells of the cochlea or the auditory nerve.[3] SNHL involves a loss of both the ability to detect quiet sound (acuity) and the ability to make sense of sound

TABLE 1 Definitions of deafness and hearing loss

Audiometric descriptor[1]	Hearing threshold level (dB)[a]
Mild hearing loss	20–40
Moderate hearing loss	41–70
Severe hearing loss	71–95
Profound hearing loss	>95

dB, decibels.
a Average pure-tone hearing threshold levels at 250, 500, 1000 and 4000 hertz.

(discrimination) and is the most common form of hearing loss in more developed countries[4] (approximately 90%).[3,5] It is often attributed to natural deterioration with ageing and prolonged noise exposure.[3] SNHL can have almost any frequency configuration and extent (from mild to profound).

Conductive hearing loss (CHL) involves a loss of acuity only and is the result of damage or blockage in the outer or middle ear due to a variety of causes such as infection, fluid (otitis media with effusion), ostosclerosis (growth of extra bone tissue) or trauma/damage to the eardrum. CHL may be caused by congenital abnormalities, which can affect any or all of the outer and middle ear structures,[6] or may be part of a syndrome such as Treacher Collins, Crouzon, branchio-oto-renal or Goldenhar syndrome.[7] It can also occur following mastoid surgery or in Down syndrome[8,9] (although SNHL can also occur with Down syndrome). The most common cause of CHL in children is otitis media with effusion and although this hearing loss is often only temporary,[10] it may be permanent in a very small number of cases.[6] CHL is most commonly of a flat frequency configuration, and its maximum extent can only be that of the contribution of the conductive pathway to audition [40–50 decibels (dB)].

A less common type of hearing loss is neural deafness, caused by the absence of, or damage to, the auditory nerve. This type of hearing loss does not benefit from sound amplification as the nerve is unable to pass on any or enough sound information,[2] and is therefore not considered further in this review.

The majority of people with hearing loss benefit from conventional air conduction hearing aids (ACHAs). These aids receive, amplify and transmit sound down the ear canal to the cochlea and are fitted behind the ear, in the ear or in the ear canal. However, people with an obstructed conduction process (via air) are unable to benefit fully or at all from ACHAs. For those with an infected ear, ACHAs may prevent adequate ventilation of the ear and thereby exacerbate the infection, whereas congenital abnormality or atresia of the pinna (external ear) may prevent an ACHA being fitted.[6] Some people with CHL can be treated with surgery in the form of repairing perforated eardrums, reconstruction or stapedectomy (surgical removal of the stapes ossicle of the middle ear),[3,6,10] but for those for whom surgery is not an option, bone conduction hearing aids (BCHAs) may be an alternative.

Conventional BCHAs use a vibrator pressed firmly against the skin of the skull via a spring headband or special spectacles to conduct sound directly through the bone to the cochlea of the inner ear, bypassing the impaired or diseased external or middle ear.[11] However, BCHAs are associated with a number of drawbacks: they are uncomfortable to wear owing to the pressure needed to apply the device effectively and can cause skin irritations and headaches; they have poor aesthetics and are difficult to hide; and speech recognition can be affected by insecure positioning or shifting of the transducer and by the attenuation of sound by tissue layers between the vibrator and the skull.[12–14]

An alternative type of hearing aid which utilises bone conduction (BC) is the BAHA, where contact with the skull is maintained by a surgical implant. It should be noted that the term 'Baha®' is a registered trademark of Cochlear Bone Anchored Solutions AB, a Cochlear™ group company; however, reference to BAHA in this report applies to all such BC devices and not to the manufacturer, supplier or trade name. BAHAs are used to help people with conductive or mixed hearing loss who cannot benefit from conventional hearing aids or from ear surgery, or in some cases as an alternative to surgery (stapedectomy). BAHAs have undergone a number of developments since they were first introduced in 1977, and are discussed in further detail in *Description of BAHAs*.

Epidemiology of hearing loss

Although an understanding of the epidemiology of hearing loss is key to the assessment of the clinical effectiveness and cost-effectiveness of a technology and to the subsequent development of guidance on its provision and use, limited research has been undertaken.[15] Assessments of the epidemiology of hearing loss have tended to focus on retrospective cohort studies of its prevalence and have been limited in the type of hearing loss considered. They often use surrogate measures of prevalence such as use of health services, rather than population-based studies, resulting in the potential to underestimate needs. In addition, studies have tended to be affected by differences in the methods for assessing and diagnosing hearing loss (e.g. self-assessment) and variations in definitions and classification of hearing loss (including arbitrary nature of thresholds). Several studies have been undertaken within the UK and elsewhere and those most relevant to this evaluation are discussed in the following section. As there is little evidence focusing on CHL, the epidemiology of hearing loss in general is discussed to give some context to this evaluation.

Prevalence of hearing loss

Children

The prevalence of hearing loss in children has been assessed in several population-based surveys within the UK and elsewhere. These studies have shown variations in prevalence, with rates differing depending on the type of loss, its severity, temporal factors and its aetiology. Within the UK a series of retrospective studies (population surveys) has been undertaken by the Medical Research Council Institute for Hearing Research, providing prevalence data for several cohorts within cities, regions and nationally. In a retrospective survey of providers of health and educational services to children with a hearing loss conducted in 1995, Fortnum and Davis[16] assessed the prevalence of permanent hearing loss [≥ 40 dB HL averaged over 0.5, 1.0, 2.0 and 4.0 kilohertz (kHz)] in children resident in the Trent Health Region (UK) who were born between 1985 and 1990. They found an overall prevalence of permanent hearing loss of 1.33 cases per 1000 live births. For those with severe hearing loss (70–94 dB HL) the prevalence was 0.28 per 1000 and for profound hearing loss (≥ 95 dB HL) 0.31 per 1000. Congenital hearing loss was more prevalent than acquired hearing loss, accounting for 1.12 cases per 1000 live births (*Table 2*). Fortnum and Davis found that SNHL was more common than purely conductive loss.[16] The prevalence of congenital and acquired SNHL (≥ 40 dB HL) was 1.27 per 1000 live births compared with 1.33 per 1000 for all hearing losses. The higher prevalence of impairments that were congenital compared with acquired, and of sensorineural compared with conductive impediments, was evident for different levels of severity of hearing loss (see *Table 2*).

Fortnum and colleagues[17] undertook a similar study to estimate the prevalence of permanent bilateral hearing loss (greater than 40 dB averaged over pure-tone threshold of 0.5, 1.0, 2.0 and 4.0 kHz in the better hearing ear) in children born between 1980 and 1995 who were resident in the UK in 1998. The retrospective survey of all health and educational providers for hearing-impaired children identified 17,160 cases, finding a prevalence of 0.91 per 1000 live births for children aged 3 years and a prevalence of 1.65 per 1000 for those aged 9–16 years (data for children aged 4–8 years were not provided numerically). Adjustment of these rates for underascertainment resulted in an increase to 1.07 and 2.05 per 1000 live births for children aged 3 years and 9–16 years, respectively. When comparing the prevalence by the severity of hearing loss, rates were higher among those children with a moderate loss than among those with a severe or profound loss. Some 0.45 per 1000 children aged 3 years and 0.89 per 1000 children aged 9–16 years had a hearing loss of 41–70 dB HL compared with 0.20 per 1000 and 0.35 per 1000 for children aged 3 years and 9–16 years, respectively, for a loss of 71–95 dB HL (severe) and 0.26 per 1000 and 0.39 per 1000 for children aged 3 years and 9–16 years, respectively, for a loss of ≥ 95 dB HL (profound).

TABLE 2 Prevalence of hearing loss in children

Study		Hearing loss (kHz) All	Moderate	Severe	Profound
Fortnum and Davis 1997[16]		***(≥ 40 dB HL)***	***(40–69 dB HL)***	***(70–94 dB HL)***	***(≥ 95 dB HL)***
Retrospective cohort survey	***All cases***				
Population: children born in Trent health region 1985–90, survey 1995	Congenital and acquired	1.33 (1.22 to 1.45)	0.74 (0.65 to 0.83)	0.28 (0.23 to 0.35)	0.31 (0.26 to 0.37)
Outcome: prevalence per 1000 live births (95% CI)	Congenital	1.12 (1.01 to 1.23)	0.64 (0.56 to 0.73)	0.23 (0.19 to 0.29)	0.24 (0.20 to 0.30)
	Permanent SNHL				
	Congenital and acquired	1.27 (1.16 to 1.39)	0.68 (0.61 to 0.78)	0.28 (0.23 to 0.34)	0.31 (0.26 to 0.37)
	Congenital	1.06 (0.96 to 1.17)	0.59 (0.52 to 0.68)	0.23 (0.18 to 0.28)	0.24 (0.20 to 0.30)
Fortnum* et al. *2001[17]		***(≥ 40 dB HL)***	***(41–70 dB HL)***	***(71–95 dB HL)***	***(> 95 dB HL)***
Retrospective cohort survey	Children aged 3 years	0.91 (0.85 to 0.98)	0.45 (0.40 to 0.50)	0.20 (0.17 to 0.24)	0.26 (0.22 to 0.29)
Population: children born in the UK 1980–95, survey 1998		1.07 (1.03 to 1.12)	0.60 (0.54 to 0.66)	0.22 (0.21 to 0.24)	0.27 (0.26 to 0.29)
	Children aged 9–16 years	1.65 (1.62 to 1.68)	0.89 (0.86 to 0.91)	0.35 (0.33 to 0.36)	0.39 (0.38 to 0.41)
Outcome: prevalence per 1000 live births (95% CI)		2.05 (2.02 to 2.08)[a]	1.21 (1.18 to 1.24)[a]	0.41 (0.40 to 0.42)[a]	0.44 (0.43 to 0.44)[a]
Fortnum* et al. *2002[18]		***(≥ 40 dB HL)***	***(41–70 dB HL)***	***(71–95 dB HL)***	***(> 95 dB HL)***
Retrospective cohort survey	Total known aetiology	50.6	47.7	50.9	57.5
Population: children born in the UK 1980–95, survey 1998	Genetic	20.2	18.3	20.8	24.1
Outcome: percentage of total study population with aetiology	Syndromal	9.5	11.6	6.6	7.4
	Prenatal	4.1	2.4	5.2	6.8
	Perinatal	8.0	7.4	11.4	6.7
	Postnatal	6.9	5.3	6.3	11.3
	Other	1.9	2.8	0.6	1.2
MacAndie* et al. *2003[19]		***(≥ 40 dB HL)***			
Retrospective cohort survey	All	1.23			
Population: children born in the UK 1985–94, survey 2000	Congenital	1.09			
Outcome: prevalence per 1000 live births					

a Adjusted by capture–recapture.

Comparisons between the studies in the Trent health region and the UK showed limited difference between the prevalence rates when cohorts were matched for age. In the Trent health region the overall prevalence rate was 1.33 per 1000 [95% confidence interval (CI) 1.22 to 1.45] for children born in the period 1985–90 compared with 1.44 per 1000 (95% CI 1.41 to 1.48) for children born in the period 1988–93 in the UK survey. Similar prevalence rates were evident when comparing the different severities of hearing loss. In the Trent health region prevalence rates were 0.74 (95% CI 0.65 to 0.83) for moderate, 0.28 (95% CI 0.23 to 0.35) for severe and 0.31 (95% CI 0.26 to 0.37) for profound hearing loss compared with 0.80 (95% CI 0.77 to 0.82)

for moderate, 0.29 (95% CI 0.28 to 0.31) for severe and 0.34 (95% CI 0.32 to 0.35) for profound hearing loss in the UK study.

Fortnum and colleagues[18] assessed the annual prevalence of hearing loss and profound hearing loss among children born between 1980 and 1995 in the UK to see if there were any temporal patterns. They found that the prevalence of hearing loss increased from 634 cases in 1980 to 1342 cases in 1987, declining to 669 cases in 1995. The proportion of children with profound hearing loss has ranged from 31.5% of children with hearing loss in 1980 to 20.4% in 1989.

Fortnum and colleagues[18] compared the aetiology for the different levels of severity of hearing loss. It was evident that for around 50% of all children with hearing loss, the cause was not known or specified. For all hearing losses, 20.2% were genetic, 9.5% syndromal, 8.0% perinatal, 6.9% postnatal and 4.1% prenatal. When comparing the aetiology for the different severities of hearing loss it was evident that there were significant differences. Fortnum and colleagues found that children with moderate hearing loss were more likely to have an unknown aetiology than those with severe loss (moderate 52.3%, severe 49.1%, profound 42.5%, $p < 0.001$) or a syndromal aetiology (moderate 11.6%, severe 6.6%, profound 7.4%, $p < 0.001$). Also, severely impaired children were more likely to have a perinatal cause (moderate 7.4%, severe 11.4%, profound 6.7%, $p < 0.001$) and profoundly impaired children to have a genetic (moderate 18.3%, severe 20.8%, profound 24.1%, $p < 0.001$), prenatal (moderate 2.4%, severe 5.2%, profound 6.8%, $p < 0.001$) or postnatal aetiology (moderate 5.3%, severe 6.3%, profound 11.3%, $p < 0.001$) than the other groups.

Similar prevalence rates for hearing loss were shown by MacAndie and colleagues[19] in a retrospective study in Greater Glasgow (UK). The study focused on children born between 1985 and 1994 who were identified from the Educational Audiology database. Of the 105,517 live births in Greater Glasgow between 1985 and 1994, 130 children had a permanent hearing loss (≥ 40 dB HL), which equates to an incidence of 1.23 cases per 1000 live births. Some 116 children had a congenital hearing loss (1.09 per 1000 live births), with only 14 children having a hearing loss that was postnatally acquired or progressive. When assessing the aetiology of bilateral hearing loss, MacAndie and colleagues found that 31% of children had a family history of congenital hearing loss, 12% craniofacial syndrome, 15% had an admission to neonatal intensive care unit that may have contributed to their hearing loss, 7% had a postnatal infection, 3% a prenatal infection and 28% had an unknown or uncategorised aetiology.

Adults

Davis[20] surveyed the prevalence of hearing loss among a cohort of 35,330 people within four cities in the UK between 1980 and 1986. The study found that 16.1% of people aged 17–80 years had mild (≥ 25 dB HL), 3.9% moderate (≥ 45 dB HL) and 1.1% severe (≥ 65 dB HL) hearing loss in both ears. The prevalence of bilateral hearing loss was shown to increase with age. Prevalence rates for moderate bilateral loss increased from 0.2% for those aged 17–30 years to 1.1% for 31- to 40-year-olds, 1.7% for 41- to 50-year-olds, 4.0% for 51- to 60-year-olds, 7.4% for 61- to 70-year-olds and 17.6% for 71- to 80-year-olds. Similar variations by age group were evident for those people with a severe bilateral hearing loss, although the prevalence rates were approximately a quarter of those for people with moderate hearing loss. For those with severe bilateral loss the rates varied from less than 0.1% for those aged 17–30 years to 0.7% for 31- to 40-year-olds, 0.3% for 41- to 50-year-olds, 0.9% for 51- to 60-year-olds, 2.3% for 61- to 70-year-olds and 4.0% for 71- to 80-year-olds.

Davis[20] assessed the effects of age, sex, occupational group and occupational noise on hearing loss through logistic regression analysis. The prevalence of hearing loss (≥ 45 dB HL) was shown to significantly increase with a person's age [odds ratios (OR) 7.6 ($p < 0.05$) for 41–50 years; 17.3

($p < 0.005$) for 51–60 years; 32.1 ($p < 0.005$) for 61–70 years; 95.4 ($p < 0.005$) for 71–80 years], occupation [OR 2.2 ($p < 0.005$) for manual occupations] and exposure to occupational noise [OR 2.3 ($p < 0.01$) for ≥ 91 dB(A) equivalent continuous sound level (L_{eq}].

Lee and colleagues[21] assessed the prevalence of self-reported hearing loss among adults (107,100 White and 17,904 African-American people aged ≥ 18 years) in the USA using the National Centre for Health Statistics National Health Interview Survey between 1986 and 1995 (annual survey of approximately 50,000 civilian households). The annual age-adjusted rates for 'some hearing impairment' and 'severe bilateral impairment' were higher among Whites than among African Americans. The rates for 'some hearing impairment' ranged from 11.0% to 12.7% for Whites and from 5.9% to 8.5% for African Americans. The prevalence of 'severe bilateral impairment' was lower for both groups, with rates ranging from 0.7% to 1.1% for Whites and from 0.1% to 0.5% for African Americans. Although the rates varied temporally during the 10 years, there were no significant upward or downward trends in prevalence.

Unsurprisingly, analysis of the prevalence of 'any hearing impairment' among different age groups showed that the older age groups had a higher prevalence of impairment. This was evident for both the White and African American groups, although the prevalence was higher for all age groups among Whites than among African Americans. Comparison of the prevalence of impairment for the different age groups during the 10-year period showed limited variation for all the age groups in the White population and among the 18–39, 40–49, 50–59 and 60–69 years age groups in the African-American population. In contrast, the 70–79 and ≥ 80 years age groups in the African-American population showed considerable variation, although there were no discernible trends in the prevalence data.

Estimates of burden of hearing loss in England and Wales

Using the studies of the prevalence of hearing loss and population estimates for England and Wales,[16,17,19,22,23] it is possible to provide a provisional estimate of the burden of bilateral hearing loss in England and Wales (*Table 3*). These estimates should be interpreted with caution owing to the differences in the nature of the studies and the classifications of hearing loss used. Estimates of the prevalence of bilateral hearing loss among children in England and Wales indicate that there could be between 900 and 1000 children in each annual birth cohort with a bilateral hearing loss of ≥ 40 dB HL. Although the majority of children would have a moderate hearing loss of 41–70 dB HL, around 400 would have either a severe (71–95 dB HL) or profound loss (≥ 95 dB HL). It was evident that the majority of impairments among children would be congenital in origin, accounting for hearing impairment in around 750–775 children per annual birth cohort in England and Wales.[16,19] Most of these congenital hearing impairments are thought to be permanent sensorineural (approximately 730 per annual birth cohort).[16] It was estimated that among adults in England and Wales there would be around 1.6 million people aged 17–80 years with a hearing loss of ≥ 45 dB, with around a quarter of these having a loss of

TABLE 3 Estimated number of children with a diagnosed bilateral hearing loss in England and Wales in each annual birth cohort

Severity of hearing loss	Range of prevalence of bilateral hearing loss (rate per 1000 live births)[16,17]	Range of estimated number of children with a bilateral hearing loss in England and Wales[a]
Moderate (41–70 dB)	0.74–0.80	511–552
Severe (71–95 dB)	0.28–0.29	193–200
Profound (≥ 95 dB)	0.31–0.34	214–235
All impairments (≥ 40 dB)	1.33–1.44	918–994

a Rates calculated using a denominator of the 690,013 live births in England and Wales in 2007.[22]

TABLE 4 Estimates of the number of adults with a bilateral hearing loss in England and Wales per age group

Age group (years)	Population in England and Wales (mid-2008) (000s)[23]	Prevalence of bilateral hearing loss (%)[20]		Estimated number of people with hearing loss in England and Wales	
		Severity of loss		Severity of loss	
		≥ 45 dB	≥ 65 dB	≥ 45 dB	≥ 65 dB
17–30	10,186.1	0.2	0.1	20,372	10,186
31–40	7516.0	1.1	0.7	82,676	52,612
41–50	7907.4	1.7	0.3	134,426	23,722
51–60	6563.6	4.0	0.9	262,544	59,072
61–70	5411.4	7.4	2.3	400,444	124,462
71–80	3718.8	17.6	4.0	654,509	148,752
Total	41,303.3	3.9	1.1	1,610,829	454,336

≥ 65 dB (*Table 4*). Around 60% (275,000) of those with a hearing loss ≥ 65 dB HL would be aged 60–80 years.

Impact of hearing loss

For those people with hearing loss who identify with the 'Deaf community' (people whose first or preferred language is British Sign Language), being deaf is seen as part of their total identity and not as a deficiency.[24] However, deafness and hearing loss can have a profound effect on individuals and have been associated with a range of negative consequences, including educational and employment disadvantages, social isolation and stigmatisation.[25,26] According to a report by the World Health Organization, hearing loss is the second leading cause globally of 'years lived with disability' and has a larger non-fatal burden than alcohol use disorders.[26] The impact of hearing loss is influenced by the severity of the loss and age at onset. Deafness present at birth or during early childhood (the pre-lingual period) has considerable effects on speech acquisition and cognitive and psychosocial development.[27] Deafness acquired post-lingually requires the individual to adopt new communication strategies and often an entirely different lifestyle,[24] and can result in isolation and compromised quality of life (QoL).[27] Hearing loss affects not only individuals, but also the people around them such as family and co-workers.[28] These people have to put more effort into communication with the individual, for example speaking more slowly and with better articulation, turning their face to allow lip-reading and moving closer.[28] As a consequence, there is a risk that people will make less contact and the individual will become more isolated.

Early hearing loss delays the development of basic auditory skills, including auditory detection, discrimination, recognition, comprehension and attention, which negatively affects the child's ability to learn and use an auditory–oral language system.[29] Difficulties with the rules of language, the meaning of words and the use of language in social contexts lead to comprehension, expressive communication and learning problems, and can result in reduced academic achievement.[29] In contrast, a number of studies have shown that children with hearing loss who are raised by parents with hearing loss often have psychosocial advantages over those who are born to hearing families, as they grow up in an environment where communication is naturally dependent on visual, not oral, cues.[24]

A recent study used both parent-report and videotaped data from 116 severely and profoundly deaf and 69 hearing preschool-age children, and demonstrated that hearing-impaired children displayed more behaviour problems and greater difficulties with oral language, parent–child communication and sustained attention than hearing children.[30] High rates of behavioural and

emotional problems and a high rate of social maladjustment according to general population norms were also found by a cross-sectional study of 84 children and adolescents (age 2–18 years) attending schools for the deaf.[31] According to parents' descriptions, children were socially isolated and not participating in structured activities. Similar results were demonstrated by a study in Upper Austria to evaluate mental health and QoL in a representative sample of deaf pupils with a bilateral impairment of at least 40 dB, from both mainstream schools and a school for the hearing impaired.[32] Using the strengths and difficulties questionnaire,[33] deaf children scored higher for conduct, emotional and peer problems than children from a normative sample, though differences were less marked for hyperactivity/inattention. Whereas parents of deaf children had a generally positive view of their child's QoL, deaf children provided a more complex picture, stressing areas of dissatisfaction.

A non-systematic review of mild bilateral hearing loss described studies demonstrating that many children with even mild hearing loss do not perform at expected academic levels, especially in the areas of vocabulary, reading comprehension and language use, and that they expend more effort in listening to speech in quiet and in the presence of background noise than children with normal hearing.[34] The author suggests that children with even a relatively mild degree of bilateral hearing loss may exert more energy than their normal-hearing peers to listen in a classroom setting, leaving them with less energy or attention capacity for processing what they hear, taking notes and other activities required of school children.[34]

There is evidence to suggest that the effects of hearing loss in adults differ according to age group. For example, a large Norwegian health-screening survey examined the association between hearing loss, measured by pure-tone audiometry, and self-report symptoms of mental health and well-being in a normal population sample of over 50,000 people aged between 20 and 101 years.[35] The survey found a moderate but clear effect of hearing loss on anxiety, depression, self-esteem and well-being among young and middle-aged people. The strongest effects were found for depression and self-esteem among young men; however, the effects were almost absent among elderly people.

These findings are supported by a recent cross-sectional study which used the internet to determine both hearing status and self-reported psychosocial health in 1511 adults aged 18–70 years.[36] Hearing status was assessed using the 'National Hearing test', an online speech-in-noise screening test (also available via telephone) that has been implemented in the UK and the Netherlands. The study found significant associations between hearing status and distress, somatisation, depression and loneliness, but not between hearing status and self-efficacy or anxiety. For every dB signal-to-noise ratio (SNR) reduction in hearing status, the odds for developing moderate or severe depression increased by 5%, and the odds for developing severe or very severe loneliness increased by 7%. The study also found that different age groups exhibit different associations between hearing status and psychosocial health; increased loneliness was an issue for the 18–29 years group and the 40- to 49-year-olds had the greatest number of significant associations (distress, somatisation, self-efficacy, depression and anxiety), but in the 60–70 years group none of the adjusted associations reached statistical significance. The authors suggest that the differences in age groups could be due to differences in the time of onset of hearing loss, in the use of health care, or in the way hearing loss is regarded; it may be considered as part of the normal ageing process by older adults,[36] whereas younger people may suffer from being different in terms of not being fully able to function as expected for people of their age.[35] Nevertheless, hearing loss can still affect the lives of older adults, as demonstrated by a population-based longitudinal study of 2688 participants aged 53–97 years.[37] This study used pure-tone audiometry to assess hearing loss, and reported a significant association between severity of hearing loss and reduced QoL.

Current service provision

In the UK, babies are screened for hearing loss as part of the NHS Newborn Hearing Screening Programme (within 26 days) and further monitoring and tests can confirm any diagnosis of hearing loss. Although there is no national school-based hearing screening programme in the UK, a 2007 survey found that most areas (over 90% of state schools) apply a hearing test at school entry,[38] and the UK National Screening Committee recommended in 2006 that screening for hearing loss in school-age children should continue.[38] Those whose hearing loss develops during later childhood or adulthood generally present to their general practitioner, who will undertake tests and refer on to an ear, nose and throat (ENT) department for assessment and treatment if necessary. In many cases people are referred on to an audiology department, where treatment is the supply of a hearing aid.

As described previously, there are different hearing aid options available to those with deafness, including ACHs, BCHAs and BAHAs. In the UK NHS, most ACHAs are now digital and the types prescribed are typically behind-the-ear types; hearing aids that sit in the ear are less often prescribed in the NHS but people may purchase their own privately. For those with congenital hearing loss, or who cannot wear ACHAs owing to infection, BCHAs can be used. However, as discussed previously, they can be uncomfortable and so many people do not use them in all situations, and some prefer not to use them at all. In some cases of bilateral deafness surgical procedures (such as stapedectomy) are considered and can lead to improved hearing, but for many there are no surgical options. In these instances BAHAs may be considered.

Bone-anchored hearing aids are available on the NHS, but are usually fitted at a specialised centre rather than in a local ENT department.[2] In general, a referral for a BAHA will come to a specialist centre from an ENT surgeon; however, in some cases an audiologist will make this referral. In either situation, an audiological assessment to ascertain suitability for a BAHA will be made. Thereafter, BAHA availability can depend on local reimbursement policies (see *Variation in services* below). Follow-up visits are required to assess if the healing process and BAHA fixture are satisfactory. There are currently 89 BAHA centres in the UK, with around 10 more planned.[39]

Quality standards in BAHAs for children and young people suggest that a child with a significant hearing loss must be provided with suitable amplification soon after diagnosis, prior to the referral to the BAHA service.[6] For some children, an ACHA may be tried in the first instance, although where a chronic CHL is present, BCHAs should always be considered, tried and evaluated and children should be provided with the opportunity to be referred for assessment to the BAHA service.[6] Until the child is old enough for BAHA surgery, a BAHA® Softband (Cochlear Bone Anchored Solutions, Sydney, Australia) may be used. This is an elastic band with a BAHA sound processor connected to a plastic snap connector disc sewn into the band. The plastic snap connector disc is held against the skin behind the ear, or at another bony location of the skull, through the pressure from the band, and works in the same way as a conventional bone conductor.

Variation in services

The number of BAHAs in use is unknown as there are no formal records, but it is thought that there are about 6000–7000 BAHAs in current use in the UK (David Proops, Birmingham Children's NHS Hospital Trust, March 2010, personal communication). Services for BAHA users vary throughout the NHS and funding is not universally available. Primary care trusts (PCTs) differ in policy on the provision of BAHAs, the eligibility criteria for funding BAHAs and sometimes the number of aids funded.[40] For most children with bilateral hearing loss, PCTs

will fund a unilateral BAHA as long as a range of criteria around the nature of the hearing loss, the indication and the social and psychological impact have been met. Few PCTs, however, will fund bilateral BAHAs. Furthermore, as stated previously, not all NHS hospitals have an audiology department or one that specialises in the fitting of BAHAs;[2] therefore, people referred for BAHAs may have to travel a considerable distance for treatment.[41]

Current service cost

Bone-anchored hearing aid funding is recovered on an individual cost-per-case basis via the PCT.[39] BAHAs are more costly than other hearings aids,[42] with the sound processor having to be replaced every 3–5 years.[13] The *NHS reference costs 2007/08*,[43] report that a day-case admission for one-stage insertion of fixture for a BAHA costs £1918. This does not include the cost of fixtures, surgical consumables and the BAHA sound processor, which are reimbursed separately, through a high-cost low-volume top-up payment. Prices from Cochlear UK suggest that the product cost for an implant, abutment and processor ranges from £2700 to £3800, this price being dependent on the type of processor used. The 2010 price list from Oticon Medical AB (William Demant Holding) gives a package deal (processor, implant and abutment) for the Ponto (Oticon Medical, Askim, Sweden) and Ponto Pro (Oticon Medical, Askim, Sweden) of £2654.64 and £2886.60, respectively. Prices for surgery, inpatient episode and internal device cost published by the Nottingham University Hospitals[44] in 2007–8 were £2683 for adults and £1588 for children. Additional maintenance costs amounted to £3800 in the first year, reducing to £1250 annually, and these did not differ for adults or children.[44]

People with BAHAs require lifelong rehabilitation. Every individual should be on a rolling maintenance programme, and therefore funding is required for ongoing maintenance and replacement. These costs, however, need to be considered in the light of the often considerable costs of hearing loss to the person, the NHS and wider society.

Description of BAHAs

Criteria for treatment

Bone-anchored hearing aids are indicated for people with conductive or mixed hearing loss who can benefit from amplification of sound. BAHAs are also indicated for unilateral sensorineural deafness, also known as single-sided deafness, which is beyond the scope of this report. Otological indications for BAHAs include:[45]

- congenital malformation of the middle/external ear or microtia
- chronically draining ear or other infective state that does not allow use of an ACHA (e.g. external otitis, draining mastoid activity)
- patients with bilateral CHL due to ossicular disease (and not appropriate for surgical correction) or unable to be aided by conventional hearing aid devices.

Chronic suppurative otitis media and recurrent ear canal infections are the most common diagnoses for adults fitted with BAHAs, as these make it difficult to wear conventional ACHAs.[46] For children, the most common diagnoses are congenital ear malformations, with the BAHA often used instead of a conventional BCHA.[46] Bone thickness is critical for implant integration[47] and is often insufficient in children under 4 years of age.[48] While it has been suggested that children as young as 3 years can be fitted with a BAHA,[2] the devices are indicated for children aged ≥ 5 years.[49,50]

Intervention

The BAHA consists of:

1. A permanent titanium implant (3–4 mm), which is surgically placed in the mastoid bone behind the ear, where it fuses with the living bone (osseointegration). The implant transfers sound vibrations to the functioning cochlea.
2. An abutment, which protrudes through the skin and connects the titanium implant to the sound processor, transferring sound vibrations.
3. A small sound processor, which picks up sound vibrations and transfers them to the abutment. The processor can be attached to the abutment and disconnected by the user. Some processors are at head level, although the more powerful are body worn.

Fitting a BAHA requires surgery and can involve either a one-stage or two-stage surgical procedure, with each stage taking around 1 hour. In the one-stage procedure, the implant and abutment are placed at the same time, whereas in the two-stage procedure, the abutment is fitted after a period of around 3 months in adults or 4–6 months in children to allow osseointegration (where bone fuses with the implant) to occur.[51] The advantage of one-stage surgery is that it requires only one surgical procedure, but it risks transmission of forces through the abutment to the fixture before osseointegration has occurred, resulting in a failure of osseointegration and loss of the fixture. The two-stage procedure is therefore most commonly used for young children, adults who may not be able to protect the abutment adequately (e.g. adults with learning difficulties) or adults with poor bone quality (e.g. irradiated bone following radiotherapy in cancer patients). The one-stage procedure is, however, being trialled in children in some centres and has been found to be safe for children as well as adults,[52,53] and can be considered for the older child aged 14–16 years. Finally, the sound processor is connected to the abutment after a period of about 1 month.[54]

Bone-anchored hearing aid surgery is generally uncomplicated. The most common potential side effects are soft tissue reactions (with poor hygiene being the most frequent reason for adverse skin reactions)[55] and loss of fixture.[54] Failures in children tend to occur soon after implantation as, relative to the adult skull, the infant skull is lower in mineral and higher in water content.[49] Re-operation rates are more common in children than adults, for example a Health Technology Assessment review[13] for the Ontario Ministry of Health and Long-Term Care found that re-operation rates for tissue reduction or repositioning were generally under 10% for adults but as high as 25% for children. Similarly, an association between younger age and increasing adverse outcomes, such as requiring revision surgery or experiencing fixture loss, was reported by a UK review of 71 children with BAHAs.[54]

If trauma or failure of osseointegration occurs, a reserve or 'sleeper' implant may be fitted during the first procedure as a backup. This allows a new vibrating part to be fitted into the second implant as soon as a problem occurs with the first, without the need for a repeated first-stage procedure and subsequent 4- to 6-month wait for osseointegration to occur, during which time the individual would be without any hearing aid. It has been usual practice to fit the sleeper approximately 5 mm from the primary fixture; however, the sleeper is rarely needed and, as the bone is thinner, it is less likely to osseointegrate successfully.[57] For bilateral hearing loss it has been recommended that the sleeper should be placed on the contralateral side at the time of the primary surgery, where it is located in an optimum position and could be used if the decision is made to proceed to bilateral BAHA placement, reducing the number of procedures needed.[57]

In the past the BAHA was fitted on just one side (unilaterally), which could be either the better hearing side if the two cochleae differ in acuity[58] or the side preferred by the individual. The vibratory patterns of bone-conducted sound would suggest that one BAHA should be sufficient for good hearing amplification in bilateral hearing loss, as sound is transmitted to both the ipsilateral (same side) and contralateral (opposite side) cochleae.[59] However, it has been suggested that people with bilateral BAHAs benefit in terms of greater stimulation levels at the cochlea, better directional hearing and space perception, and better speech recognition in noise.[14,59–61] A potential advantage of this includes road safety, especially for children. A further benefit of bilateral BAHAs is that in the event of a problem with one side, for example an infected site or malfunctioning processor, the individual still has one functioning BAHA rather than being without any hearing aid while the problem is resolved. In a consensus statement from BAHA experts in 2005,[42] bilateral application with thorough counselling was advocated in young children with severe congenital conductive hearing impairment.

However, the application of bilateral BAHAs is still debatable. Although the benefits of bilateral stimulation through air conduction (AC) are well established, the benefits with BC are less clear. One consequence of BC stimulation is crossover transmission, where the signal presented to one side of the head is transmitted to the contralateral cochlea. When bilateral stimulation occurs, the signals from each side are transmitted to both cochleae and thus interfere, potentially leading to the cancelling of the differences in signals arriving from the two ears and removing the benefits of binaural hearing.[62] The term binaural hearing 'denotes our faculty for taking advantage from comparisons of the acoustic signals at the two ears',[63] implying the involvement of specialised brain processing that compares the neural correlates of the acoustic signals at the two ears. While empirical evidence suggests that some people with two BAHAs can use some available cues for localisation of sound, the processes remain unclear.

Past BAHA models

The BAHA technique was introduced in 1977, with the first BAHA device made by Branemark and Kuikka.[64] Since then, BAHAs have undergone a series of developments. The first generation of BAHAs, HC 100 (1981–6, Wennberg finmekanik),[65] were serially produced but handmade. The second generation of BAHAs, HC 200 [1987–91, Nobel Biotech, Zurich, Switzerland (previously Nobelpharma, Göteborg, Sweden)],[65] incorporated a number of improvements such as a damped transducer and a new amplifier system. A more powerful body-worn version, known as the Superbass HC 220 (1987–97, Nobel Biotech, Zurich, Switzerland/Nobelpharma, Göteborg, Sweden) was also developed for people with poorer nerve loss.

The third generation of BAHAs included the HC 300 (1987–97, Nobel Biotech, Zurich, Switzerland/Nobelpharma, Göteborg, Sweden), later named the Classic 300 (1991–9, Nobelpharma).[65] The first Cordelle (Cochlear Bone Anchored Solutions, Sydney, NSW, Australia) (previously Mega base HC 380) was introduced in 1999, described as having the most powerful sound processor,[50] with a functional gain that exceeded older BAHAs in higher (5–7 dB) and lower (10–15 dB) frequencies.[55] This was followed by the Compact (Cochlear Bone Anchored Solutions, Sydney, NSW, Australia) (previously HC 360) in 2000.[65] Although the simple signal processing used by models such as the Classic 300 and the Compact benefitted users with CHLs, newer models use more complex signal processing schemes that also benefit users with sensorineural loss.[66] The third-generation BAHA devices marketed by Entific Medical Systems (now Cochlear Bone Anchored Solutions AB) have US Food and Drug Administration (FDA) clearance and carry the CE mark.[67]

The Xomed Audiant® (Xomed-Treace, Florida, FL, USA) was introduced in 1985 and manufactured by Xomed and Treace, but was never CE marked.[68] It was a transcutaneous type

of BAHA, which used electromagnetic energy from an external processor. The Audiant did not perform well at lower frequencies and is no longer manufactured.[47]

The above models are no longer sold in the UK and users should have received an upgrade to one of the devices described below.

Current BAHA models

There are six BAHA devices that are currently manufactured: four from Cochlear and two from Oticon Medical.

The Baha Divino™, Baha Intenso™ and Cordelle II™ were initially manufactured by Entific Medical Systems (Gothenburg, Sweden), which was acquired by the Cochlear Corporation in 2005. The Divino is described as being suitable for people with moderate-to-severe mixed hearing or symmetrical conductive loss [defined as ≤ 10 dB difference (pure-tone average, PTA) or ≤ 15 dB difference at individual frequencies]. Bilateral fitting is suitable for people with moderate-to-severe bilateral symmetrical conductive and/or mixed hearing loss. The processor's digital technology and built-in directional microphone operate entirely at head level.[69]

The Intenso device also has digital technology and operates entirely at head level.[69] It has a larger sound processor than the Divino and hence needs a larger battery. The device is indicated for people with mixed or conductive hearing loss with BC thresholds in the 0–45 dB range across speech frequencies.[70] It is also indicated for bilateral implantation in people with bilaterally symmetrical conductive or mixed hearing loss. The function gain of BC for both the Divino and the Intenso, defined as the difference between BC thresholds measured with a standard audiometer and aided sound field thresholds (expressed in dB HL), is between 5 and 10 dB.[71] The Divino is described as having good sound clarity with reduced feedback.

The Cordelle II reportedly offers even more amplification for people with a severe hearing loss and is on average 13 dB stronger than the discontinued Classic 300 model. It is indicated for CHL and mixed hearing loss, in individuals with average BC thresholds better than 45 dB (across 0.5, 1.2 and 3.0 kHz). It connects directly to external equipment such as television, MP3 players and hi-fi systems, without disconnecting the environmental microphone, and a built-in telecoil receiver allows wearers to connect to teleloop facilities. This device has a body-worn amplifier unit that powers an ear-level transducer.[69]

The latest generation of devices from Cochlear and Oticon Medical are even more sophisticated. They are digital with computer-based fitting allowing adjustment to the person's individual hearing requirements, whereas BAHAs such as the Divino are adjusted using a screwdriver. The devices also have improved quality and advanced features such as directionality.

The most recently launched BAHA sound processor by Cochlear is the BP100 (Cochlear Bone Anchored Solutions, Sydney, NSW, Australia). It is indicated for people with conductive and mixed hearing loss or single-sided sensorineural deafness and average BC thresholds of ≤ 45 dB (across 0.5, 1.0, 2.0 and 3.0 kHz). It is also indicated for bilateral implantation in people with bilaterally symmetric conductive or mixed hearing loss. The device is reported to offer improved audibility, sound quality and speech understanding owing to various automatic systems and has been attributed with a more than 25% improvement in speech understanding in noise.[72]

The Ponto and Ponto Pro processors were released in the UK in autumn 2009 by Oticon Medical. The range complies with all European medical device regulatory requirements and has FDA approval. The processors are indicated for people with conductive and mixed hearing loss with an

average BC threshold better than 45 dB HL (across 0.5, 1.0, 2.0 and 3.0 kHz), and for single-sided deafness with a PTA AC threshold of the hearing ear better than 20 dB HL (across 0.5, 1.0, 2.0 and 3.0 kHz). Bilateral fitting is applicable for most people with a symmetrical BC threshold. The Pronto Pro model contains additional advanced features such as automatic multiband adaptive directionality, noise reduction and learning volume control.

Chapter 2

Methods for the systematic review of clinical effectiveness and cost-effectiveness

The a priori methods for systematically reviewing the evidence of clinical effectiveness and cost-effectiveness are described in the research protocol (see *Appendix 1*), which was sent to experts for comment. Although helpful comments were received relating to the general content of the research protocol, there were none that identified specific problems with the methodology of the review. The methods outlined in the protocol are briefly summarised below.

Search strategy

A comprehensive search strategy was developed, tested and refined by an experienced information scientist. Separate searches were conducted to identify studies of clinical effectiveness, cost-effectiveness, QoL, resource use and costs, and epidemiology. Sources of information and search terms are provided in *Appendix 2*. The most recent search was carried out in November 2009.

A total of 19 electronic resources were searched: 13 databases listing published papers and abstracts and six databases listing ongoing studies. Searches were from database inception to the current date with no language restrictions. The following electronic databases were searched: MEDLINE (Ovid); MEDLINE In-Process & Other Non-Indexed Citations; EMBASE; The Cochrane Library including Cochrane Central Register of Controlled Trials and Cochrane Database of Systematic Reviews; Centre for Reviews and Dissemination including Health Technology Assessment Database, Database of Abstracts of Reviews of Effects and National Health Service Economic Evaluation Database; EconLit; Science Citation Index and Conference Proceedings Citation Index (Web of Science); BIOSIS; Health Management Information Consortium; National Institute for Health Research (NIHR) CRN Portfolio; Current Controlled Trials; Clinical trials.gov; CenterWatch; Health Services Research Projects in Progress; and Computer Retrieval of Information on Scientific Projects. In addition, society websites and conferences were searched for recent abstracts and ongoing studies (see *Appendix 2*). Bibliographies of retrieved articles were checked for any additional references, and the expert advisory group and BAHA manufacturers were contacted to identify additional published and unpublished studies.

Inclusion and data extraction process

Studies were selected for inclusion in the systematic review of clinical effectiveness through a two-stage process using predefined and explicit criteria. The full literature search results were independently screened by two reviewers to identify all citations that possibly met the inclusion criteria. Full papers of relevant studies were retrieved and assessed independently by two reviewers using a standardised eligibility form. As far as possible, full papers or abstracts

describing the same study were linked together, with the article reporting key outcomes designated as the primary publication.

Data were extracted by one reviewer using a standard data extraction form and checked by a second reviewer. At each stage, any disagreements between reviewers were resolved by consensus or, if necessary, by arbitration by a third reviewer.

Titles and abstracts identified by the search strategy for the systematic review of cost-effectiveness were assessed for potential eligibility by two health economists using predetermined inclusion criteria. Full papers were formally assessed for inclusion by one health economist with respect to their potential relevance to the research question.

Quality assessment

The methodological quality and the quality of reporting of the included clinical effectiveness studies were assessed following guidelines by Thomas and colleagues,[73] which were modified to accommodate the types of studies included in this review (see *Appendices 6–10*). Quality criteria were applied by one reviewer and checked by a second reviewer, with any differences in opinion resolved by consensus or by arbitration by a third reviewer.

Quality assessment for the systematic review of cost-effectiveness was based on a checklist for economic evaluation publications[74] and guidelines for good practice in decision-analytic modelling in health technology assessment.[75]

Inclusion and exclusion criteria

Participants

- Adults and children with bilateral deafness were included.
- Single-sided deafness was excluded.
- Studies reporting both bilateral and unilateral hearing loss were included only if the groups were reported separately or if the majority of participants had bilateral hearing loss.

Interventions

- Bone-anchored hearing aids, consisting of a surgically implanted titanium fixture. Devices in current use and devices no longer manufactured were included. BAHAs could be fitted unilaterally or bilaterally.

Comparisons

- Bone-anchored hearing aids versus:
  - conventional hearing aids (ACHA or BCHA)
  - unaided hearing
  - ear surgery (tympanoplasty, myringoplasty, ossiculoplasty, stapedectomy and stapedotomy).
- Unilateral versus bilateral BAHAs.
- Studies comparing different BAHA models were excluded.

Outcomes

- Hearing measures, aided hearing thresholds, speech recognition scores.
- Validated measures of QoL and patient satisfaction.
- Adverse events.

- Measures of cost-effectiveness [i.e. cost per quality-adjusted life-year (QALY), cost per life-year saved] and consequences in terms of health service resources.

Study design

For the systematic review of clinical effectiveness, studies were classified according to the criteria by Thomas and colleagues,[73] with some adaptations to meet the requirements of this review. The following study designs were eligible:

1. randomised controlled trials (RCTs)
2. controlled clinical trials
3. prospective cohort analytic studies (two groups pre and post, i.e. assessments made before and after BAHA surgery in the intervention group and the control group)
4. prospective cohort one-group pre and post studies (no control group, assessments made before and after BAHA surgery)
5. cross-sectional 'audiological comparison studies' [no control group, assessments with intervention and comparator(s) made at one point in time, after BAHA surgery]
6. prospective case series (no comparator condition, outcomes reported with BAHA only).

Where evidence from different types of study design was identified for each of the above comparisons, only studies with the most rigorous designs were included. Where higher level evidence was limited to BAHA models no longer in current use, lower level evidence for models in current use (Divino, Intenso, Cordelle II, BP100, Ponto, Ponto Pro) was considered.

Studies published as abstracts or conference presentations were included only if sufficient details were presented to allow an appraisal of the methodology and the assessment of results to be undertaken.

Only full economic evaluations, those reporting both costs and outcomes, were eligible for inclusion in the systematic review of cost-effectiveness evidence. Conference abstracts were not eligible for inclusion in the cost-effectiveness section.

Data synthesis

Studies of clinical effectiveness and cost-effectiveness were synthesised through a narrative review with full tabulation of the results of all included studies. It was considered inappropriate to combine the results of the studies in a meta-analysis owing to differences in the outcome measures and patient populations. Within *Chapter 3*, results are discussed according to the comparison to aid interpretation. Where studies report outcomes for more than one comparison (e.g. BAHA vs ACHA and BAHA vs unaided), these are discussed in each relevant section. Care should therefore be taken to avoid double-counting the BAHA data, which are repeated. This is noted where appropriate. Outcome measures are discussed throughout the review of clinical effectiveness as reported by the included studies, including the use of descriptions such as 'improvement' or 'deterioration'. To aid interpretation of the data, lower hearing thresholds are considered to be 'better' than higher thresholds, but it is acknowledged that this is a simplistic approach and, while true in many cases, it may not necessarily be so. The methods for the economic model are described in *Chapter 4, Southampton Health Technology Assessments Centre economic analysis*.

Chapter 3

Clinical effectiveness results

Quantity and quality of research available

Searching identified 665 references after de-duplication. The number of references excluded at each stage of the systematic review is shown in *Figure 1*. Selected references which were retrieved but later excluded are listed in *Appendix 3* with reasons for exclusion. Studies were often excluded for more than one reason; the most common reason being study design (16 studies), followed by outcomes (12 studies), intervention (six studies), comparator (10 studies) or participants (one

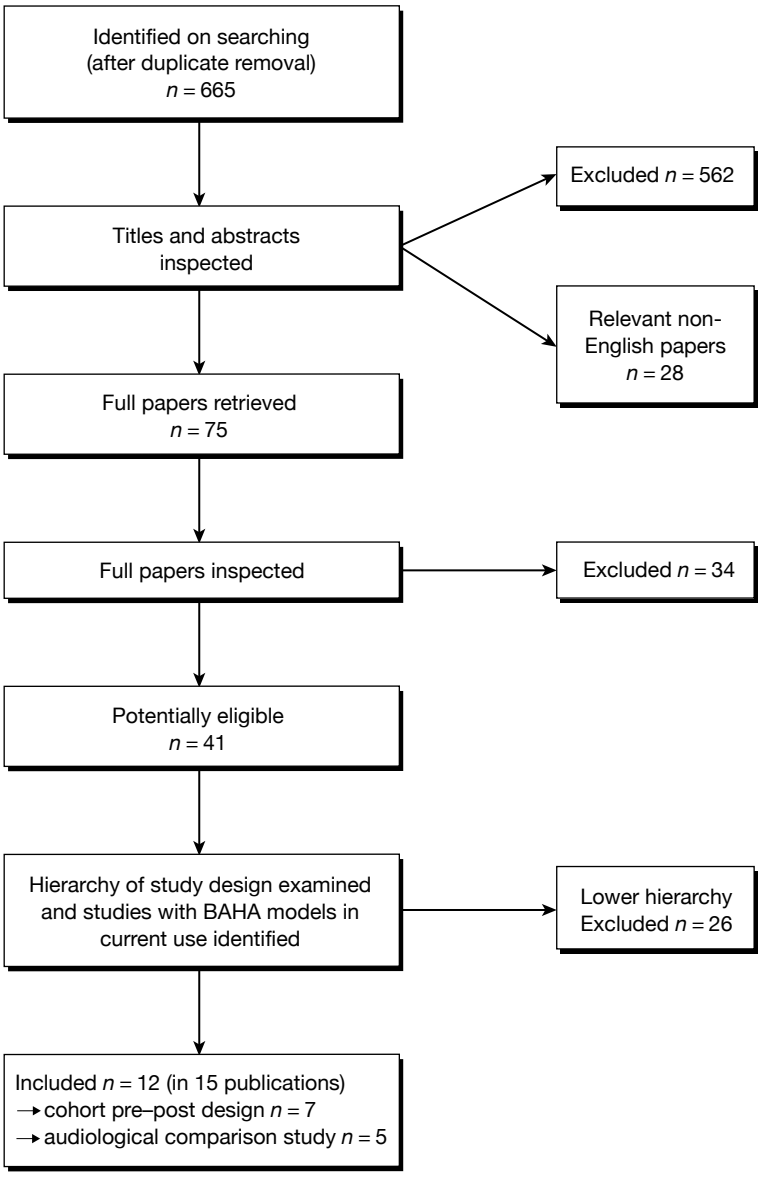

FIGURE 1 Flow chart of identification of studies.

study). Although not formally assessed, the level of agreement between reviewers for screening was good. Twenty-eight relevant non-English references were identified by the searches and can be seen in *Appendix 4*. After examination of the titles and English abstracts (where available) it was unclear whether or not any of these studies met the inclusion criteria, and none appeared to have a concurrent control group. Because it was anticipated the studies would add limited value to the review, and in view of limited resources, translation and full screening of the papers were not undertaken. Searches did not identify any eligible ongoing studies.

Forty-one potentially eligible studies were identified. After selecting the highest level of evidence available for each comparison (BAHA vs BCHA, ACHA, unaided hearing or ear surgery, unilateral vs bilateral BAHA) and checking the remaining studies for BAHA models in current use, 12 studies (in 15 publications) were included in the systematic review of clinical effectiveness.[59,60,66,76–87] The included studies were either one-group cohort pre and post studies or cross-sectional 'audiological comparison' studies (study design is discussed further in *Quality assessment*); no RCTs, controlled clinical trials or prospective cohort analytic studies were identified. Only two studies included BAHA models that are in current use.[66,76] A summary of the highest level of evidence available and the current availability of the BAHAs used (whether or not currently manufactured) for each comparison can be seen in *Table 5*. The remaining 26 lower evidence studies are listed in *Appendix 5*, and were described by reviewers as audiological comparison studies (using BAHAs no longer manufactured, 20 studies) or prospective case series with no comparator (six studies). No eligible studies comparing BAHAs with ear surgery were identified.

TABLE 5 Summary of hierarchy of evidence identified by searches

Comparison	Highest level of evidence identified and current availability of BAHA (no. of studies[a])
BAHA vs BCHA	CPP and BAHA in current use: 0 CPP and BAHA no longer manufactured: 4[77–82] ACS and BAHA in current use: 0 ACS and BAHA no longer manufactured: 14 (see *Appendix 5*)
BAHA vs ACHA	CPP and BAHA in current use: 0 CPP and BAHA no longer manufactured: 5[78–84] ACS and BAHA in current use: 1[76] ACS and BAHA no longer manufactured: 13 (see *Appendix 5*)
BAHA vs unaided	CPP and BAHA in current use: 1[66] CCP and BAHA no longer manufactured: 3[77,78,83] ACS and BAHA in current use: 0 ACS and BAHA no longer manufactured: 6 (see *Appendix 5*)
BAHA vs ear surgery	0 eligible studies
Unilateral vs bilateral	CPP and BAHA in current use: 0 CPP and BAHA no longer manufactured: 0 ACS and BAHA in current use: 0 ACS and BAHA no longer manufactured: 4[59,60,85–87]

ACS, cross-sectional audiological comparison study; CPP, cohort pre and post study (one group before and after).
a Most studies reported more than one comparison.

Characteristics of included studies

BAHAs versus BCHA or ACHA

Study design

Seven studies (one study had three associated publications[79–81]) comparing BAHAs with conventional aids, either BCHAs,[77] ACHAs[76,83,84] or both (in separate subgroups),[78–82] were included (*Table 6*, *Appendices 6–8*). Three of the studies also tested participants unaided[77,78,83] (see *BAHA versus unaided hearing*). Six[77–84] of the studies were described by reviewers as cohort pre- and post-studies (before and after studies) and either assessed BAHAs models that are no longer manufactured[77–83] or did not report the model used (although this study was published in 1998 so is unlikely to have used a BAHA that is currently manufactured[84]). Only one study was identified that assessed a BAHA model in current use;[76] this study compared the BAHA Intenso with an ACHA, and was described by reviewers as a cross-sectional audiological comparison study.

Post-operative assessment with the BAHA was undertaken after either 4–6 weeks[79,80,84] or 6 months.[77,78,82] The post-operative duration was not reported in one study.[83] Participants in the cross-sectional audiological comparison study had ≥ 12 months experience with the BAHA before being assessed with a BAHA and an ACHA at the same time point.[76]

Participants

The study from the Netherlands by Snik and colleagues[79–81] was associated with three eligible publications, which had considerable overlap of participants. It appears that the participants who formed the BAHA HC 200 subgroup ($n = 42$) and the HC 220 subgroup ($n = 16$) in the 1994 study by Snik and colleagues[80] were also reported in the 1998 study (BAHA HC 200, $n = 41$)[81] and the 1992 study (BAHA HC 220, $n = 12$),[79] respectively. Participants from another centre were also included in the 1992 study. To avoid double-counting of participants these three publications have been considered as one study with the appropriate publication referenced when discussed. It is not clear whether there is an overlap with the participants from the studies by Mylanus and colleagues[84] or Hol and colleagues,[82] which were also conducted by the same group in the Netherlands. There may also be some overlap of participants between two of the UK studies, which were undertaken by the Birmingham group, but again this is not clear.[78,83]

The studies comparing BAHAs with conventional aids included participants with inoperable bilateral congenital microtia atresia,[77] otosclerosis,[83] chronic otitis media/externa or otorrhoea,[78,79,81,84] aural atresia[80] and congenital hearing loss.[78] Details of aetiology were not reported in the studies by Flynn and colleagues[76] or Hol and colleagues.[82]

All seven studies[76–84] reported PTA thresholds for AC and/or BC, although the frequencies over which these were determined varied between the studies (see *Table 6*). Mean PTA thresholds for AC varied from 55 dB HL (range 30–90 dB HL) at 0.5, 1.0 and 2.0 kHz in the 1998 publication by Snik and colleagues[81] to 91.1 dB HL [standard deviation (SD) 14.3, range 70–108 dB HL] (frequencies not reported) in the BCHA subgroup of the 1992 publication by Snik and colleagues.[79] Similarly, mean PTA thresholds for BC varied from 16 dB HL (range 0–28 dB HL) at 0.5, 1.0 and 2.0 kHz in the 1998 publication by Snik and colleagues[84] to 49.6 dB HL (SD 7.3, range 40–57 dB HL) (frequencies not reported) in the ACHA subgroup of the 1992 publication by Snik and colleagues.[79]

One study included children only (age 5–17 years)[77] and five studies[76,78,79,82,84] included adults only (mean age from approximately 43[78] to 59[82] years). In the study by Snik and colleagues, two of the publications included children and adults (age range from 10 to 70[81] or 77[80] years), while the 1992 publication[77] included adults only (age range 34–78 years). The proportion of men and women varied between those studies reporting characteristics of the study sample.[76,77,82,84]

These studies are generalisable only to people with bilateral conductive or mixed hearing loss who had previously used either an ACHA or a BCHA.

Outcomes

Data were reported in a variety of ways in the studies. Five of the studies reported pure-tone or warble-tone average thresholds,[76–78,83,84] and one study reported the average difference between warble-tone thresholds.[79] The range of frequencies over which these were assessed varied between the studies, and although in the USA it is mandatory to include 3 kHz, this is not required in Europe. One study did not report any audiological measures at follow-up.[82] Outcomes for speech audiometry included 100% speech audiometry discrimination with background noise at 65 dB HL,[77] speech recognition threshold (level at which 50% of the presented phonemes were repeated properly by the participant)[79] and speech discrimination score at 63 dB.[78,83] One study reported the speech-to-noise ratio with BAHAs and ACHAs,[76] whereas two other studies reported only the change in speech-to-noise ratio[81,84] or the change in speech reception threshold (SRT) in quiet.[81] The maximum phoneme score was reported by two studies.[79,84] One study reported the number of participants with a statistically significant change in the speech recognition in quiet score and speech-to-noise ratio score.[80] Accurate directional identification of location of a sound source was evaluated by one study.[78]

Two studies[77,82] reported using a validated measure of QoL, although limited details and data were reported in one of these.[77] The second study[80] did not report any audiological measures, but presented before and after data from the Short Form questionnaire-36 items (SF-36), European Quality of Life-5 Dimensions (EQ-5D) and the Hearing Handicap and Disability Index (HHDI). Five studies reported the results of subjective questionnaires on patient preference,[77,81,83,84] satisfaction,[78] comfort[79,83] and opinions on speech recognition in noise and quiet,[78–81] but none of these questionnaires appears to have been validated.

In five of the seven studies,[76,78,80,81,83,84] some data were reported only in figures and had to be estimated by reviewers, which increases potential for error. In two of the studies,[79,83] individual patient data without any summary statistics were reported for some outcomes, therefore means and SDs presented in this review for these studies were calculated by reviewers. Data estimated from figures or summary statistics calculated by reviewers are indicated in *Tables 6, 8* and *10*.

Country

The studies by Snik and colleagues,[79–81] Mylanus and colleagues[84] and Hol and colleagues[82] were conducted in the Netherlands, two studies were conducted in the UK,[78,83] and one study was conducted in each of Sweden[76] and Mexico.[77]

Funding

Two of the studies stated that they were funded by non-commercial organisations (public bodies),[77,79–81] four did not report funding[78,82–84] and one was funded by Cochlear Bone Anchored Solutions.[76]

BAHA versus unaided hearing

Four studies included a comparison of BAHAs with unaided hearing (see *Table 6* and *Appendices 6–9*).[66,77,78,83] Three of these studies have been described above, as they compared BAHAs with conventional aids but also reported outcomes unaided. The BAHAs used in these studies are no longer manufactured. Audiological assessment was undertaken 6 months post-operatively in two studies,[77,78] but this was not reported in the other study.[83]

A study by Kompis and colleagues[66] was identified that compared BAHAs with the unaided condition and included a BAHA model in current use (BAHA Divino). This study is defined

TABLE 6 Characteristics of included studies

Study	Intervention and timing of audiology	Participants indication and characteristics
BAHAs vs BCHA		
Béjar-Solar *et al.* 2000[77] Mexico Cohort pre–post	One group: $n=11$ 1. Unaided (pre-op) 2. BCHA (pre-op) 3. BAHA Classic 300 (at 6 months)	Inoperable bilateral congenital microtia atresia. BC PTA ≥ 45 dB HL with 100% speech discrimination. Low socioeconomic background *Age, mean years (range)*: 10 (5–17) *Sex (M:F)*: 7:4 *PTA thresholds (1.25–3.00 kHz), mean, dB HL*: AC right ear 69, left ear 69; BC right ear 20, left ear 14; sound field PTA 64
BAHA vs ACHA		
[a]Burrell *et al.* 1996[83] UK Cohort pre–post	One group: $n=9$ 1. ACHA (pre-op) 2. Unaided (unclear if pre- or post-op) 3. BAHA Superbass and ear level BAHA, model NR (time of assessment NR)	Otosclerosis. Average BC thresholds (0.5–4.0 kHz) < 40 dB HL for ear level BAHA, < 60 dB HL for body-worn Superbass *Age, mean years (range)*: NR for study sample *Sex (M:F)*: NR for study sample *Average BC thresholds (0.5–4.0 kHz), dB HL*: NR for study sample
Flynn *et al.* 2009[76] Sweden Audiological comparison study	One group: $n=10$ 1. BAHA Intenso 2. ACHA Oticon Sumo DM (digital superpower hearing aid) Assessments at same session	Mixed hearing loss, no further details. Sensorineural component ≥ 25 dB HL plus air-bone gap > 30 dB *Age, mean years (range)*: 59 (32–75) *Sex (M:F)*: 5:5 *PTA thresholds (0.5, 1.0 and 2.0 kHz), mean dB HL (range)*: AC 77 (55–80); BC 41 (25–66)
[c]Mylanus *et al.* 1998[84] Netherlands Cohort pre–post	One group: $n=34$ 1. ACHA (pre-op) 2. BAHA, model NR (4–6 weeks post-fitting)	Bilateral conductive or mixed hearing loss with chronic otitis. No audiological criteria stated *Age, mean years (range)*: 48 (26–72) *Sex (M:F)*: 12:22 *PTA thresholds (0.5, 1.0, 2.0 and 4.0 kHz), mean dB HL (range)*:[d] AC 60 (25–90); BC 26 (6–46)
BAHA vs BCHA and ACHA (in separate subgroups)		
[a]Cooper *et al.* 1996[78] UK Cohort pre–post	Four subgroups [previous aid AC or BC, aetiology congenital (CON) or CSOM]: 1. Unaided[b] 2. Previous aid: (i) ACHA ($n=33$); (ii) BCHA ($n=35$), (pre-op) 3. BAHA HC 200, 300, 220 (at 6 months post-fitting)	CSOM or congenital aetiology. Average BC thresholds (0.5–4.0 kHz) < 40 dB HL (ear level) or < 60 dB HL (body-worn), speech discrimination score ≥ 60% *Age, mean years*:[e] CSOM/ACHA 58; CSOM/BCHA 61; CON/ACHA 30; CON/BCHA 24 *Sex (M:F)*: NR *PTA thresholds (0.5–4.0 kHz), mean dB HL*:[e] AC: CSOM/ACHA 58, CSOM/BCHA 65, CON/ACHA 70, CON/BCHA 60; BC: CSOM/ACHA 24, CSOM/BCHA 30, CON/ACHA 20, CON/BCHA 13
[c]Hol *et al.* 2004[82] Netherlands Cohort pre–post	Two subgroups (previous aid AC or BC): 1. Previous aid: (i) ACHA ($n=36$); (ii) BCHA ($n=20$), (pre-op) 2. BAHA Classic (51) or Cordelle (5) (at 6 months)	Acquired conductive or mixed hearing loss, no further details *Age, mean years (range)*: ACHA 47.9 (24–73), BCHA 62 (42–82) *Sex (M:F)*: ACHA 12:24; BCHA 9:11 *PTA thresholds (0.5, 1.0 and 2.0 kHz), mean dB HL (range)*: AC: ACHA 63.2 (30–103), BCHA 76.5 (40–107); BC: ACHA 26.8 (9–51), BCHA 43.4 (17–63)

continued

by reviewers as a pre and post cohort study, but differs from the other included studies as the participants had prior experience with BAHAs when tested unaided at baseline. The aim of the study was to compare the BAHA Divino with the BAHA Compact (this comparison did not meet the inclusion criteria of the review), and the participants had at least 2 years' experience with a BAHA Compact or Classic 300 prior to the study. However, the study also assessed the unaided condition at baseline and then assessed the BAHA Divino after 3 months' use, so these data were included in the systematic review (see *Appendix 9*).

TABLE 6 Characteristics of included studies *(continued)*

Study	Intervention and timing of audiology	Participants indication and characteristics
[c]Snik *et al.* 1992,[79] 1994,[80] 1998[81] Netherlands Cohort pre–post	Two subgroups (previous aid AC or BC):[79] 1. Previous aid: (i) BCHA ($n=7$); (ii) ACHA ($n=5$), (pre-op) 2. BAHA HC220 (at least 4 weeks)	Recurrent otorrhoea. Severe mixed hearing loss with sensorineural components of 45–60 dB HL[79] *Age, mean (SD, range)*: (i) 60.6 (18.8, 34–84) years; (ii) 62 (13.9, 46–78) years *Sex (M:F)*: NR *PTA thresholds (frequencies NR), mean dB HL (SD, range)*:[f,g] AC (i) 91.1 (14.3, 70–108), (ii) 84.8 (12.3, 72–100); BC (i) 46.2 (12.6, 28 to >62), (ii) 49.6 (7.3, 40–57)
	Four subgroups (previous aid AC or BC, current BAHA HC200 or HC220):[80] 1. Previous aid: (i) BCHA ($n=44$); (ii) ACHA ($n=14$), (pre-op) 2. BAHA: (i) HC200; (ii) HC220 (≥4 weeks)	Chronic otitis media/externa, aural atresia. Both normal to moderate and more severe SNHL[80] *Age, range*: 10–77 (mean NR) years *Sex (M:F)*: NR *PTA thresholds (0.5, 1.0 and 2.0 kHz) for BC, range dB HL*: HC 200 ($n=42$) 0–44; HC 220 ($n=16$) 33–63
	Two subgroups:[81] 1. Previous aid: (i) BCHA ($n=33$); (ii) ACHA ($n=8$), (pre-op) 2. BAHA HC 200 (at least 6 weeks)	Conductive or mixed binaural hearing loss, SNHL of ≤30 dB HL. No details of aetiology[81] *Age, range*: 10–70 (mean 43) years *Sex (M:F)*: NR *PTA thresholds (0.5, 1.0, 2.0 kHz), mean dB HL (range)*: AC 55 (30–90); BC 16 (0–28)
BAHA vs unaided (see also three studies from above: Béjar-Solar* et al. *2000,[77] Burrell* et al. *1996[83] and Cooper* et al. *1996[78])		
Kompis *et al.* 2007[66] Switzerland Cohort pre–post	One group: $n=7$ 1. Unaided (at baseline) 2. BAHA Divino (at 3 months)	Bilateral CHL, some mild-to-moderate SNHL. No further details. All had at least 2 years' experience with BAHAs *Age, mean (range)*: 49 (19–66) years *Sex (M:F)*: 3:4 *PTA AC and BC thresholds*: IPD presented in figure but could not be extracted
Unilateral vs bilateral BAHAs		
Bosman *et al.* 2001[60,85] Netherlands Audiological comparison study	One group: $n=25$ 1. Unilateral (first implant side) 2. Bilateral (HC 200 or Classic 300) Assessments at same session	Recurrent otorrhoea, otitis externa, congenital atresia *Age, mean (range)*: 44.3 (12–74) years *Sex (M:F)*: 14:11 *BAHA experience, mean (range)*:[f] unilateral 49.1 (18–105) months; bilateral 13.6 (3–105) months *PTA thresholds (0.5, 1.0 and 2.0 kHz), mean dB HL (SD, range)*: AC first fitted side 59.5 (13.7, 32–82),[f] second fitted side 63.6 (10.9, 38–82);[f] BC first fitted side 21.0 (10.7, –5 to 36);[g] second fitted side 21.9 (12.4, –8 to 48)[g]
Dutt *et al.* 2002[86] UK Audiological comparison study	One group: $n=11$ 1. Unilateral (best response, or R and L) 2. Bilateral (Compact) Assessments at same session	Treacher Collins syndrome, Goldenhar syndrome, bilateral: mastoid cavities, CON, chronic otitis media, microtia, acquired otosclerosis *Age, mean (range)*:[f] 42.3 (22–54) years *Sex (M:F)*: 3:9 (one patient chose not to participate) *BAHA experience, mean (range)*:[f] unilateral 6.3 (3–12) years; bilateral 2.2 (1–5) years PTA AC and BC thresholds: NR
Priwin *et al.* 2004[87] Sweden Audiological comparison study	One group: $n=12$ 1. Unilateral (best and shadow side) 2. Bilateral (Compact and Classic 300) Assessments at same session	Chronic otitis, otosclerosis, congenital ear canal atresia *Age, mean (range)*:[f] 51.7 (27–68) years *Sex (M:F)*: 3:9 *BAHA experience, mean (range)*:[f] unilateral 14.3 (5.8–21) years; bilateral 6.8 (1–19.6) years *PTA thresholds (0.5, 1.0 and 2.0 kHz), mean dB HL (SD, range)*:[f,g] AC first fitted side 58.3 (15.3, 38–87), second fitted side 59 (20.7, 27–102); BC first fitted side 29.8 (15.2, 8–53),[h] second fitted side 30.9 (13.4, 7– 50)

TABLE 6 Characteristics of included studies *(continued)*

Study	Intervention and timing of audiology	Participants indication and characteristics
Priwin *et al.* 2007[59] Sweden Audiological comparison study	Two groups: 1. Unilateral (unaided and 1 BAHA), $n=3$ 2. Bilateral (unaided, 1 and 2 BAHAs), $n=6$ (Compact and Classic) Assessments at same session	Majority had symmetrical maximal or near-maximal conductive bilateral hearing loss *Age, mean (range)*: 11.3 (6–17) years *Sex (M:F)*: 3:6 *BAHA experience*: at least 3 months (no further details) *PTA (M4) thresholds (0.5, 1.0, 2.0 and 4.0 kHz) mean dB HL (SD)*: AC better ear 61.3 (15.5), worse ear 72.1 (12.1); BC better ear 14.1 (12.7); worse ear 13.8 (10.7)

CON, congenital hearing loss; CSOM, chronic suppurative otitis media; F, female; IPD, individual patient data; L, left; M, male; NR, not reported; R, right.
a There may be overlap of participants between these studies conducted in the UK (see *BAHAs versus BCHA or ACHA*).
b Unaided condition was assessed pre- and post-operatively and it is not clear which of these data are presented.
c There may by overlap of participants between these studies conducted in the Netherlands (see *BAHAs versus BCHA or ACHA*).
d Ear ipsilateral to the side of implantation, always ear with best cochlear reserve.
e Data estimated from figure by reviewer.
f Means calculated by reviewer.
g SD calculated by reviewer.
h In three patients, at one or more frequency, no fixed value was attained, highest measurable value used for mean.
See *Appendices 6–10* for further details.

Participants

Kompis and colleagues[66] included participants described as having substantial bilateral CHL, some combined with mild-to-moderate SNHL. No details of aetiology were reported. PTA thresholds for AC and BC for each ear of individual participants were presented in figures in this study, but data could not be extracted. The participants in this study were adults with a mean age of 48.6 years.[66] The studies by Burrell and colleagues,[83] Cooper and colleagues[78] and Béjar-Solar and colleagues[77] have already been described in *BAHAs versus BCHA or ACHA*.

The generalisability of the study by Kompis and colleagues[66] may be limited to people with bilateral conductive or mixed hearing loss with previous experience of BAHAs.

Outcomes

Kompis and colleagues[66] reported the average improvement in sound field pure-tone thresholds over all frequencies compared with unaided. Outcomes for speech audiometry included mean speech recognition threshold and speech recognition scores in quiet, and speech recognition in noise when noise was presented from the front or back. QoL was not reported. Outcomes reported by Burrell and colleagues,[83] Cooper and colleagues[78] and Béjar-Solar and colleagues[77] have already been described in *BAHAs versus BCHA or ACHA*.

Data had to be estimated from figures by reviewers for the study by Kompis and colleagues;[66] this is indicated in *Tables 6* and *12*.

Country

The study by Kompis and colleagues[66] was conducted in Switzerland and, as described in *BAHAs versus BCHA or ACHA*, two studies were conducted in the UK[78,83] and one in Mexico.[77]

Funding

The BAHA Divinos used in the study by Kompis and colleagues[66] were provided by Entific Medical Systems. The other three studies are reported in *BAHAs versus BCHA or ACHA*.[77,78,83]

Unilateral versus bilateral BAHAs

Four studies[59,60,85–87] comparing unilateral and bilateral BAHAs were included, and all were described as audiological comparison studies by reviewers (see *Table 6*, *Appendix 10*). None of these studies (or any eligible study from a lower level of evidence) compared unilateral and bilateral BAHAs using a model in current use.

Participants

The studies comparing unilateral with bilateral BAHAs included participants with various diagnoses, including recurrent otorrhoea,[60] chronic otitis,[60,86,87] congenital atresia,[60,87] otosclerosis,[86,87] congenital syndromes and hearing loss,[86] mastoid cavities[86] and microtia.[86] One study did not describe aetiology, simply stating that the majority of participants had symmetrical maximal or near-maximal conductive bilateral hearing loss.

Three of the four studies reported PTA thresholds (of frequencies 0.2, 1.0 and 2.0 kHz[60,87] or 0.5, 1.0, 2.0 and 4.0 kHz[59]) for AC and BC. Mean PTA thresholds for AC were ~ 60 dB HL in these studies, and mean PTA thresholds for BC ranged from ~ 14 dB HL[59] to ~ 30 dB HL.[87]

Some of the participants in the study by Bosman and colleagues[60] had previously used a unilateral BCHA and had been deprived of binaural cues since early life,[85] whereas participants in the study by Dutt and colleagues[86] were required to have previous experience of binaural hearing. Previous experience of binaural hearing was not explicitly stated by the other two studies.[59,87]

Implantation of the bilateral BAHAs was not simultaneous in three of the studies;[60,85–87] the participants in two of these studies consisted of self-selected volunteers applying for a second BAHA.[60,86] Participants had on average 4.1[60] to 14.3[87] years experience with unilateral BAHAs, and 1.1[60] to 6.8[87] years experience with bilateral BAHAs at the time the studies were conducted. It is not clear whether bilateral implantation was simultaneous or sequential in the fourth study, which simply stated that participants had at least 3 months' experience with BAHAs.[60]

One study included children only (mean age 11.3 years, range 6–17 years)[59] and two studies included adults only [mean age 42.3 years (SD 10 years)[86] and 51.7 years (SD 13.3 years)[87]]. Bosman and colleagues[60] included both adults and children, with a mean age of 44.3 years (SD 16.3 years, range 12–74 years). Three studies had a higher proportion of women than of men.[59,86,87]

Three of these studies[60,85–87] are generalisable only to people with bilateral conductive or mixed hearing loss and previous experience with unilateral and bilateral BAHAs. The fourth study is relevant to a paediatric population who have at least 3 months' experience with unilateral and/or bilateral BAHAs.[59]

Outcomes

Two of the four studies reported data on sound field tone thresholds; one of these reported data separately for unilateral and bilateral BAHAs where sound was presented from the front[59] and one reported average improvement with bilateral BAHAs where sound was presented from the front, at best side, at shadow side and from behind the participant.[87] Outcomes for speech audiometry included SRT in quiet;[60,87] SNR with noise from baffle and shadow side;[60] change in the SNR with bilateral BAHAs with noise at best side, shadow side and as surrounding noise;[87] scores for speech in quiet at a range of intensity levels;[86] and speech-in-noise scores with noise presented at front, left and right.[86] Three studies reported directional hearing, including correct localisation,[59,60] localisation within 30°[60,87] and correct lateralisation.[59,60] The set-up of the loudspeakers for the assessment of directional hearing was different in each of the three studies, using either five loudspeakers positioned at 45° intervals in a frontal semicircle,[59] seven or nine loudspeakers positioned at 30° intervals in an arc spanning 180° or 240° with a 1-m radius,[60] or

using 12 loudspeakers positioned at 30° in a circle with a 1-m radius[87] (see data extraction forms in *Appendix 10* for further details). The 'binaural masking level difference' was reported by two studies in an attempt to demonstrate the existence of binaural hearing (defined as the ability to use binaural cues, i.e. use the different sound information at the two cochleae to improve hearing) with bilateral BAHAs.[60,87] In this test, a pure-tone signal is presented in noise, and the task is to detect the tone. Three conditions were tested: for the first condition, the pure-tone signal and noise were presented equally at both ears; for the second condition, the phase of the tones presented at the two sides had an opposite phase (180° out of phase), but the noises were in phase (the levels were equal at both sides); and for the third condition, the noises at both sides were 180° out of phase, but the tones were in phase.

One study[59] reported data from the validated Meaningful Auditory Integration Scale (MAIS) and Meaningful Use of Speech Scale (MUSS)[45] to assess hearing skills in 'meaningful, real world situations', and the International Outcome Inventory for Hearing Aids (IOI-HA)[88] to assess hearing aid outcomes.

All four studies[59,60,85–87] presented individual patient data without any summary statistics for some outcomes, while other outcomes were presented in figures only. Data estimated from figures or summary statistics calculated by reviewers are indicated in *Tables 6* and *13*.

Country

Two studies were conducted in Sweden[59,87] and one study was conducted in each of the Netherlands[60,85] and the UK.[86]

Funding

Two of the studies stated that they were funded by non-commercial organisations (public bodies),[59,87] one did not report funding[86] and one was funded by Entific Medical Systems.[60]

Quality assessment

All 12 included studies were rated overall as 'weak' for their methodological quality and quality of reporting (*Table 7*).[59,60,66,76–87] The studies were not described using recognised study types or descriptions, and therefore were termed 'cohort pre and post studies' (seven studies[66,77–84]) or cross-sectional 'audiological comparison' studies (five studies[59,60,76,85–87]) by reviewers. In the cohort pre and post studies, baseline measurements were undertaken with the individual's previous aid or unaided or both, *before* BAHA surgery, and measurements with the BAHA were undertaken after a given period of use. In the audiological comparison studies, the measurements were undertaken with the intervention and comparator at the same point in time. With both of these designs, a potential source of bias is that, as with any hearing aid trial, participants are likely to prefer the second hearing aid tested.[89,90] As the study design method was not described by any of the included studies, all were assigned a rating of 'weak', although in the hierarchy of evidence, cohort pre and post studies are ranked higher than audiological comparison studies.

Six studies were rated as 'moderate' for selection bias,[60,81–84,87] indicating that the selected individuals are at least somewhat likely to be representative of the target population, and at least 60% of those identified agreed to participate. The remaining six studies were rated as 'weak' for selection bias,[59,66,76–78,86] as participants may not be representative of the target population if they are self-referred, or because selection method or the level of participation were not described.

Although some studies described some baseline characteristics of the participants, none of the included studies reported these as potential confounding variables or described how these may be causally related to the outcomes of interest. All 12 studies were therefore rated as 'weak' for the assessment of potential confounders.

TABLE 7 Summary of ratings of methodological quality

Study	Selection bias	Study design (description)	Confounders	Blinding	Data collection methods	Withdrawals and dropouts	Intervention integrity	Analysis appropriate to question?		Global rating
								Appropriate statistical methods?	Handling of missing data reported?	
BAHAs vs BCHA										
Béjar-Solar *et al.* 2000[77]	Weak	Weak (CPP)	Weak	Weak	Strong	Weak	No	No	No	**Weak**
BAHA vs ACHA										
Burrell *et al.* 1996[83]	Moderate	Weak (CPP)	Weak	Weak	Strong	Weak	No	No	No	**Weak**
Flynn *et al.* 2009[76]	Weak	Weak (ACS)	Weak	Weak	Strong	Weak	No	Can't tell	No	**Weak**
Mylanus *et al.* 1998[84]	Moderate	Weak (CPP)	Weak	Weak	Strong	Weak	Can't tell	Yes	No	**Weak**
BAHA vs BCHA/ACHA										
Cooper *et al.* 1996[78]	Weak	Weak (CPP)	Weak	Weak	Strong	Weak	Yes	Yes	No	**Weak**
Hol *et al.* 2004[82]	Moderate	Weak (CPP)	Weak	Weak	Strong	Weak	Yes	Yes	Yes	**Weak**
Snik *et al.* 1998[79–81]	Moderate	Weak (CPP)	Weak	Weak	Strong	Strong	Can't tell	Can't tell	No	**Weak**
BAHA vs unaided (see also three studies from above: Béjar-Solar* et al. *2000,[77] Burrell* et al. *1996[83] and Cooper* et al. *1996[78])										
Kompis *et al.* 2007[66]	Weak	Weak (CPP)	Weak	Weak	Strong	Weak	No	Yes	No	**Weak**
Unilateral vs bilateral BAHAs										
Bosman *et al.* 2001[60,85]	Moderate	Weak (ACS)	Weak	Weak	Strong	Weak	Can't tell	No	No	**Weak**
Dutt *et al.* 2002[86]	Weak	Weak (ACS)	Weak	Weak	Strong	Strong	Can't tell	No	No	**Weak**
Priwin *et al.* 2004[87]	Moderate	Weak (ACS)	Weak	Weak	Strong	Weak	Can't tell	Yes	No	**Weak**
Priwin *et al.* 2007[89]	Weak	Weak (ACS)	Weak	Weak	Strong	Weak	No	Yes	No	**Weak**

ACS, cross-sectional 'audiological comparison study', reviewer's description; CPP, cohort pre and post (one group, before and after) design, reviewer's description.
Global rating: strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.[71] Further details on these criteria for each study can be seen in the data extraction forms in *Appendices 6–10*.

It was assumed that outcome assessors were not blinded to the intervention as this was not reported by any of the studies. This can lead to bias in the care provided (performance bias) and how the outcomes are assessed (measurement or detection bias). Participants were blinded to the use of unilateral or bilateral BAHAs in one study;[87] however, it was assumed that participants were not blinded to the research question in the remaining studies, leading to reporting bias. While it may be difficult to blind participants to the intervention, it is important to note that this bias is present when interpreting the results.

All 12 studies were rated as strong for data collection methods. The 11 studies that reported audiometric data all used methods known to be valid and reliable; however, some of these studies also reported self-reported subjective outcomes using methods that are not valid and reliable and are therefore not discussed in the systematic review (but can be viewed in *Appendices 6–10*). The study by Hol and colleagues[82] used QoL measures that are known to be valid and reliable.

Two studies were rated 'strong' for withdrawals and dropouts,[81,86] as they described both the numbers and reasons for withdrawals and dropouts, and had a follow-up rate of ≥ 80%. This information could not be deduced from the remaining studies, which may therefore be at risk from attrition bias.

Intervention integrity describes whether all participants received the intervention in the same way, for example the same frequency or duration of use. Only two studies[78,82] reported the amount of use with the BAHA (such as number of hours per day), five studies reported that the BAHA was used 'daily' with no further description,[60,79–81,84,86,87] and the remaining five studies did not report how often the BAHA was used during the study period.[59,66,76,77,83]

Statistical analysis of the results was not undertaken by four studies.[60,77,83,86] The remaining eight studies reported statistical analyses, six of which were judged appropriate to the research question[59,66,78,82,84,87] and in two studies this was not clear.[76,81] Only one of the studies reported on how missing data were dealt with in the analysis.[82]

Assessment of clinical effectiveness: BAHA versus BCHA

Audiological measures

Three included studies (reported in five publications)[77–81] provided a comparison of audiological measures between BAHA and BCHA (*Table 8*). All three studies were cohort pre and post studies and none used BAHA models that are in current use. The high risk of bias in all three included studies should be considered when interpreting the results.

Audiometry

All three included studies reported data on hearing threshold tests, two[78,79] used sound field warble-tone thresholds and the remaining study used sound field pure-tone thresholds (see *Table 8*).[77]

Béjar-Solar and colleagues[77] report meaned sound field PTA thresholds in their 11 participants (at 0.125–3.000 kHz) with inoperable bilateral congenital microtia atresia. The study found a 37% improvement with the Classic 300 BAHA compared with the BCHA (BCHA 30 dB HL, BAHA 19 dB HL), although statistical analysis was not undertaken.

Cooper and colleagues[78] tested two subgroups: 19 participants who had chronic suppurative otitis media and 16 participants who had congenital hearing loss. Mean sound field warble-tone threshold (over a 0.5–4.0 kHz range) was reported to be statistically significantly better with

TABLE 8 Audiologic measures: BAHA versus BCHA

Study and outcomes	BCHA	BAHA model	Comparison
***Béjar-Solar* et al. *2000*[77] (n = 11)**	***BCHA***	***Classic 300***	***Difference***
Sound field PTA (125–3000 Hz), dB HL	30	19	–11 (37% improvement)
Sound field 100% speech audiometry discrimination, background noise at 65 dB, dB HL	62	48	–14 (23% improvement)
Accurate directional identification of location of a sound source (% of cases)	0	80	
Cooper* et al. *1996*[78] *two subgroups: CSOM (n = 19), CON (n = 16)	***BCHA***	***HC 200, 300, 220***	**p-*value***
Mean sound field warble-tone thresholds, dB [dB(A), 500–4000 Hz)][a]	CSOM 42	CSOM 35	$p < 0.01$
	CON 31	CON 26	$p < 0.01$
Mean sound field speech discrimination score (at 63 dB),% correct[a]	CSOM 65	CSOM 72	p = NS
	CON 86	CON 85	p = NS
***Snik* et al. *1992,*[79] *1994,*[80] *1998*[81]**	***BCHA***	***HC 200, 220***	**p-*value***
Snik et al. *1992*[79] *(n = 7)*	*BCHA*	*HC 220*	
Maximum phoneme score,%, mean (SD) range[b]	36.1 (28.9), 0–85	48.7 (31.7), 0–100	NR
Speech recognition threshold, dB(A), mean (SD) range[b]	(*n* = 2)	(*n* = 4)	NR
	40 (7.1), 35–45	38.8 (11.1), 25–50	
Average difference between the sound field warble thresholds (BCHA minus BAHA), dB[a,c]			
250 Hz	2		
500 Hz	3		
1000 Hz	–2		
2000 Hz	–10		
4000 Hz	–14		
8000 Hz	NR		
Snik et al. *1994*[80] *(n = 44)*	*HC 220, HC 200*		
Patients with a statistically significant change in:	*HC 220 (n = 11)*		
speech recognition in quiet score with BAHA[d]	Improved: 6 of 11 (54%); deteriorated: 0 of 11		
speech-to-noise ratio score with BAHA[d]	Improved: 5 of 11 (44%); deteriorated: 0 of 11		
Patients with a statistically significant change in:	*HC 200 (n = 33)*		
speech recognition in quiet score with BAHA[d]	Improved: 4 of 33 (12%); deteriorated: 0 of 33		
speech-to-noise ratio score with BAHA[d]	Improved: 20 of 33 (60%); deteriorated: 0 of 33		
Snik et al. *1998*[81] *(n = 33)*	*HC 200*		
Change in SRT in quiet (BCHA minus BAHA), mean dB (SD)	2.7 (4.4), improvement $p < 0.05$		
Change in speech-to-noise ratio, mean dB (SD)	2.5 (2.2), improvement $p < 0.05$		

CON, congenital hearing loss; CSOM, chronic suppurative otitis media; NR, not reported; NS, not significant.
a Data estimated by reviewer from figure.
b Means and SDs calculated by reviewer.
c Note that the legend on this figure[77] appears to be incorrectly labelled.
d Percentages reported in paper, numerator calculated by reviewer. There appears to be a slight rounding error in paper.

the BAHA than with the BCHA in the CSOM subgroup [42 dB(A) BCHA vs 35 dB(A) BAHA, $p < 0.01$]. In the CON subgroup the BAHA was also seen to be statistically significantly better than the BCHA [31 dB(A) BCHA vs 26 dB(A) BAHA, $p < 0.01$]. In this study the BAHA tested was one of the HC 200, 300 or 220.

Snik and colleagues[79] presented data from seven participants using the HC 220 on the average difference between the sound field warble thresholds (BCHA minus BAHA) across a range of frequencies, where tones were presented from the front through a loudspeaker. At 0.25 and 0.50 kHz, positive values indicate that hearing was better with the BCHA; at 1, 2 and 4 kHz, negative values indicate that hearing was better with the BAHA. The study did not report the results for testing at 8 kHz. No statistical analysis was undertaken.

Speech audiometry

All three[77–81] included studies provide details of speech discrimination using BCHA and BAHAs (see *Table 8*). Tests varied between the studies and are discussed in turn below.

Béjar-Solar and colleagues[77] reported sound field 100% speech audiometry discrimination at a background noise of 65 dB. The authors adapted a test by Håkansson and colleagues, using colloquial language common to Mexico City and sentences with a high degree of difficulty. This showed a 23% improvement with the BAHA compared with the BCHA (BCHA 62 dB HL, BAHA 48 dB HL, difference –14). In addition, accurate directional identification of the location of a sound was demonstrated in 80% of cases with the BAHA (0% with the BCHA).

Cooper and colleagues[78] reported mean sound field speech discrimination scores, using Boothroyd word lists undertaken at 63 dB, for their two subgroups. The proportion of correct scores was not statistically significant between BCHA and BAHA in the chronic suppurative otitis media subgroup ($n = 19$; BCHA 65% vs BAHA 72%) or in the congenital hearing loss group ($n = 16$; BCHA 86% vs BAHA 85%).

Snik and colleagues[79] reported both the maximum phoneme score and the speech recognition threshold (presentation level at which 50% of presented phonemes were repeated properly by the individual), which were determined using standard Dutch phonetically-balance (PB) word lists consisting of 10 monosyllables. The study showed that the maximum phoneme score was better with the BAHA HC 220 [48.7% (SD 31.7)] than with the BCHA [36.1% (SD 28.9)] although no statistical significance testing was undertaken and the sample size was small ($n = 7$). The mean speech recognition threshold in four participants with the BAHA HC 220 [38.8 (SD 11.1)] was similar to that with the BCHA in two participants [40.0 (SD 7.1)]; however, as noted previously, no statistical significance testing of these differences was undertaken. Two later publications reported additional outcomes and also a group of participants using the BAHA HC 200.[80,81] In the 1994 publication,[80] the proportion of participants with a statistically significant improvement or deterioration with the BAHA in speech recognition in quiet and speech-to-noise ratio was reported. The speech recognition in quiet value in this study was the maximum phoneme score obtained using standard phonetically balanced lists of monosyllables, presented at 60, 70 and 80 dB. The speech-to-noise ratio was the difference between the SRT (established with an adaptive procedure, using the test described by Plomp and Mimpen[91,92] with lists of 13 sentences) and the steady-state, speech-shaped noise presented at a fixed level of 65 dB). The study reports results separately for those using the HC 200 ($n = 33$) BAHA and those using the HC 220 ($n = 11$). This showed that four participants (12%) with the HC 200 and six (54%) with the HC 220 improved on their speech recognition in quiet score. For speech-to-noise ratio, 20 participants (60%) with the HC 200 and five (44%) with the HC 220 improved. No participants had a statistically significant deterioration in either outcome with the HC 200 or 220. No further details were reported. In the 1998 publication,[81] a statistically significant improvement with the BAHA HC 200 ($n = 33$) was seen in mean SRT in quiet [2.7 dB (SD 4.4 dB), $p < 0.05$] and speech-to-noise ratio [2.5 dB (SD 2.2 dB), $p < 0.05$] compared with the BCHA. This publication reported using the tests as described in the 1994 publication above.[80]

Self-reported measures

Béjar-Solar and colleagues[77] reported QoL using a validated measure described as the Coop/Dartmouth test.[93] However, limited details were reported; the authors simply stated that 'results uniformly showed the response "hardly could have done better" (options: hardly could have done better, pretty good, indifferent, pretty bad, hardly could have done worse)'. They also stated that physical and emotional condition was reported as very improved. Given that limited details were reported, these results should be interpreted with caution.

Hol and colleagues[82] assessed QoL using a BCHA prior to BAHA surgery and again after 6 months' experience with a BAHA. There were no statistically significant differences found by the SF-36 or EQ-5D (*Table 9*). The SF-36 showed increased emotional problems [indicated by a decrease in role limitations (emotional) score, $p = 0.19$], more pain experienced ($p = 0.3$) and improved mental health (p = not significant) with the BAHA, but the clinical effects were small (0.33, 0.24 and –0.36, respectively). Scores from the EQ-5D suggested that participants were slightly less mobile ($p = 0.26$) and experienced more pain/discomfort ($p = 0.26$) with the BAHA, but again the effect sizes were small (–0.30 and –0.28, respectively). However, statistically significant improvements in disability ($p < 0.01$) and handicap ($p < 0.01$) were found with the BAHA by the HHDI, and effect sizes indicated a large clinical impact (1.42 and 0.79, respectively).

TABLE 9 Self-reported measures: BAHA versus BCHA

Study and outcomes	BCHA	BAHA model: Classic, Cordelle	Mean difference	Effect size[a]
***Hol* et al. *2004*[82] (n=20)**				
SF-36 mean (SD)				
Physical functioning	69.2 (25.4)	70.8 (24.6)	1.4 (p=NS)	–0.06
Role limitations (physical)	61.3 (40.1)	57.5 (45.2)	–3.8 (p=NS)	0.09
Role limitations (emotional)	76.7 (39.1)	63.3 (41.8)	–13.4 (p=0.19)	0.33
Vitality	60.8 (16.6)	61.0 (21.9)	0.2 (p=NS)	–0.01
Mental health	68.4 (17.6)	74.2 (14.2)	5.8 (p=NS)	–0.36
Social functioning	80.6 (17.9)	82.2 (18.3)	1.6 (p=NS)	–0.09
Pain	73.8 (20.0)	67.9 (27.9)	–5.9 (p=0.30)	0.24
General health	61.0 (19.8)	59.5 (20.3)	–1.5 (p=NS)	0.07
EQ-5D mean (SD)				
Five domains (score 1–3)				
Mobility	1.35 (0.49)	1.50 (0.51)	0.15 (p=0.26)	–0.3
Self-care	1.20 (0.41)	1.10 (0.31)	–0.10 (p=NS)	0.28
Usual activities	1.60 (0.68)	1.55 (0.60)	–0.05 (p=NS)	0.08
Pain/discomfort	1.70 (0.57)	1.85 (0.49)	0.15 (p=0.26)	–0.28
Anxiety/depression	1.26 (0.45)	1.20 (0.41)	–0.06 (p=NS)	0.13
Utility (score 0–1)	0.71 (0.23)	0.70 (0.19)	–0.01	0.05
Visual analogue scale (0–100)	74.0 (16.0)	72.4 (17.4)	–1.6	0.10
HHDI mean (SD)				
Disability	31.0 (6.0)	20.8 (8.2)	–10.2 (p<0.01)	1.42
Handicap	27.4 (6.2)	21.8 (8.0)	–5.6 (p<0.01)	0.79

NS, not significant.
a Authors report effect size 0.2–0.5 = small effect, 0.5–0.8 = moderate effect, greater than 0.8 = large effect.[82]

Hol and colleagues[82] also reported the number of otolaryngology visits over the preceding 6 months for draining ears, and found these to reduce from a mean of 5.40 (SD 4.19, range 0–20) visits with BCHA to a mean of 1.50 (SD 2.10, range 0–6) visits with the BAHA (statistical significance not reported).

Four studies reported the results of subjective questionnaires on patient preference,[77,81,82] satisfaction,[78] comfort[79] and opinions on speech recognition in noise and quiet.[78–81] None of these questionnaires appeared to have been validated and are therefore not discussed here. The data from these studies can be viewed in the data extraction forms in *Appendices 6* and *8*; however, care should be taken when interpreting results because of the issues associated with non-validated questionnaires.

Assessment of clinical effectiveness: BAHA versus ACHA

Audiological measures

Five included studies (in seven publications)[76,78–81,83,84] provided a comparison of audiological measures between a BAHA and an ACHA (*Table 10*). Four studies were pre–post comparisons and the BAHAs used in these studies are no longer manufactured. One audiological comparison study assessed a BAHA model in current use (BAHA Intenso).[76] The high risk of bias in all five included studies should be considered when interpreting the results.

Audiometry

All five included studies reported data on hearing threshold tests (see *Table 10*); four used sound field warble-tone thresholds[76,78,79,83] and the remaining study states that it used sound field thresholds, but whether this was pure-tone or warble-tone was not reported.[84]

Cooper and colleagues[78] reported mean sound field warble-tone thresholds over a 0.5–4.0 kHz range. In this study two subgroups were tested, those whose condition was caused by chronic suppurative otitis media ($n = 19$) and those whose condition was congenital hearing loss ($n = 16$). Mean sound field warble-tone threshold was reported to be statistically significantly better with the BAHA than with the ACHA in the chronic suppurative otitis media subgroup [40 dB(A) ACHA vs 33 dB(A) BAHA, $p < 0.01$]. In the congenital hearing loss subgroup the BAHA was also seen to be statistically significantly better than the ACHA [41 dB(A) ACHA vs 28 dB(A) BAHA, $p < 0.01$]. In this study the BAHA tested was one of the HC 200, 300 or 220. Flynn and colleagues[76] reported data on the average aided sound field warble-tone thresholds at a range of frequencies (0.25–8.0 kHz). Testing 10 participants with either an ACHA or the BAHA Intenso, this study reports that, clinically, audibility improved with the BAHA by 5–15 dB at 1, 2, 3 and 4 kHz and that the BAHA provided a statistically significant improvement ($p < 0.01$) in audibility. All participants in this study had used BAHAs for at least 1 year prior to assessment with the ACHA.

Snik and colleagues[79] presented data from five participants on the difference between the sound field warble thresholds (ACHA minus HC 220 BAHA) across a range of frequencies. At 0.25 and 0.50 kHz, negative values indicate that hearing was better with the BAHA, while at higher frequencies (1–4 kHz), the positive values indicate that hearing was better with the ACHA (with a zero difference at 8 kHz). However, there is no statistical significance testing to aid interpretation of these data. Participants in this study had severe mixed hearing loss (mean hearing aid use of 23 years). Burrell and colleagues[83] reported mean sound field warble-tone thresholds over a 0.5–4.0 kHz range. Participants in this study had otosclerosis for which stapedectomy was declined, not indicated or had previously failed. In the nine participants tested, the mean (SD) sound field warble-tone thresholds (0.5–4.0 kHz) were 33.0 dB(A) (5.4) with the ACHA compared with

TABLE 10 Audiological measures: BAHA versus ACHA

Study and outcomes	ACHA	BAHA model	Comparison
[a]***Burrell* et al. *1996*[83] (n = 9)**		***Model NR***	
Average sound field warble-tone thresholds (0.5–4.0 kHz), mean (SD), range,[b] dB(A)	33 (5.4), 28–40	30.6 (8.1), 22–43	NR
Sound field speech discrimination at 63 dB(A), % correct, mean (SD), range[b]	91.6 (14.7), 60–100	84 (22.3), 30–100	NR
[a]***Cooper* et al. *1996*[78] *two subgroups: CSOM (n = 24), CON (n = 6)***		***HC 200, 300, 220***	**p-*value***
Mean sound field warble-tone thresholds, [dB(A), 500–4000 Hz][c]	CSOM 40	CSOM 33	$p<0.01$
	CON 41	CON 28	$p<0.01$
Mean sound field speech discrimination score (at 63 dB), % correct[c]	CSOM 69	CSOM 72	$p=$NS
	CON 57	CON 82	$p<0.05$
***Flynn* et al. *2009*[76] (n = 10)**		***Intenso***	***Difference***
Average aided warble-tone thresholds (dB SPL)[c]			$p<0.01$ overall
250 Hz	39	47	
500 Hz	42	39	
1 kHz	37	30[d]	
2 kHz	43	31[d]	
3 kHz	46	39[d]	
4 kHz	50	41[d]	
6 kHz	75	53	
8 kHz	68	55	
Speech-in-noise ratio, dB	3.44	0.88	2.56
[e]***Mylanus* et al. *1998*[84] (n = 34)**		***Model NR***	***Difference and* p-*value***
Mean sound field threshold, dB HL[a] (SD[f])			
0.25 kHz	40	39	$p=$NS
0.50 kHz	36	36	$p=$NS
1 kHz	28	22 (8.3)	$p<0.01$
2 kHz	22 (11.9)	25	$p=$NS
4 kHz	37	33	$p=$NS
8 kHz	55 (21.3)	43 (22.3)	$p<0.001$
Maximum phoneme score (mean ± SD)	Data NR	Data NR	1.0% ± 5.4%, $p=$NS
Speech-to-noise ratio improvement	Data NR	Data NR	1.1 ± 2.1 dB $p<0.01$

30.6 dB(A) (8.1) with the BAHA. No statistical analyses are presented; however, the authors state that results were comparable to the ACHA and 'significantly' better in one case. The BAHA model used in this study, which was conducted in 1996, was not reported.

Mylanus and colleagues[84] tested mean sound field thresholds over a 0.25–8.00 kHz range in 34 participants. The BAHA (model not reported) was seen to be statistically significantly better than the ACHA at 1 kHz and at 8 kHz. There were no statistically significant differences between the two devices at the other frequencies tested.

Speech audiometry

All five[76,78–81,83,84] included studies provide details of speech discrimination using ACHA and BAHAs (see *Table 10*). A variety of different tests were used, as discussed below.

TABLE 10 Audiological measures: BAHA versus ACHA *(continued)*

Study and outcomes	ACHA	BAHA model	Comparison
[e]***Snik* et al. *1992,[79] 1994,[80] 1998[81]***		***HC 200, 220***	
Snik et al. *1992*[79] *(n = 5)*	*ACHA*	*HC 220*	
Maximum phoneme score,%, mean (SD), range[g]	81.6 (8.7), 70–90	67.6 (22.2), 43–90	
Speech recognition threshold, dB(A), mean (SD), range[g]	39 (10.8), 20–45	(*n* = 3) 45 (5), 40–50	
Average difference between the sound field warble thresholds, dB[c] (ACHA minus BAHA)			
250 Hz	–6		
500 Hz	–5		
1000 Hz	3		
2000 Hz	4		
4000 Hz	15		
8000 Hz	0		
Snik et al. *1994*[80] *(n = 14)*	*HC 220, HC 200*		
Patients with a statistically significant change in:	*HC 220 (n = 5)*		
■ speech recognition in quiet score with BAHA[h]	Improved: 2 of 5 (40%); deteriorated: 1 of 5 (20%)		
■ speech-to-noise ratio score with BAHA[h]	No results for HC 220		
Patients with a statistically significant change in:	*HC 200 (n = 9)*		
■ speech recognition in quiet score with BAHA[h]	Improved: 0 of 9; deteriorated: 1 of 9 (11%)		
■ speech-to-noise ratio score with BAHA[h]	Improved: 5 of 9 (56%); deteriorated: 1 of 9 (11%)		
Snik et al. *1998*[81] *(n = 8)*	*HC 200*		
Change in SRT in quiet (ACHA minus BAHA), mean dB (SD)	–6.4 (3.7), $p < 0.05$ significant deterioration		
Change in speech-to-noise ratio, mean dB (SD)	1.6 (1.0), $p < 0.05$ significant improvement		

CON, congenital hearing loss; CSOM, chronic suppurative otitis media; NR, not reported; NS, not significant; SPL, sound pressure level.
a There may be overlap of participants between these studies conducted in the UK.
b Individual patient data estimated from figure and means and SDs calculated by reviewer.
c Data estimated from figure by reviewer.
d States that clinically, audibility improved by 5–15 dB at these frequencies (although figure does not seem to show that for 500 Hz).
e The study by Mylanus *et al.*[82] is conducted in the same centre in the Netherlands as the study by Snik *et al.*,[79–81] but it is not clear whether there is an overlap of participants.
f Standard deviations for each frequency NR. States that the SD varied between 11.9 dB at 2 kHz and 21.3 dB at 8 kHz for ACHA, and between 8.3 dB at 1 kHz and 22.3 dB at 8 kHz for BAHA.
g Means and SDs calculated by reviewer.
h Percentages reported in paper, numerator calculated by reviewer. There appears to be a slight rounding error in paper.

Cooper and colleagues[78] report mean sound field speech discrimination scores, using Boothroyd word lists undertaken at 63 dB, for their two subgroups (chronic suppurative otitis media $n = 24$, congenital hearing loss $n = 9$). The difference in proportion of correct scores between ACHA and BAHA in the chronic suppurative otitis media subgroup was not statistically significant [ACHA 69% vs BAHA 72%, $p =$ not significant (NS)], but in the COM subgroup there was a statistically significant difference in favour of BAHA (ACHA 57% vs BAHA 82%, $p < 0.05$). Burrell and colleagues[83] also reported mean sound field speech discrimination scores, using Boothroyd word lists at 63 dB(A). The proportion of correct scores was not tested for statistical significance; however, scores were better (91.6%, SD 14.7) with the ACHA than with the BAHA (84.0%, SD 22.3).

Flynn and colleagues[76] measured speech understanding in noise using the adaptive procedures from the Swedish version of the Hearing In Noise Test,[94] and presented speech through a loudspeaker from 0° and noise from 180°. They found a 2.56 dB improvement in the

speech-to-noise ratio with the BAHA compared with the ACHA in their sample of 10 (ACHA 3.44 dB vs BAHA 0.88 dB). No statistical analysis was presented for this difference, but the authors suggested that the improvement in speech understanding with BAHA was 'large and clinically significant'. In this cross-sectional audiological comparison study, all 10 participants had experience with BAHAs prior to testing with the ACHA.

Mylanus and colleagues[84] reported both the maximum phoneme score, which was calculated from the sound field speech audiogram, and the speech-to-noise ratio improvement. The speech-to-noise ratio was determined according to the criteria of Plomp and Mimpem[91] at a fixed noise level of 65 dB. In this study of 34 participants it was shown that there was no statistically significant difference between devices on the maximum phoneme score (difference 1.0% ± 5.4%), but a statistically significant improvement in the speech-to-noise ratio between ACHA and BAHA (difference 1.1 ± 2.1 dB, $p < 0.01$) in favour of the BAHA.

Snik and colleagues[79] found improved outcomes with the ACHA compared with the BAHA HC 220 for maximum phoneme score [ACHA 81.6% (SD 8.7%), BAHA 67.6% (SD 22.2%); $n = 5$)] and speech recognition threshold [ACHA 39.0 (SD 10.8) db (A), BAHA 45.0 (SD 5.0) db (A); $n = 3$], which were determined using standard Dutch PB word lists consisting of 10 monosyllables. However, no statistical significance testing was undertaken and the sample size was small, therefore the meaningfulness of the data is uncertain. Two later publications reported additional outcomes and also a group of participants using the BAHA HC 200.[82,83] The 1994 publication[80] simply reported the proportion of participants with a statistically significant improvement or deterioration with the BAHA in speech recognition in quiet and speech-to-noise ratio, determined using methods previously described in *Audiological measures*. This showed that with the HC 200 ($n = 9$) no participants improved on their speech recognition in quiet score, whereas 11% deteriorated, and with the HC 220 ($n = 5$) 40% of participants improved and 20% showed a deterioration. No further detail was reported. For speech-to-noise ratio, 55% of participants with the HC 200 improved and 11% deteriorated. No results were presented for the HC 220 on this measure. The 1998 publication[81] reported the change in SRT in quiet and the change in speech-to-noise ratio. The authors reported a statistically significant deterioration in mean SRT in quiet with the BAHA HC 200 [–6.4 dB (SD 3.7), $p < 0.05$] among their eight participants, although there was a statistically significant improvement in speech-to-noise ratio [1.6 dB (SD 1.0), $p < 0.05$]. The authors reported that these ambiguous results do not mean that the ACHA would be used in preference to the BAHA in their sample, owing to their chronic draining ears.

Self-reported measures

Hol and colleagues[82] assessed QoL using an ACHA prior to BAHA surgery and again after 6 months' experience with a BAHA. There were no statistically significant differences found by the SF-36 (*Table 11*), and although mental health improved slightly with the BAHA ($p =$ not significant), the effect size was small (–0.28). The EQ-5D found a statistically significant increase in anxiety/depression ($p < 0.01$) with the BAHA, but again the clinical effect was small (–0.3). There were no other statistically significant differences in the EQ-5D. However, statistically significant improvements in disability ($p < 0.01$) and handicap ($p < 0.01$) were found with the BAHA by the HHDI, and effect sizes indicated a large clinical impact (0.79 and 0.86, respectively).

Hol and colleagues[82] also reported the number of otolaryngology visits over the preceding 6 months for draining ears, and found these to reduce from a mean of 12.7 (SD 10.5, range 0–30) visits with ACHA to a mean of 3.3 (SD 4.8, range 0–25) visits with the BAHA (statistical significance not reported).

TABLE 11 Self-reported measures: BAHA versus ACHA

Study and outcomes	ACHA	BAHA model: Classic, Cordelle	Mean difference	Effect size[a]
***Hol* et al. *2004*[82] (n=36)**				
SF-36 mean (SD)				
Physical functioning	80.3 (21.8)	79.8 (22.4)	–0.5 (p=NS)	0.02
Role limitations (physical)	71.5 (39.7)	68.9 (40.5)	–2.6 (p=NS)	0.06
Role limitations (emotional)	76.2 (40.1)	73.2 (38.1)	–3.0 (p=NS)	0.07
Vitality	60.4 (20.0)	59.9 (19.9)	–0.5 (p=NS)	0.02
Mental health	62.4 (18.0)	67.9 (21.3)	5.5 (p=NS)	–0.28
Social functioning	69.8 (28.3)	75.0 (27.8)	5.2 (p=NS)	–0.19
Pain	74.7 (25.2)	79.2 (25.0)	4.5 (p=NS)	–0.18
General health	63.2 (21.4)	63.6 (21.2)	–0.4 (p=NS)	–0.18
EQ-5D mean (SD)				
Five domains (score 1–3)				
Mobility	1.29 (0.46)	1.31 (0.47)	0.02	–0.04
Self-care	1.03 (0.17)	1.03 (0.17)	0.00	0.00
Usual activities	1.47 (0.66)	1.44 (0.50)	–0.03 (p>0.05)	0.05
Pain/discomfort	1.49 (0.51)	1.47 (0.51)	–0.02 (p>0.05)	0.04
Anxiety/depression	1.26 (0.44)	1.42 (0.60)	0.16 (p<0.01)	–0.30
Utility (score 0–1)	0.78 (0.17)	0.77 (0.17)	–0.01	0.06
Visual analogue scale (0–100)	76.1 (14.1)	73.4 (17.1)	–2.7	0.17
HHDI mean (SD)				
Disability	25.8 (6.5)	20.9 (6.2)	–5.0 (p<0.01)	0.79
Handicap	25.0 (5.9)	19.6 (6.7)	–5.4 (p<0.01)	0.86

a Authors report effect size 0.2–0.5 = small effect, 0.5–0.8 = moderate effect, greater than 0.8 = large effect.[82]

Five studies reported the results of subjective questionnaires on patient preference,[81–84] satisfaction,[78] comfort[79,83] and opinions on speech recognition in noise and quiet.[78–81] None of these questionnaires appeared to have been validated and they are therefore not discussed here. The data from these studies can be viewed in the data extraction forms in *Appendices 7* and *8*, although care should be taken when interpreting results owing to the issues associated with non-validated questionnaires.

Assessment of clinical effectiveness: BAHA versus unaided hearing

Audiological measures

Four cohort pre and post studies were identified that provide a comparison between BAHAs and unaided hearing (*Table 12*).[66,77,78,83] Three studies used BAHA models that are no longer manufactured.[77,78,83] The study by Kompis and colleagues[66] included the BAHA Divino, which is in current use. As discussed in *Characteristics of included studies*, this study differs from the other included studies as the participants had prior experience with BAHAs when tested unaided at

baseline. The high risk of bias in all four included studies should be considered when interpreting the results.

The studies by Béjar-Solar and colleagues,[77] Burrell and colleagues[83] and Cooper and colleagues[78] also reported data comparing the BAHA with the hearing aids previously used, BCHAs,[77] ACHAs[83] or ACHAs and BCHAs.[78] In order to aid the narrative synthesis of studies for similar comparators, the BAHA data from these studies are repeated in *Tables 8*, *10* and *12*. Care should therefore be taken to ensure that data for BAHAs are not 'double-counted' when interpreting the results. The entire data set for each study can be seen in the data extraction forms in *Appendices 6–9*.

Audiometry

All four studies[66,77,78,83] included data on audiometry: two[78,83] stated using sound field warble-tone thresholds and one used sound field pure-tone.[77] All four studies found an improvement with BAHAs compared with the unaided condition (see *Table 12*).[66,77,78,53] Mean sound field PTA threshold improved from 64 dB HL unaided to 19 dB HL with the BAHA in 11 participants with inoperable bilateral congenital atresia in the study by Béjar-Solar and colleagues.[77] Burrell and colleagues[83] included nine participants with otosclerosis who had previously used an ACHA. Average sound field warble-tone thresholds (0.5–4.0 kHz) improved from 49.4 (SD 11.9) dB(A) unaided to 30.6 (SD 8.1) dB(A) with the BAHA, although no statistical analysis was reported. It should be noted that the unaided condition was assessed pre- and post-operatively in this study, and it is not clear which of these data are presented. Another study from the same centre (and therefore note the possibility of overlap of participants) by Cooper and colleagues[78] included a total of 68 participants split into four subgroups according to previous aid (ACHA or BCHA) and aetiology (chronic suppurative otitis media or congenital hearing loss). In all subgroups, the mean sound field warble-tone thresholds showed statistically significant improvement with the BAHA ($p < 0.01$ for each comparison; see *Table 12*). The models used in this study were the HC 200, 220 or 300.

The study by Kompis and colleagues[66] included seven participants who had at least 2 years' experience of a BAHA Compact or Classic 300. Sound was presented through two loudspeakers placed just off one diagonal axis, which the participant sat in between. The study found a statistically significant improvement of 28 dB ($p < 0.001$) in average sound field thresholds over all frequencies after 3 months with the BAHA Divino compared with unaided.

Speech audiometry

Three studies[66,78,83] reported data on speech audiometry. In nine participants with otosclerosis, Burrell and colleagues[83] demonstrated an improvement in sound field speech discrimination scores, using Boothroyd word lists at 63 dB(A), from 74% (SD 19.5%) unaided to 84% (SD 22.3%) with a BAHA (model not reported), although no statistical analysis was conducted. Cooper and colleagues[78] also found improvements in this outcome using either a BAHA HC 200, 300 or 220 compared with unaided in each of their subgroups (see *Table 12*), but again no statistical analysis was undertaken.

Speech recognition thresholds in quiet (levels required for 50% speech understanding, using Freiburger two-digit numbers) improved from 54 dB unaided to 23 dB with the BAHA Divino in the study by Kompis and colleagues.[66] There was also an improvement in speech recognition scores in quiet, which were defined as the percentage of correctly repeated Freiburger monosyllabic words at 50 dB SPL (decibel sound pressure level) (unaided 5%, BAHA Divino 45%), 60 dB SPL (unaided 15%, BAHA Divino 90%) and 80 dB SPL (unaided 50%, BAHA Divino 95%). Speech audiometry in noise was assessed using the Basler sentence test,[95] an adaptive test in which speech was presented at 70 dB from a loudspeaker in front of the participant and

TABLE 12 Audiological measures: BAHA versus unaided hearing

Study and outcomes	Unaided	BAHA model	Comparison
***Béjar-Solar* et al. *2000*[77] (n = *11*)**		***BAHA Classic 300***	
Sound field PTA threshold (1.25–3.00 kHz), dB HL	64	19	
[a]*Burrell* et al. *1996*[83] (n = *9*)		***Model NR***	
Average sound field warble-tone thresholds (0.5–4.0 kHz), mean (SD), range[b], dB(A)	49.4 (11.9), 40–78	30.6 (8.1), 22–43	
Sound field speech discrimination at 63 dB(A), % correct, mean (SD), range[b]	74 (19.5), 50–98[c]	84 (22.3), 30–100	
[a]*Cooper* et al. *1996,*[78] *four subgroups: previous aid BC, CSOM (n = 19), CON (n = 16); previous aid AC, CSOM (n = 24), CON (9)*		***HC 200, 300, 220***	**p-*value***
Mean sound field warble-tone thresholds, dB [dB(A) 0.5–4.0 kHz][d]	Previous aid BC	Previous aid BC	
	CSOM 63	CSOM 35	$p<0.01$
	CON 62	CON 26	$p<0.01$
	Previous aid AC	Previous aid AC	
	CSOM 60	CSOM 33	$p<0.01$
	CON 68	CON 28	$p<0.01$
Mean sound field speech discrimination score (at 63 dB), % correct[d]	Previous aid BC	Previous aid BC	
	CSOM 17	CSOM 72	$p=$NR
	CON 3	CON 85	$p=$NR
	Previous aid AC	Previous aid AC	
	CSOM 19	CSOM 72	$p=$NR
	CON 17	CON 82	$p=$NR
***Kompis* et al. *2007*[66] (n = *7*)**		***Divino***	**p-*value***
Average improvement in sound field thresholds over all frequencies compared with unaided, dB		28.0	$p<0.0001$
Speech recognition thresholds in quiet using two-digit numbers, dB (assume value is mean)[d]	54	23	$p=$NR
Speech recognition scores for monosyllabic words in quiet, % correct (assume mean)[d]			
50 dB SPL	5	45	$p=$NR
65 dB SPL	15	90	
80 dB SPL	50	95	
Speech recognition threshold in noise (noise presented from front or back), dB[d]		*Omnidirectional/ directional mode*	
Front	12	3/4	
Back	9	3/1	

CON, congenital hearing loss; CSOM, chronic suppurative otitis media; NR, not reported.
a Burrell *et al.*[83] and Cooper *et al.*:[78] BAHA data are also presented in *Tables 8* and *10* for the comparisons with BCHA and ACHA.
b Individual patient data estimated from figure and means and SDs calculated by reviewer.
c Data missing for two participants.
d Data estimated from figure by reviewer.
Priwin *et al.*[59] also reported data for the unaided condition; however, as this is a audiological comparison design and the studies in the table above are of a higher level of evidence, the study has not been summarised here (see *Appendix 9* for details).

noise was emitted either from the same direction or from the back of the participant (180°). The SNR, in dB, at which 50% of the key words were understood correctly, was measured. The speech recognition threshold in noise was reduced approximately from 12 dB unaided to 3 dB with the Divino in the omnidirectional model and to 4 dB in directional mode when noise was

presented from the front; and when noise was presented from the back the threshold was reduced approximately from 9 dB unaided to 3 and 1 dB, respectively. However, no statistical analysis was undertaken for any of these comparisons.

Self-reported measures

No self-reported measures were described by the included studies comparing BAHAs with unaided hearing.

Assessment of clinical effectiveness: unilateral versus bilateral BAHAs

Audiological measures

Four audiological comparison studies[59,60,85–87] compared unilateral and bilateral BAHAs (one study was reported in two publications[60,85]) (*Table 13*, *Appendix 10*). None of these studies

TABLE 13 Audiological measures: unilateral versus bilateral BAHAs

Study and outcomes	Unilateral		Bilateral	Comparison (*p*-value)
Bosman* et al. *2001,[60,85] BAHA HC 200 or Classic 300 (n = 25)	***Unilateral***		***Bilateral***	
SRT in quiet [dB(A)]	41.5		37.5	$p<0.001$
SNR ([dB(A)], noise from the baffle side	–0.7		–3.2	$p<0.001$
SNR [dB(A)], noise from shadow side	–3.4		–4.0	$p>0.05$
Directional hearing at 500 Hz, %[a]				
Correct localisation	23		42[b]	
Localisation within 30°	56		90[b]	
Lateralisation	54		85[b]	
Directional hearing at 2 kHz, %[a]				
Correct localisation	24		45[b]	$p<0.001$ across all observations
Localisation within 30°	58		89[b]	
Lateralisation	64		87[b]	
Proportion of responses corresponding to the fitted BAHA side at				
500 Hz	75.3%		45.7%	
2 kHz	70.3%		48.8%	
Binaural masking level difference SNR[a,c]		Bilateral BAHAs ($n=9$)		
	S_0N_0	$S_\pi N_0$	S_0N_π	
125 Hz	2.2	3.8	–3.7	$p<0.001$
250 Hz	0.1	–6.0	–5.1	$p<0.001$
500 Hz	0.4	–5.9	–3.9	$p<0.001$
1 kHz	0.4 -	–3.3	–4.9	$p>0.05$ (NS)
Dutt* et al. *2002,[86] BAHA Compact (n = 11)	***Unilateral***		***Bilateral***	
Speech-in-quiet (Arthur-Boothroyd word list cumulative scores, 30 words) at[a]	*Best response*			
30 dB intensity levels	1		5	
40 dB intensity levels	13		19	
50 dB intensity levels	20		24	
60 dB intensity levels	25		28	

TABLE 13 Audiologic measures: unilateral versus bilateral BAHAs *(continued)*

Study and outcomes	Unilateral	Bilateral	Comparison (*p*-value)
70 dB intensity levels	27	29	
80 dB intensity levels	30	30	
Speech-in-quiet (Bamford–Koval–Bench sentences)	All 11 patients scored 100% with right, left and bilateral BAHAs		
Speech-in-noise (Bamford–Koval–Bench cumulative sentence scores) at[a]	*Best response*		
Plus 10 SNR	99	100	
Zero SNR	80	81	
Minus 10 SNR	0	1	
Plomp test, % correct score [mean (SD), range][d]			
Sound front, noise front	Left side: 76 (11.7), 56–93; right side: 77.3 (11.7), 58–90	82.4 (13.3), 60–97	
Sound front, noise left	Left side: 40.1 (25.3), 2.–71; right side: 84.1 (11.2), 55–97	71.1 (14.9), 44–95	
Sound front, noise right	Left side: 88.2 (9), 72–100; right side: 45.8 (22.1), 13–88	79.5 (11.6), 58–93	
***Priwin* et al. *2004*[87] *BAHA Compact or Classic* (n = *12*)**			
Average difference in sound field tone thresholds (at 0.25–8 kHz), dB			
Sound presented in front, at best side and from behind patients		2–7 dB improvement with bilateral	
Sound presented at shadow side		5–15 dB improvement with bilateral	
Speech recognition in quiet, average threshold, dB HL	38.7	33.3	$p = 0.001$
Speech-in-noise (change in SNR with bilateral BAHA), masking noise presented:			
At best side		3.1 dB improvement	
At shadow side		1.0 dB deterioration	
As surrounding noise		2.8 dB improvement	
Directional hearing	*Best/shadow side*		
Per cent of correct answers[a,e]			
0.5 kHz	12/11	25	
2.0 kHz	8/10	23	
Per cent of answers within 30° of correct response[a,e]			
0.5 kHz	23/30	53	
2.0 kHz	28/27	51	
Binaural masking level difference (relative threshold change in dB from the condition 'signal and noise in phase at both sides')		*Bilateral BAHAs (n = 12)*	
0.25 kHz			
$S_\pi N_0$		Threshold changes within 3 dB except for two patients	

continued

TABLE 13 Audiologic measures: unilateral versus bilateral BAHAs *(continued)*

Study and outcomes	Unilateral	Bilateral	Comparison (*p*-value)
S_0N_π		Threshold changes between –18 and 3 dB, mean –5 dB	
0.5 kHz			
$S_\pi N_0$		Average threshold change 2 dB	
$S_0N\pi\tau$		Average threshold change –4 dB	
1 kHz			
$S_\pi N_0$		Average threshold change 3 dB	
S_0N_π		Average threshold change –3 dB	
Priwin* et al. *2007*[59] *BAHA Classic or Compact. Two groups: (unilateral BAHA* n = *6, bilateral BAHA* n = *3)	***One BAHA (unilateral* n = *6/bilateral* n = *3)***	***Two BAHAs (bilateral* n = *3)***	
Sound field average tone thresholds, dB HL;[d] mean (SD, range)	24 (5, 20–32)/30 (5, 25–35)	25 (5, 20–30)	
Speech recognition in noise, median score (%)[a]			
SNR 0 dB	87/69	88	
SNR 4 dB	92/79	93	
SNR 6 dB	98/97	90	
Localisation of sound at 0.5 kHz,[a] *mean %*			
Correct score[f]			
50 dB	20/20	50	
60 dB	28/20	50	
Lateralisations score[f]			
50 dB	68/60	86	
60 dB	70/68	94	
Localisation of sound at 3 kHz,[a] *mean %*			
Correct score[f]			
50 dB	28/16	50	
60 dB	37/18	57	
Lateralisations score[f]			
50 dB	60/68	80	
60 dB	72/56	96	

NS, not significant; SNR, signal-to-noise ratio; S_0N_0, in-phase tone stimuli and in-phase noise bands; $S_\pi N_0$,180° out-of-phase tone stimuli and in-phase noise bands; S_0N_π, in-phase tone stimuli and 180° out-of-phase noise bands.

a Data estimated from figure by reviewer.

b $p < 0.05$ vs the chance level for that outcome. For correct localisation the chance level is 14.3%, (95% upper confidence limit 32%[a]), for localisation within 30° the chance level is 42.9% (95% upper confidence limit 64%[a]) and for lateralisation the chance level is 50% (95% upper confidence limit 32%[a]).

c Note discrepancy between table and text; see *Appendix 10* for details.

d Means and SDs calculated by reviewer.

e For correct score, the chance level is 8.3%; for answers within 30° the chance level is 25%.

f For correct localisation score, the chance level is 20%; for lateralisation score, the chance level is 68%.

assessed a BAHA model that is currently available. In each of the four studies, participants had several months' experience with bilateral BAHAs before being tested with unilateral and bilateral BAHAs during the same session, which could lead to bias, as discussed in *Quality assessment*. The high risk of bias in all four included studies should be considered when interpreting the results.

Audiometry

Two included studies reported data on hearing thresholds, one in an adult population[87] and one in children[59] (see *Table 13*). Priwin and colleagues[87] included 12 adults and found an average improvement of 2–7 dB with bilateral BAHAs in free sound field tone thresholds (at 0.25, 1, 1.5, 2, 3, 4, 6 and 8 kHz) when sound was presented in front, at the best side (usually aid first implanted) and from behind participants, and an improvement of 5–15 dB when sound was presented at the shadow side. A comparative strength of this study is that the BAHAs, which were either Compact or Classic models, were electronically controlled by research personnel (BAHAs could be switched on and off by the investigator without the participant's knowledge) and tests were randomised so that participants were blinded to unilateral or bilateral use of BAHAs.

A later study by the same group[59] included two small groups of children, unilateral BAHA users ($n = 6$) and bilateral BAHA users ($n = 3$), although no statistical comparison was made between the groups. The bilateral BAHA users group were tested using one BAHA and two BAHAs, but the unilateral BAHA users group were only tested with one BAHA. Mean sound field average-tone thresholds were 24 (SD 5) dB HL for the unilateral BAHA user group, 30 (SD 5) dB HL for the bilateral group using one BAHA and 25 (SD 5) dB HL for the bilateral group using two BAHAs.

Speech audiometry

All four included studies[59,60,85–87] reported speech audiometry with unilateral and bilateral BAHAs (see *Table 13*).

In a study of 25 consecutive patients by Bosman and colleagues,[60,85] mean SRTs in quiet were measured with sentences by Plomp and Mimpen[91] and Smoorenburg,[96] with speech presented in front of the participant and an adaptive procedure used to determine the presentation level providing a whole-sentence correct score of 50%. SRTs in quiet were found to be significantly lower with bilateral than unilateral BAHAs [37.5 dB(A) vs 41.5 dB(A), $p < 0.001$]. A statistically significant difference was also found in favour of bilateral BAHAs in the SNR for noise presented at 65 dB(A) from the baffle side [–3.2 dB(A) vs –0.7 dB(A), $p < 0.001$], but not for noise presented from the shadow side [–4.0 dB(A) vs –3.4 dB(A), $p > 0.05$]. This study used the BAHA HC 200 or Classic 300.

Dutt and colleagues[86] assessed 11 adults whose 'professional needs warranted binaural hearing' and used a BAHA Compact. All had voluntarily applied for the second BAHA and all had previous experience of binaural hearing. No statistical comparison was made between unilateral and bilateral use of BAHAs; therefore, comments are based on observation of the data. For sound field speech in quiet using Bamford–Kowal–Bench sentences, all 11 participants scored 100% with right, left and bilateral BAHAs. For speech in quiet using Arthur Boothroyd word list cumulative scores at 30–80 dB intensity levels, bilateral BAHAs appeared to be slightly better than the best unilateral response at lower intensities (see *Table 13*). Speech-in-noise cumulative sentence scores appeared similar between the best unilateral response and bilateral BAHAs at SNRs of + 10, 0 and –10 dB. When using the Modified Plomp multitalker noise test, similar results were obtained with unilateral and bilateral BAHAs when both sound and noise were

presented from the front. However, when the noise was presented from the BAHA side (baffle situation, i.e. noise from left and using left BAHA only, or noise from right and using right BAHA only), scores were lower with a unilateral BAHA than with bilateral BAHAs, and when noise was presented from the opposite side (shadow side, i.e. noise from right and using left BAHA only, or noise from left and using right BAHA only), scores were better with a unilateral BAHA than a bilateral BAHA.

In their study of 12 adults, Priwin and colleagues[87] found the average threshold for speech recognition in quiet (measured with phonetically balanced three-word sentences extracted from Hagerman[97] and presented at 0°) was statistically significantly lower with bilateral BAHAs than the best unilateral side (33.3 dB HL vs 38.7 dB HL, $p = 0.001$). SRT in noise was also tested, where speech was presented at the participant's most comfortable level, between 65 and 80 dB HL, and noise was speech weighted. An improvement of around 3 dB in the SNR was found with bilateral BAHAs when masking noise was presented at the best side and as surrounding noise. A deterioration of 1.0 dB in the SNR was found with bilateral BAHAs when noise was presented at the shadow side.

Similar scores for speech recognition in noise were found between the unilateral BAHA users group ($n = 6$) and the bilateral BAHA users group ($n = 3$) in the study of children by Priwin and colleagues[59] (see *Table 13*), although the group of bilateral BAHA users had lower scores when tested with just one BAHA at 0 and 4 dB SNR. However, as the sample sizes were very small and no statistical analysis was undertaken, this should be interpreted with caution. This study used phonemically balanced Swedish three-word sentences extracted from Hagerman.[97] Speech and noise were presented at 0°, with speech presentation level set at 60 dB SPL and noise presented at SNRs of 0, 4 and 6 dB; thus, noise was presented at 60, 56 and 54 dB SPL.

Directional hearing

Directional hearing was assessed in three included studies[59,60,85,87] (see *Unilateral versus bilateral BAHAs* and *Appendix 10* for details on methods). Correct localisation, localisation within 30° and lateralisation measured at 0.5 and 2.0 kHz were significantly better than chance ($p < 0.05$) with bilateral BAHAs, but not with unilateral BAHAs, in the study by Bosman and colleagues.[60,85] Bilateral scores were statistically significantly better than unilateral scores across all observations ($p < 0.001$) (see *Table 13*). The study also found that sounds appeared to come from the fitted side when just one BAHA was in use. The proportion of responses corresponding to the fitted (baffle) side for unilateral BAHAs was 75.3% at 0.5 kHz and 70.3% at 2.0 kHz, whereas for bilateral BAHAs the responses were more symmetrical at 45.7% at 0.5 kHz and 48.8% at 2.0 kHz.

Priwin and colleagues[59] found similar results in their studies of 12 adults[87] and nine children, although no statistical analyses were undertaken. In the first study, the proportion of correct answers with a unilateral BAHA on the best or shadow side (scores between 8% at 2.0 kHz and 12% at 0.5 kHz) were close to the chance level of 8.3%, while with a bilateral BAHA the proportion of correct answers increased to 25% at 0.5 kHz and 23% at 2.0 kHz. The results for the proportion of answers within 30° of a correct response followed the same pattern (see *Table 13*).[87] This suggests that sound localisation was better with bilateral BAHAs. Similarly, the second study[59] found an improvement in sound localisation and sound lateralisation ability with bilateral BAHAs, while with unilateral BAHAs the results were close to chance levels (see *Table 13*).

Binaural hearing

Two studies used the masking-level difference test to investigate binaural hearing with bilateral BAHAs,[60,87] and both claimed that their results indicate that binaural hearing with bilateral BAHAs is possible, at least in some situations. However, the interpretation of this test with BC, being more complex than the interpretation with AC, remains to be established.[98]

Self-reported measures

Priwin and colleagues[59] reported the validated MAIS and MUSS to assess hearing skills in 'meaningful, real world situations', and the IOI-HA to assess hearing aid outcomes. Scores appeared similar between unilateral and bilateral BAHA users for most items; however, given the very small sample sizes ($n = 2–6$), these results should be interpreted with caution (*Table 14*).

Adverse effects

Included studies

Only 3 of the 12 included studies reported any data on adverse effects[77,81,84] (*Table 15*). Béjar-Solar and colleagues[77] reported that no irritation was present at most follow-up visits (71 of 82 visits); however, it is not clear how many of the 11 participants experienced irritation. No participants in this study experienced infection leading to loss of the implant or any major complications. Osseointegration could not be achieved in one participant following an impact to the mastoid area. Re-operations to remove or replace implants or to reduce the thickness of the subcutaneous layer around the implant were required in 15% of participants in the study by Snik and colleagues.[81] Two of 34 participants in the study by Mylanus and colleagues[84] stopped using their BAHA owing to pain of unknown cause; this was after 3 months' use in one participant and 2.5 years' in the second participant.

Prospective case series

To supplement the limited data from the included studies, prospective case series reporting adverse events were identified from the list of potentially eligible studies (see *Appendix 5*). As they were not included in the systematic review, these studies did not undergo the same process of data extraction and quality assessment. Six prospective case series reporting adverse events were identified; five of these can be seen in *Table 16*. Further examination of the remaining

TABLE 14 Self-reported measures: unilateral versus bilateral BAHAs

Study and outcomes	Unilateral BAHA users ($n = 6$)	Bilateral BAHA users ($n = 3$)	*p*-value
***Priwin* et al. *2007*[59]**			
MAIS and MUSS,[a] *mean (SD)*	($n = 6$)	($n = 3$[b])	p = NR
Hearing aid use	3.4 (1.3)	4.0	
Reaction to sounds	3.1 (0.9)	3.5	
Sound discrimination	3.5 (0.8)	3.8	
Verbal communication	3.8 (0.6)	3.7	
Speech intelligibility	3.1 (1.2)	3.3	
IOI-HA,[c] *mean (SD)*	($n = 6$)	($n = 2$[b])	p = NR
Use	5.0 (0.0)	5.0	
Benefit	5.0 (1.0)	5.0	
Residual activity limitation	4.2 (0.5)	4.0	
Satisfaction	4.3 (1.0)	5.0	
Residual participation	4.2 (1.3)	3.0	
Impact on others	4.8 (0.4)	2.5	
QoL	4.8 (0.4)	5.0	

NR, not reported.
a Scored 1–5, never to always. Completed by children's guardian and teacher.
b No SD owing to small number of participants.
c Scored 1–5, worst to best outcome. Mean scores below 3.5 in participants with mild-to-moderate hearing impairment and below 3.6 in moderate-to-severe hearing impairment indicate poor habilitation outcome.

TABLE 15 Adverse effects reported in included studies

Study	Results
***Béjar-Solar* et al. *2000*[77]**	***BAHA Classic 300* (n = 11)**
Unable to obtain osseointegration (following impact to mastoid area 24 hours after discharge from first stage)	1/11
Major complications	0/11
Types of skin reactions, n *of observations (%)*	
No irritation	71/82 (87)
Slight erythema	7/82 (8)
Erythema and moisture	3/82 (4)
Red and moist with granulation tissue	1/82 (1)
Infection leading to loss of implant	0
Total number of observations	82 (71 at scheduled visits, 11 at unscheduled visits)
***Mylanus* et al. *1998*[84]**	***BAHA model NR* (n = 34)**
States surgery was uneventful in all patients. Two stopped using their BAHA after 3 months and 2.5 years respectively, owing to pain – no explanation for this found	
***Snik* et al. *1998*[81]**	***BAHA HC 200* (n = 39)**
Lost implant owing to inflammation after 2 years of use	1 – implant not replaced
Requested implant removal owing to pain after 3 years	1
Implants loss due to inflammation	1 – implant replaced
Lost implant owing to trauma	2 – implant replaced
Reduction of thickness of the subcutaneous layer around implant to minimise risk for inflammation	2
Total re-operations	6/39 (15.4%)
Rejections of BAHA due to insufficient amplification	0
Severe deterioration in sensorineural hearing (25–65 dB HL) after surgery for cholesteatoma in cerebellopontine angle and refitted with more powerful BAHA (NBC-HC-220). However, result poor owing to severe deterioration of cochlear function	1
Non-users after at least 4.5 years (all others using BAHA on daily basis)	2/39 (5%)

NR, not reported.

study revealed no useful data for the purposes of this review so it is not discussed further.[99] The discussion below focuses on loss of implants and skin reactions as these were identified as being the most relevant to this report.

Loss of implants

Bonding[100] followed 31 adults for a median of 6 years (range 1–12 years). Seven (19.4%) implants were lost in six participants after a median of 42 months (range 27–78 months); the estimated causes were minor trauma (three), malignant disease with emaciation (one), severe dermatitis (psoriasis) with granuloma at the point of skin penetration (one), and uncertain causes (two). The success rate of the implants, expressed in a life table, was 100% at 2 years, reducing to 85% at 3–4 years and about 75% after 7 years (see *Table 16*).

In the study by Håkansson and colleagues,[101] 147 participants with 167 implants were followed for between 1 month and 11.5 years during 1977–87. Sixteen (9.6%) of the abutments were removed, owing to no hearing improvement (in seven participants with SNHL), unexplained discomfort

TABLE 16 Loss of implants and skin reactions reported in prospective case series

Study details and patient characteristics	Adverse events			Results	
Bonding 2000[100]					
Denmark	Implants lost			7 (19.4%) in 6 patients	
Prospective case series				After 27–78 months (median 42 months)	
Study period: 1986–98					
Length of follow-up (years): median 6, range 1–12	Estimated causes of loss			Minor trauma (3)	
				Malignant disease with emaciation (1)	
Number of participants: 31					
Sample attrition/dropout: NR				Severe psoriasis with granulation (1)	
Indication for treatment:					
Chronic otitis media (28)				Uncertain (2)	
External otitis (2)	*Success rate of implants, expressed in a life table sample*[a]				
Congenital atresia of ear canal (1)	**Time period (years)**	***n***	**Implants lost**	**Success rate (%)**	
Age (years): median 58, range 36–80				**Within-group**[b]	**Cumulative**
	1	31	0	100	100.0
Sex (M:F): 17:14	2	29	0	100	100.0
Funding: NR	3	26	3	88.5	88.5
	4	21	1	95.2	84.3
	5	19	1	94.7	79.8
	6	15	0	100	79.8
	7	14	1	92.9	74.1
	>7	10	0	100	74.1
Håkansson* et al. *1990[101]					
Sweden	167 implants in 147 participants				
Prospective case series	*Type of skin reactions*[c]			*Observations, % (n)*	
Study period: 1977–87	0: no irritation			93.2	
Length of follow-up: 1 month to 11.5 years	1: slight redness			4.1	
	2: red and moist tissue			1.3 (16 in 16 patients)	
Number of participants: 147	3: granulation tissue			1.3 (16 in 13 patients)	
Sample attrition/dropout: 0	4: infection and removal of abutment			0.1 (1)	
Indication for treatment	Total observations			1236	
Otitis media/external (107)	*Abutments removed*			16	
External ear canal/ossicular malformation (24)	■ Skin infection			1	
	■ Discomfort, psychological			5	
Otosclerosis (9)	■ No hearing improvement			7 (sensorineural hearing loss)	
SNHL (7)	■ Trauma			2	
Age (years): mean 50.8 (SD 17.4), range (5–82)	■ Implant not integrated			1	
	■ Abutments changed owing to inadequately hygienic and loose coupling			9 (all early in study)	
Sex (M:F): 78:69					
Funding: NR					
	■ Tissue reduction, thick and moveable skin			10 (all early in study)	

continued

TABLE 16 Loss of implants and skin reactions reported in prospective case series *(continued)*

Study details and patient characteristics	Adverse events	Results
***Jacobsson* et al. *1992*[103]**		
Sweden	*Grade of skin reactions*[e]	*Observations,* n *(%)*
Prospective case series	0: no irritation	99 (91.7)
Study period: NR	1: slight redness	7 (6.5)
Length of follow-up: 40 (range 1–144)[d] months	2: red and moist, no granulation	1 (0.9)
	3: as in 2, with granulation tissue	0
Number of participants: 16	4: revision of skin-penetration necessary	0
Sample attrition/dropout: NR		
Indication for treatment: NR	R: removal of implant owing to non-integration	1 (0.9)
Age (years): mean 10 (range 3–16)[d]		
	Total observations	108
Sex: NR		
Funding: NR		
***Mylanus* et al. *1994*[102]**		
Netherlands	33 implants	
Prospective case series	*Type of skin reactions*[c]	
Study period: 1991–2	0: no irritation	NR
Length of follow-up (months): 9–25	1: slight redness	11 (in 8 implants)
	2: red and moist tissue	7 (in 6 implants)
Number of participants: 33	3: red and moist tissue and/or granulation, revision surgery	0
Indication for treatment:	4: infection and removal of abutment	1
Chronic otitis media (28)	Implants lost	2/33
Chronic otitis externa (3)	Cause of loss	Severe inflammatory reaction around implant site (1), trauma (1)
Congenital anomaly (2)		
Age (years): mean 50, range 15–76		
Sex (M : F): 13 : 20	*Life table: cumulative proportion of implants which did not suffer from any skin reaction in follow-up period*	
Funding: reported[e]		

Interval months	No. followed	No. of reactions	Proportion within interval (%)	Cumulative proportion size	Effective sample size
0–4	33	7	79	79	26
4–8	26	3	88	70	18
8–12	17	1	94	66	11
12–16	10	0	100	66	7
16–20	6	0	100	66	4
20–24	3	0	100	66	3

or for psychological-cosmetic reasons (five), trauma (two), skin infection (one) and the bone implant not integrated (one). Nine abutments were changed owing to inadequate hygiene and loose coupling, and in 10 participants the subcutaneous tissue reduction had not been extensive enough, although these all occurred early in the study while improvements were still being made.

Mylanus and colleagues[102] followed 33 participants with 33 implants for 9–25 months. Two (6.1%) of the implants were lost owing to a severe inflammatory reaction around the implant

TABLE 16 Loss of implants and skin reactions reported in prospective case series *(continued)*

Study details and patient characteristics	Adverse events	Results
***Portmann* et al. *1997*[104]**		
France	*Type of skin reactions*[c,e]	*Observations,* n *(%)*
Prospective case series	0: no irritation	232 (87.5)
Time period: 1991–6	1: slight redness	22 (8.3)
Length of follow-up: 6 months to 5 years	2: red and slightly moist tissue	8 (3)
Number of participants: 36	3: reddish and moist, granulation, revision may be indicated	1 (0.4)
Sample attrition/dropout: 1	4: removal of abutment due to infection	2 (0.8)
Indication for treatment:		
Bilateral agenesia of ear (16)	Total observations	265
Chronic otitis (20)	Implants removed	3 [osseointegration did not occur (2), head trauma (1)]
(44 fixtures: five with bilateral implants, three re-implantation)		
Age (years): 5.5–62.0		
Sex: NR		
Funding: NR		

NR, not reported.
a Individuals who lost two implants included only once here.
b Proportions based on those followed up, not whole sample.
c Classification by Holgers *et al.*[105]
d This is for the entire sample of 30 children, including 14 cases with bone-anchorage for auricular epistheses.
e Classification by Holgers *et al.*[106]
f Public body grants, details reported.

site (one) and trauma (one). Jacobsson and colleagues[103] followed 16 children for a median of 40 months (range 1–144 months). One (6.3%) implant was removed owing to non-integration. Portmann and colleagues[104] followed 36 participants with 44 fixtures (five with bilateral implants and three re-implantation) for between 6 months and 5 years. Three (6.8%) implants were removed, two because of lack of osseointegration and one because of head trauma.

Skin reactions

Skin reaction definitions used by the studies can be seen in *Table 16*. From a total of 1236 observations of skin reactions, Håkansson and colleagues[101] found that 93.2% had no skin irritation and 4.1% had slight redness. Red and moist tissue, and granulation tissue, were each observed on 1.3% of occasions. Infection leading to the removal of the abutment occurred in one case (0.1% of observations). A total of 265 observations were made in the study by Portmann and colleagues;[104] 87.5% of these had no irritation, 8.3% had slight redness and 3.0% had red and slightly moist tissue. Skin reactions Type 3 and Type 4 were observed on 0.4% and 0.8% of occasions, respectively. Mylanus and colleagues[102] did not report the total number of observations made, but reported that skin reaction Type 1 was observed 11 times in eight implants and Type 2 was observed seven times in six implants. Skin reaction Type 3 was never observed during the study, and a skin reaction leading to the loss of an implant was observed once. A life table of the cumulative proportion of implants that did not suffer from any skin reaction can be seen in *Table 16*. In the small study of 16 participants by Jacobsson and colleagues,[103] 91.7% of 108 observations had no irritation and 6.5% had slight redness. No grade 3 or 4 skin reactions were observed.

Summary of clinical effectiveness

No trials with a concurrent control group were identified; the 12 included studies[59,60,66,76–87] were either one-group cohort pre and post studies or cross-sectional audiological comparison studies. The methodological quality and quality of reporting of the included studies was weak, putting them at high risk of bias.

BAHA versus BCHA

Four cohort pre and post studies[77–82] provided a comparison of BAHAs and BCHAs. Improvements in sound field PTA and warble-tone thresholds were found with a BAHA by all three studies[77–81] that reported this outcome, but statistical analysis was reported by only one study ($p < 0.01$).[78] A statistically significant improvement in SRT in quiet and speech-to-noise ratio was found by one study,[81] while another study found no statistically significant difference in speech discrimination score.[78] Statistical analysis was not reported for other results, which included a 23% improvement in 100% speech audiometry discrimination in noise,[77] but little difference in speech recognition threshold.[79] Statistically significant improvements with large clinical effects were found with BAHAs compared with BCHAs for disability and handicap using the HHDI.[82]

BAHA versus ACHA

Five cohort pre and post studies[78–84] and one cross-sectional audiological comparison study[76] provided a comparison of BAHAs and ACHAs. Results for sound field pure-tone or warble-tone thresholds were inconsistent among the five studies reporting audiological data.[76,78–81,83,84] Where statistical analysis was undertaken, one study[83] found a statistically significant improvement in mean warble-tone thresholds (0.5–4.0 kHz) with a BAHA, while a different study[76] comparing each frequency individually found a statistically significant improvement with a BAHA at 1 and 8 kHz, but not at 0.25, 0.5, 2 or 4 kHz. The remaining three studies[79,80,83] did not compare data statistically; one study found that the ACHA was better at 0.25 kHz but there was improvement with BAHA at other frequencies,[76] another study found that the BAHA was better at 0.25 and 0.50 kHz, but the ACHA was better at higher frequencies,[79] while in another study data on average warble-tone thresholds (0.2–4.0 kHz) were described as 'comparable' between BAHAs and ACHAs.[83]

The direction of the effect was also unclear for speech audiometry, with some studies finding improved outcomes with the ACHA and some with the BAHA. One study reported better outcomes with the ACHA for speech discrimination scores,[83] and another for maximum phoneme score[79] or speech recognition threshold,[79] although statistical analysis was not conducted. A later publication by the same authors as the latter study found a statistically significant deterioration in SRT in quiet with BAHA ($p < 0.05$), but statistically significant improvement in speech-to-noise ratio ($p < 0.05$).[81] One study found no statistically significant difference in maximum phoneme score, but a statistically significant improvement in speech-to-noise ratio with BAHA.[84] Speech discrimination score was statistically significantly better with the BAHA in the congenital hearing loss group but not the chronic suppurative otitis media group in one study.[78] The final study reported an improvement in speech-in-noise with the BAHA described as 'large and clinically significant'.[84]

Statistically significant improvements with large clinical effects were found with BAHAs compared with ACHAs for disability and handicap using the HHDI.[82]

BAHA versus unaided hearing

Four studies[66,77,78,83] reported improvements in sound field thresholds and speech audiometry (where reported) with BAHA compared with unaided hearing, which were statistically significant where analysis was undertaken.

Unilateral versus bilateral BAHAs

An improvement in sound field average tone thresholds with bilateral BAHAs compared with unilateral BAHAs was found in adults[87] and a small group ($n = 3$) of children[59] with previous experience of BAHAs. Two studies found that speech recognition thresholds in quiet were statistically significantly lower with bilateral BAHAs,[60,87] although one study found similar results between unilateral and bilateral BAHAs.[86] Bilateral BAHAs produced better results when noise was presented from baffle/best side, but not when noise was presented from the shadow side.[60,86,87] Three studies found that localisation of sound was improved with bilateral BAHAs.[59,60,87] Two studies[60,87] reported the binaural masking level difference test and suggested that BAHAs give binaural hearing, although the validity of their methods is uncertain.

Adverse events

The included studies reported very limited data on adverse events. Prospective case series reported rates of loss of implants between 6.1% (9–25 months follow-up)[102] and 19.4% (median 6 years' follow-up).[100] The vast majority of participants in the prospective case series experienced no or minor skin reactions.

Chapter 4

Economic analysis

The aim of this section is to assess the cost-effectiveness of BAHAs with respect to conventional hearing aids or unaided hearing, and unilateral and bilateral BAHAs in adults or children with bilateral deafness who would be considered suitable for a BAHA. The economic analysis comprises:

- a systematic literature review of economic evaluations, QoL and cost studies in BAHAs and other potentially relevant comparator hearing aids
- the development of a de novo economic model and presentation of cost-effectiveness results.

Systematic review

Published economic evaluations

A systematic literature search was undertaken to identify full economic evaluations and cost studies that included BAHAs and other potentially relevant hearings aids (BCHAs and ACHAs). The methods for the systematic review are described in *Chapter 2*. Details of the inclusion and exclusion criteria are shown in *Chapter 2, Inclusion and exclusion criteria* and the search strategies are documented in *Appendix 2*.

A total of 225 potentially relevant publications were identified by the searches. No relevant full economic evaluations involving BAHAs were found after screening titles and abstracts. The searches identified 29 economic evaluations in hearing aids. None of the identified economic evaluations (19 in cochlear implants and 10 in other hearing aids) was found to be directly applicable to the aim of this study.

Two studies[107,108] that reported costs, but not outcomes, associated with BAHAs were reviewed for their relevance to estimating resource use and costing in our economic model. Catalano and colleagues[107] assessed the costs (in terms of participants' and physicians' time, as well as fees for treatment) of outpatient and inpatient insertion of BAHAs in a retrospective study with 19 US participants. However, the costs in the study were not directly applicable to the development of our economic model because of the US perspective of the study. Moreover, day case surgery would be the current service standard for surgical implantation, preparatory to fitting of the BAHA processor, in the NHS. Therefore, this study was not used to inform the economic model.

Watson and colleagues[108] in a retrospective analysis compared service use before and after BAHA insertion in 26 adults with suppurative otitis media exacerbated by behind-the-ear hearing aids. They identified a reduction in the number of treatments and visits after BAHA insertion compared with behind-the-ear hearing aids. As part of the analysis they outlined treatment protocols for people undergoing surgical implantation and post-surgical management (within the ENT clinic), as well as audiological management to fit and commission the BAHA processor in a UK district general hospital. This study was used, in conjunction with current service standards[109] relevant to the NHS and expert opinion, to identify the management pathway for individuals considered eligible for a BAHA as a basis for costing the intervention in our economic model (see *Resource use and cost data* for more details).

Unpublished economic evaluations

An unpublished, UK-based, economic evaluation of BAHAs conducted for an MSc thesis was identified.[110] This study does not strictly meet the inclusion criteria for the review, as it appears that a minority of participants had bilateral hearing loss (33.3% were reported as having bilateral hearing loss, the remainder were unilateral/single-sided or not stated). However, given that this is the only economic evaluation of BAHAs compared with conventional hearing aids that was identified, it is briefly reviewed below.

The initial sampling frame for the study was all adults (greater than 16 years old) undergoing primary BAHA implantation by the Departments of Audiology and Otolaryngology at University Hospitals Birmingham NHS Foundation Trust (UHB) between April 2007 and June 2008. All patients were invited to participate in the study and were sent the Health Utilities Index (HUI) 15-item self-completion questionnaire, along with a set of questions concerning current hearing aid use and duration of hearing impairment. Of the 147 eligible participants, 89 returned the first questionnaire (61% response rate). Of those 89 who completed the baseline questionnaire, 70 completed a second questionnaire (at least 3 months after the fitting of their BAHA).

The mean age of participants responding to the initial questionnaire was 55 years, with 44% of respondents being male. The mean time from surgery to receiving their BAHA was 2.8 months and the mean time from fitting of their BAHA to receiving the second questionnaire was 6.1 months.

Costs for BAHA provision were based on charges by the provider to PCTs. These were £5689 for the first year, to cover surgical and audiological assessment, implantation surgery, post-surgical care and acquisition and fitting of the BAHA sound processor, and a cost for the annual maintenance contract. The contribution of each component to this total charge was not reported. For subsequent years, the continuing cost of providing BAHAs was based on the annual maintenance contract fee of £1004. The analysis appears not to take account of costs associated with adverse events or treatment failure in BAHA users. The comparator group for the analysis was modelled by assumption, using participants' reported usage of hearing aids prior to receiving their BAHA [39/70 (56%) reported using one or more hearing aids prior to receiving their BAHA]. Costs of conventional hearing aids were based on provision of an NHS digital hearing aid (£260) replaced every 5 years.

Outcomes in the analysis were based on differences in utility (before and after BAHA provision), scored using the HUI3 and HUI2 algorithms, and participants' age-sex-specific life expectancy, derived from UK life tables. The mean HUI3 utility at baseline was 0.57 (95% CI 0.52 to 0.62) for all 89 respondents to the initial questionnaire and 0.59 (95% CI 0.53 to 0.65) for the 70 participants who responded to both outcome questionnaires. The mean HUI3 utility score for participants post BAHA was 0.66 (95% CI 0.60 to 0.72). It is unclear whether individual utility values or the mean values were used to derive QALYs, as the study report refers both to using 'pooled [utility] results' and to 'each subject[s]… utility scores' in calculating QALYs.

Table 17 reports the costs, QALYs and incremental cost-effectiveness ratio (ICER) for BAHAs compared with participants' previous hearing aid provision. Costs and outcomes are discounted at 3.5%.

A series of scenario analyses were reported, assuming that all participants in the modelled comparator group were using standard hearing aids (rather than the observed proportion, 56%) and adopting shorter time horizons. The study[110] also reported a subgroup analysis, breaking the

TABLE 17 Cost-effectiveness results from unpublished economic evaluation of BAHAs[108] compared with participants' previous hearing aid

	Previous hearing aid	BAHA	Difference	ICER (£/QALY gained)
Costs (£) (95% CI)	827 (644 to 1022)	21,430 (20,263 to 22,535)	20,604	17,610
QALYs (95% CI)	NR	NR	1.17 (0.50 to 1.91)	

NR, not reported.
95% CIs were derived using bootstrapping methods.[111]

total population of participants down by the main indication for treatment. The ICER results were generally robust to variables in the scenario analyses, with the ICER reducing fractionally (to £17,224 per QALY gained) for increasing the proportion of the modelled comparator group using conventional hearing aids. Reducing the time horizon of the model led to higher ICERs (£18,820 at 20 years, £22,097 at 10 years and £28,928 at 5 years). In the analysis by indication for treatment, BAHAs were least cost-effective in participants with bilateral CHL (£32,331 compared with £7459 for unilateral CHL and £19,391 for single-sided deafness). However, the number of cases included for each comparison was low (13–31 cases).

Published quality-of-life studies

In addition to the searches for economic evaluations and cost studies, a systematic literature search was undertaken to identify QoL studies of BAHAs and other potentially relevant hearing aids (see *Appendices 2* and *11* for details).

A total of 322 potentially relevant publications were identified by the searches. After screening titles and abstracts, only one relevant study (by Hol and colleagues,[82] discussed in *Chapter 3, Self-reported measures*) was identified. This study reported statistically non-significant differences before and after BAHA implantation using generic measures (SF-36 and EQ-5D), but statistically significant differences using a condition-specific measure (HHDI). The difference between the condition-specific and the generic health-related QoL instruments was probably due to a lack of hearing dimension on the EQ-5D and SF-36 questionnaires. As has been noted elsewhere,[112,113] the HUI3 appears to be a more suitable generic measure for QoL in a population with hearing difficulties rather than the EQ-5D or SF-36. Owing to a lack of sensitivity in the EQ-5D and SF-36 it was decided that the study by Hol and colleagues[82] should not be used in the decision model.

Southampton Health Technology Assessments Centre economic analysis

We developed a new model to estimate the cost-effectiveness of BAHAs in separate cohorts of eligible adults and children. In view of the lack of relevant clinical data (see *Chapter 3*), expert advice was sought to determine the comparator most appropriate to clinical practice. This suggested that the model should be limited to comparing BAHAs against BCHAs. The outcomes used in the model are in terms of cost per case and cost per successful implantation. An exploratory analysis using cost per QALY is also presented. Scenario and sensitivity analyses were undertaken to consider the impact of parameter and structural uncertainty.

Model type and rationale for model structure

The management pathway for individuals considered eligible for a BAHA, as outlined in the *Bone Anchored Hearing Aids Service Standards*[109] (discussed further in *Resource use and cost data*), indicates an initial phase of intensive activity (to assess eligibility, perform surgical implantation, fit and commission the BAHA processor) involving care from a multidisciplinary team, followed by a less intensive phase of long-term maintenance using the device. The clinical effectiveness section of this report (see *Chapter 3, Adverse effects*) has highlighted the risk of periodic occurrence of adverse outcomes including skin reactions or failure of the titanium implant, which may lead to revision surgery, a repeat of the original implantation procedure or possibly to people stopping using the BAHA. In addition to these longer term adverse event risks, there may also be short-term adverse events associated with the initial implantation procedure.[54,114] To take account of the changes in intensity of management, the periodic occurrence of adverse events and the potential for users to abandon the use of their BAHA, we developed a simple state transition model.

The model includes three states:

- success
- success with adverse outcome
- failure.

Individuals in the success state may experience an adverse event and move to the success with adverse outcome state, or they may remain in the success state. Individuals in the success with adverse outcome state may undergo surgical or non-surgical management (depending on the nature of the adverse event) and may move to the success state (if the adverse outcome resolves), may move to the failure state (if they choose not to continue with the BAHA) or may remain in the success with adverse outcome state. Those in the failure state may remain there or may elect for a repeat of the original implantation procedure (with success, success with adverse outcome or failure as possible outcomes).

This conceptual model was implemented as a decision tree with embedded Markov processes using the software package TREEAGE PRO (Williamstown, MA, USA), as shown in *Figure 2*. People enter the model with bilateral deafness, currently managed using BCHA, but are considered potentially suitable for BAHA. If they choose to accept this option, they follow the branch from the root node of the decision tree marked BAHA and will immediately be assigned all the costs related to assessing eligibility for BAHA and the costs of surgical implantation. The BAHA node is associated with a Markov process containing four health states (the failure state described above has been split into a temporary 'failure' state, where people make a decision whether to accept a re-operation, and a CeaseBAHA state for those who decide not to undergo a repeat operation), plus an absorbing state marked death. This does not indicate that BAHA use or implantation surgery is expected to be associated with a significant risk of death, but is included to take account of general, all-cause, mortality over time in the population being modelled. People undergoing initial implantation surgery are allocated to potential short-term outcomes (success, success with adverse outcome and failure) on the basis of probabilities estimated from the literature. Success with adverse outcome or failure might be expected to incur higher costs or poorer outcomes (in terms of QoL) than successful surgery. Individuals whose initial implantation procedure was successful are subject to risks of adverse events (in the model these are limited to skin reactions at site of implantation and loss of integration of titanium implant) for each cycle of the model. Skin reactions may be treated non-surgically (grades 1 or 2, assumed managed by cleaning regimes and antibiotics) or surgically (grade 3 requiring revision surgery

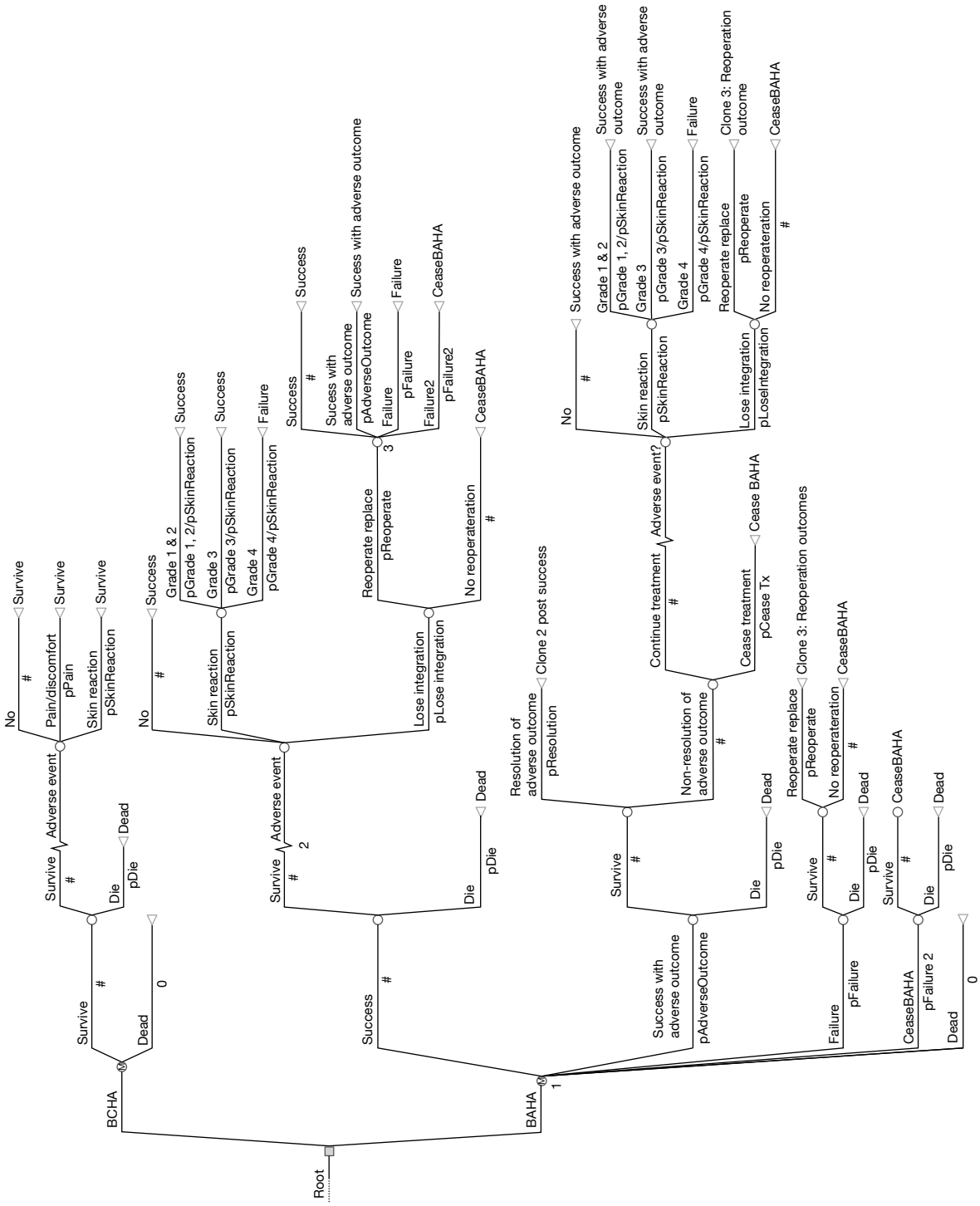

FIGURE 2 Outline of model.

and grade 4 resulting in removal of implant). Loss of bone integration is managed by a repeat operation, although the model allows for people to choose not to undergo repeat surgery, with a similar range of outcomes as for the initial operation.

People whose initial surgery resulted in success, but with adverse outcome, may choose to cease treatment, in which case they move to the CeaseBAHA state. If they continue treatment, the adverse outcome may resolve, in which case they would move to the success state. If the adverse outcome is not resolved they continue similarly to those in the success state, but will stay in the success with adverse outcome arm of the model.

Individuals whose initial surgery was a failure (defined as failure to achieve bone integration) or whose implants subsequently lose bone integration may choose to have a re-operation with a similar possible range of outcomes as initial surgery.

The model has an annual cycle and the principal outcome for the model is the incremental cost of BAHAs compared with BCHA. BAHA users who choose not to undergo repeat operations, owing to loss of bone integration or severe skin reactions, are identified as treatment failures and are assumed either to revert to BCHA or to continue unaided.

Baseline cohort

The population in the base-case analysis are those with bilateral deafness who were already provided with BCHAs, but are considered for BAHA owing to convenience and improved wearability. For the purposes of the model, adults are considered to be aged ≥ 18 years. Children are those less than 18 years of age. The sex composition of the included cohorts reflects the general population and is applicable only in calculation of death rates.

Data sources used in the model

Clinical effectiveness data

Gain in hearing

Chapter 3 of this report, *Assessment of clinical effectiveness: BAHA versus BCHA*, presents the systematic review of outcomes for BAHAs compared with BCHA in terms of audiological outcomes. No quantitative summary of the reported outcomes could be produced and there was little consistency of reporting between included studies. Consideration was given to using one of these outcomes for inclusion in the economic model, but it is not clear from the included studies what these outcomes mean to those receiving BAHAs. We were not able to identify any robust methods to map from audiological outcome measures to QoL measures. As a result, the outcomes in the model are based on potential gains using a generic QoL scale that is sensitive to changes in hearing. *Quality of life* reports the approach adopted for outcome assessment (potential QoL gain) in the model.

Adverse events

Table 18 presents estimates of the annual risk of implant failure (loss of bone integration), using data reported and discussed in *Chapter 3, Prospective case series*. Data in columns 1–5 of *Table 18* were extracted from the study by Bonding,[100] which reported the number of patients followed up for up to 7 years and the count of implants lost each year. Annual risk of failure (column 6) has been estimated from the reported cumulative success proportions, using the declining exponential approximation to life expectancy (DEALE) method,[115] which estimates a constant risk over time. The estimated annual risk of failure, beyond year 2, varies between 3.8% and 4.5%.

Figure 3 illustrates the fit of the predicted annual risk [based on the cumulative success proportion at 7 years (0.741)] to the data reported by Bonding,[100] treating the cumulative success proportions as Kaplan–Meier survival estimates. This figure suggests that applying the constant risk may overestimate the failure rate in the first 2 years from implantation. For subsequent years

TABLE 18 Loss of implants, estimated cumulative success and annual risk of failure

Time (years)	*n*	Implants lost	Success (% success per year)	Cumulative success (%)	Annual risk of failure[a] (%)
0	31	0	100.0	100.0	
1	31	0	100.0	100.0	0.00
2	29	0	100.0	100.0	0.00
3	26	3	88.5	88.5	4.09
4	21	1	95.2	84.2	4.28
5	19	1	94.7	79.8	4.51
6	15	0	100.0	79.8	3.76
7	14	1	92.9	74.1	4.28
>7	10	0	100.0	74.1	

a Annual failure risk at each time point is calculated based on the cumulative success up to that time point, e.g. annual failure risk at 3 years was calculated as $1/3 \times -\ln(0.885)$.

Annual failure risk (R) estimated using the DEALE method: $R = 1/t \times -\ln(S)$, where $\ln(S)$ is natural log of cumulative survival at time t.

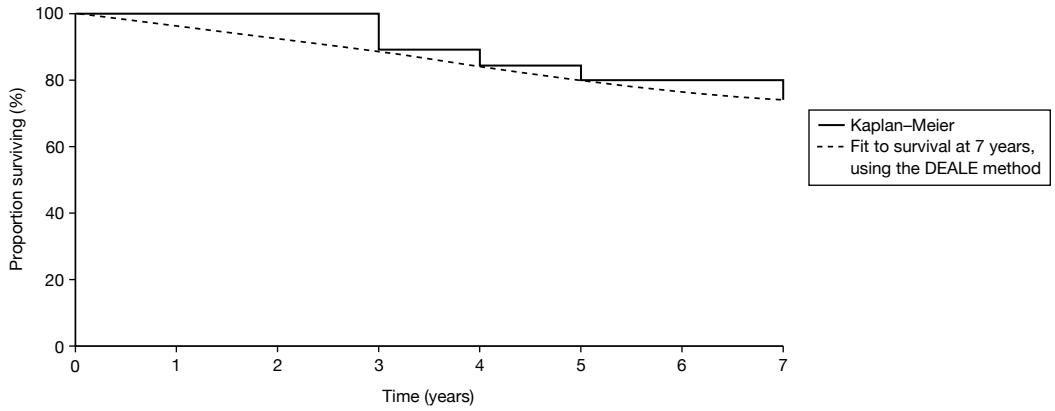

FIGURE 3 Goodness of fit of annual risk of titanium implant failure (derived using the DEALE method) and Kaplan–Meier estimates.

there appears to be reasonable agreement between the Kaplan–Meier estimate and the line of fit using the DEALE method. The annual failure risk estimated at year 7 (4.28%) is applied in the base-case analysis.

The clinical data discussed in *Chapter 3, Prospective case series*, typically report the proportion of BAHA users experiencing skin reactions, rather than an estimate of rates. The latter would be more appropriate given the differential follow-up periods shown for participants in all of the included studies. *Table 19* reports our estimates of the rates of skin reactions by grade for people using BAHAs, based on the reported counts and cumulative person-years of observation in each study shown in *Chapter 3, Prospective case series* (95% CIs have been estimated using exact confidence limits for Poisson counts[116]). The estimated rates show considerable variation, with substantially higher rates of all skin reactions, except grade 3, reported by Portmann and colleagues[104] and Mylanus and colleagues,[102] than in other studies. It is difficult to interpret the significance of the results of these studies or suggest explanations for variation between studies, as little information is provided on characteristics of populations in the studies [for example, the proportion of paediatric cases or the proportion of subjects (adult or children) with learning disability]. The studies are comparatively small and are not all consistent in the categorisation of grade of skin reaction. We therefore adopted rates estimated from the study by Håkansson and colleagues[101] in our model, as this is the largest study using standard definitions for grades of skin

TABLE 19 Estimated rates of skin reaction, by grade, in included studies

Source	Person-months at risk (*n*)	Count by grade			Rate per 100 person-years at risk (PYAR) (95% CI)		
		Grades 1 and 2	Grade 3	Grade 4	Grades 1 and 2	Grade 3	Grade 4
Holgers *et al.* 1988[105]	1515 (64)	15	5	1	11.88 (6.65 to 19.60)	3.96 (1.29 to 9.24)	0.79 (0.02 to 4.41)
Håkansson *et al.* 1990[101]	5542 (164)	67	16	1	14.51 (11.24 to 18.42)	3.46 (1.98 to 5.63)	0.22 (0.01 to 1.21)
Jacobsson *et al.* 1992[103]	586 (15)	8	0	0	16.38 (7.07 to 32.28)	0.00 (0.00 to 7.55)	0.00[a] (0.00 to 7.55)
Mylanus *et al.* 1994[102]	476 (33)	18	0	1	45.34 (26.89 to 71.72)	0.00 (0.00 to 9.30)	2.52 (0.06 to 14.05)
Portmann *et al.* 1997[104]	1338 (41)	30	1	2	26.91 (18.15 to 38.41)	0.90 (0.02 to 5.00)	1.79 (0.22 to 6.48)

a One implant was removed owing to non-integration at 4 months. Rate per 100 PYAR = 2.05 (95% CI 0.05 to 11.41).

reaction. The sensitivity of results to this assumption is addressed in scenario analyses, in which estimates based on other included studies are applied.

The impact of these adverse events is included in cost estimates used in the model (see *Resource use and cost data* for discussion of the costs of managing and treating adverse events). No direct estimate of the impact of adverse events on QoL was found in our literature searches and these are not included in the base-case analysis.

All-cause mortality

The most recent UK life tables were used in the model to estimate the percentage of the cohort dependent on age that dies in each cycle of the model. No increase in the mortality rate was assumed to be applicable to the baseline cohort.

Quality of life

Exploratory analysis of hearing improvements from HUI3

The lack of useable QoL data for people using BAHAs in the studies discussed earlier in this chapter (see *Systematic review*) and the absence of any robust methods to map from outcomes identified in *Chapter 3, Assessment of clinical effectiveness: BAHA versus BCHA*, to QoL/health-state utility led to further methods being sought to link potential benefits from the use of BAHAs. An exploratory analysis was undertaken using the difference between the levels of the hearing attribute in the HU13 classification system.

The HUI is a generic, preference-based system for measuring health status and comprises a health-state classification system and formulae for calculating utility scores. The classification systems consist of a number of attributes, each representing a particular dimension of health status (such as pain or emotion). The attributes are divided into levels of increasing impact on health status. The HUI is available in three versions which, although they have some common attributes, have some notable differences. These determine the version that is most appropriate for a given study group. For example, the HUI2 contains a single 'sensation' attribute relating to sight, hearing and speech, whereas the HUI3 contains separate attributes for each of these senses. As a result of the inclusion of separate attributes for the sensations and demonstrated improved sensitivity over other generic QoL measures,[112] the HUI3 has been used in previous

economic evaluations of hearing aid devices[117–119] and for descriptive studies of QoL before and after hearing aid provision.[113] As was noted in our review of QoL studies, the HUI appears to be a more suitable generic measure for QoL in a population with hearing difficulties than the EQ-5D or SF-36. *Table 20* presents the level descriptions for the hearing attribute for the HUI3 classification system.

The HUI3 has structural independence of the attributes in its classification system.[74] This is important for the explanatory analysis of QoL benefit as it is possible to strip out the other attributes and concentrate on potential changes in hearing gain as described in the classification system. However, any results derived from these methods should be interpreted with caution owing to their weak methodological basis. Furthermore, severe hearing loss and associated improvement in hearing and wearability from the use of BAHA could possibly affect other attributes in the HUI3 classification system, such as cognition.[112] Therefore, it is possible that this will underestimate the real gain in QoL that may be experienced from using a BAHA.

An estimate of potential utility gain was calculated from the difference in utility between levels in the hearing attribute of the HUI3, using the scoring algorithm for the multiattribute utility function on the Dead–Healthy Scale reported by Furlong and colleagues.[121] This was calculated while keeping all other attributes fixed at 'level 1'. For example, the associated gain in utility for moving from level 6 of the HUI3 hearing attribute, at which the respondent is 'unable to hear at all', to level 5, at which the respondent is 'able to hear what is said in conversation with one other person in a quiet room with a hearing aid, but unable to hear what is said in a group conversation with at least three other people with a hearing aid', is 0.178. Put in terms of overall health-state utility, this is a move from 0.465 to 0.644 on the scale, where 0 is dead and 1 is full health. In *Table 21* the difference in utility gains for moving between each of the hearing levels is calculated.

TABLE 20 Level descriptions for the HUI3 hearing attribute[120]

Level	Description
1	Able to hear what is said in a group conversation with at least three other people, without a hearing aid
2	Able to hear what is said in a conversation with at least one other person in a quiet room without a hearing aid, but requires a hearing aid to hear what is said in a group conversation with at least three other people
3	Able to hear what is said in a conversation with one other person in a quiet room with a hearing aid and able to hear what is said in a group conversation with at least three other people with a hearing aid
4	Able to hear what is said in a conversation with one other person in a quiet room without a hearing aid, but unable to hear what is said in a group conversation with at least three other people even with a hearing aid
5	Able to hear what is said in a conversation with one other person in a quiet room with a hearing aid, but unable to hear what is said in a group conversation with at least three other people even with a hearing aid
6	Unable to hear at all

TABLE 21 Potential health state utilities and utility gain from changes in hearing levels by providing hearing aids to an individual who is bilaterally deaf, using the HUI3 classification system

Hearing level on the HUI3 hearing attribute (health state utility)				
Before BAHA or hearing aid		After BAHA or hearing aid		Utility gain from BAHA or hearing aid
Level 6	(0.465)	Level 6	(0.465)	0.000
Level 6	(0.465)	Level 5	(0.644)	0.178
Level 6	(0.465)	Level 3	(0.849)	0.384

We assumed that the QoL benefit from improved hearing could be proxied by levels of the hearing dimension in the HUI3, that identical gains were potentially achievable through use of BAHA and BCHA, and that BAHA-eligible patients were initially at level 6 (unable to hear at all). The minimum utility gain is therefore 0.178 (level 6 to level 5) and the maximum 0.384 (level 6 to level 3). Differences between the utility associated with use of BAHA or BCHA were assumed to be realised through differences in the wearability of alternative devices. For this we required information on participants' use of their previous device and the BAHA.

Included studies were reviewed for information on post-implantation use of BAHAs (hours per day and days per week that the device was used) and for use of previous hearing aids. Additional publications reporting use of BAHAs and BCHAs were identified from the reference lists of included studies and using targeted searches. The results of these searches are summarised in *Table 22*, indicating (where reported) the key characteristics of participants included in the studies.

Only one study,[82] of relatively small size, reported results for both BAHA and BCH and this reported very limited information, simply the proportion of people using the hearing aid for more than 8 hours per day. For the base-case analysis it was assumed that this was a reasonable characterisation of the relative use of BCHA and BAHA, with BAHA use being approximately 10% greater than use of BCHA. The sensitivity of results to this assumption was tested in

TABLE 22 Reported usage of BAHA and previous hearing aid

Study	Participant characteristics	Questionnaire	Usage
de Wolf *et al.* 2009[122]	135 (of 211) BAHA compact users aged 18–77 years. 100 with bilateral conductive/mixed hearing loss; 23 with unilateral conductive/mixed hearing loss; 12 with unilateral conductive/mixed hearing loss/other ear deaf	IOI-HA	BAHA use for greater than 8 hours per day **Age range (years)** **%** 18–40 82.1 41–60 84.1 >60 70.7
Badran *et al.* 2006[123]	117 (of 152) adults who 'underwent BAHA procedure for greater than 6 months'. 64% chronic otitis media and 21% chronic otitis externa and/or acquired stenosis	Entific Medical Systems questionnaire	81% reported using BAHA everyday **Hours BAHA used per day** **%** >8 78 4–8 15 2–4 3 <2 3
Hol *et al.* 2004[82]	56 consecutive adult patients with acquired conductive or mixed hearing loss (20 using conventional BCHA)	Not stated	Of those previously using BCHA, 100% reported using their BAHA for greater than 8 hours per day compared with 90% for their previous aid
Dutt *et al.* 2002[124]	227 (of 351) children and adults implanted at Birmingham implant otology unit.[a] Cause of hearing loss not stated in this publication	Entific Medical Systems questionnaire	95% reported using BAHA everyday **Hours BAHA used per day** **%** >8 86 4–8 10 2–4 5 <2 3
Cooper *et al.* 1996[78]	68 (of 106) adults. 43 with CSOM (24 using ACHA and 19 using BCHA prior to BAHA) and 25 with congenital CHL (9 using ACHA and 16 using BCHA prior to BAHA)	Not stated	95.5% reported using BAHA greater than 8 hours per day

CSOM, chronic suppurative otitis media.
a Much higher non-response in children (under 16 years) – 60% non-response vs 11% non-response in adults.

deterministic sensitivity analyses using the upper and lower values for the exact binomial CIs as shown in *Table 23* and in scenario analyses.

The utility associated with each device, in the model, was therefore a weighted average of the unaided utility ($u_{unaided}$) and the aided utility (u_{aided}), with the weight based on the proportion of time users were able to wear their device (p_{wear}). Hence the total utility for a given time period was defined as in *Equation 1*.

$$u_{unaided} \times (1 - p_{wear}) + u_{aided} \times p_{wear} \quad \text{[Equation 1]}$$

Another potentially relevant dimension of the HUI is pain and this was incorporated in scenario analyses. This item has five levels as described in *Table 24*.

In the scenario analysis we assumed that the BCHA may be associated with 'mild to moderate' pain (level 2 of the HUI pain attribute) for the majority of users. In this scenario analysis pain is also included for BAHA users who experience pain requiring removal of the BAHA implant, assuming HUI level 5 pain for the cycle following implantation and leading to removal (in effect a very large penalty of 0.55 arising from intolerable pain).

Resource use and cost data

Both resource use and cost data are taken from an NHS perspective. The resource use associated with the implantation of BAHAs was identified using Watson and colleagues's[108] retrospective study of resource use and the *Bone Anchored Hearing Aid Service Standards*.[109] These sources were used to develop a resource use protocol (illustrated in *Figure 4*) which was discussed with clinical experts.

Four distinct phases of costs associated with BAHAs were identified. These cost phases were defined as being associated with:

- assessment of surgical and audiological eligibility
- surgery
- post-surgical management (up to 12 months following the initial surgical procedure and included fitting of external processor)
- long-term management.

TABLE 23 BAHA and BCHA usage, applied in base-case analysis

Hearing aid type	*n*	Reported usage (%)	95% CI (exact binomial)
BCHA	20	90	68.3% to 98.8%
BAHA	20	100	83.2% to 100%

Exact binomial CI calculated on assumption that 18 participants (i.e. 0.9 × 20) reported using their previous aid (BCHA) for more than 8 hours per day.

TABLE 24 Levels, description and scoring of the HUI pain dimension

Level	Description
1	Free of pain and discomfort
2	Mild-to-moderate pain that prevents no activities
3	Moderate pain that prevents a few activities
4	Moderate-to-severe pain that prevents some activities
5	Severe pain that prevents most activities

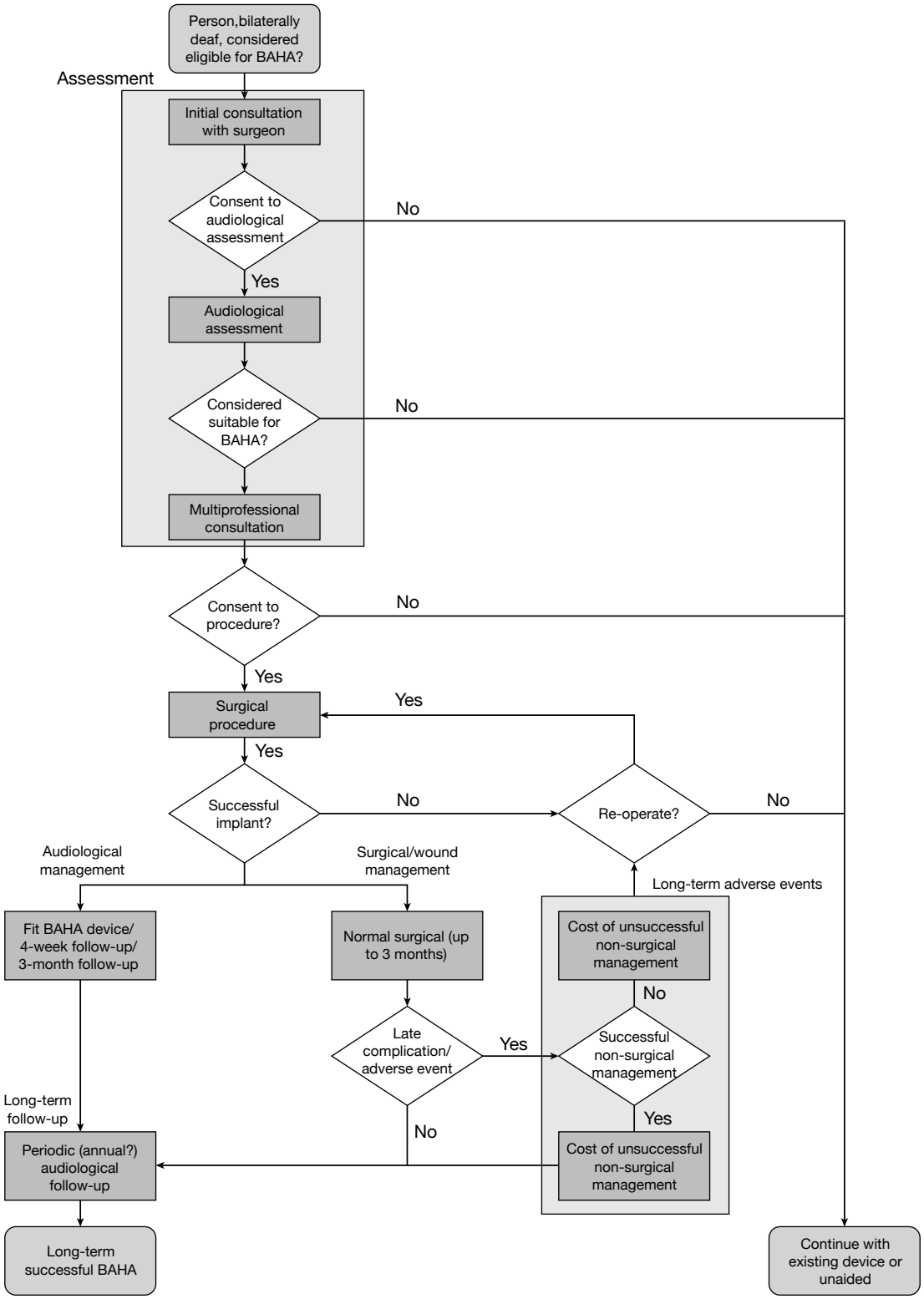

FIGURE 4 Resource use protocol for patients considered eligible for a BAHA.

All cost data and relevant sources are given and discussed in turn below. All unit costs and cost-effectiveness results are expressed in 2009 pound sterling.

Assessment of surgical and audiological eligibility costs

Potential BAHA users have an initial consultation with the specialist BAHA surgeon. This includes an examination of the middle and external ears and determination of the aetiology of hearing loss, as well as an assessment of the individual's general medical status and suitability for BAHAs. The cost of consultation with the surgeon was taken from the NHS reference costs.[43] An ENT outpatient first attendance cost of £110.78 was used for adults and a paediatric ENT outpatient first attendance cost of £131.69 was used for children.

An audiological assessment is carried out by an audiologist, who assesses current middle and external ear status. AC and BC are tested using pure-tone audiometry. There may be an evaluation of the user's current hearing aid provision, if applicable. The cost of the audiological assessment (£57.48) is taken from the NHS reference costs.[43]

The final stage of the assessment phase involves a multiprofessional consultation (£147.36) to agree to the individual's eligibility for a BAHA and to gain consent for surgery.

The breakdown of unit costs for the initial assessment of surgical and audiological eligibility for adults and children is given in *Table 25*. The total cost of the assessment of surgical and audiological eligibility cost was £315.63 for adults and £336.53 for children.

Surgery costs

The cost of one-stage surgery to implant the BAHA was taken from the NHS reference costs for a day-case BAHA operation with a cost of £2004.57 for adults. The paediatric two-stage method for implantation of a BAHA was assumed to cost twice the NHS reference cost for BAHA surgery and therefore had a cost of £4009.14.[43] Additional costs including the fixture and abutment costs and additional consumables from the surgery are given in *Table 26*. These had a total cost of £989.50. These costs are current list prices and were provided by the UHB. The breakdown of unit cost of surgery for adults and children is given in *Table 26*. The total cost for surgery in adults was estimated at £2994.07 in adults and £4998.64 in children.

Post-operative costs

Watson and colleagues[108] identified three obligatory visits to ENT and audiology after surgery. This was verified by our experts. These visits consist of changing the dressing (removal of the mastoid bandages after 24 hours), removal of healing disc and stitches and an assessment to ensure osseointegration at around 3 months. The cost for these three visits was taken from the NHS reference costs.[43] There appears to be practice variation at this stage with either the

TABLE 25 Unit and total cost of assessment of surgical and audiological eligibility for adults and paediatrics

Item	Adult unit costs (£)	Paediatric unit costs (£)
Initial consultation with surgeon[a]	110.78	131.69
Audiological assessment[b]	57.48	57.48
Multiprofessional consultation[c]	147.36	147.36
Total cost	**315.63**	**336.53**

Source: *NHS reference costs 2007/08*,[43] inflated to 2008–9 values.

a Consultant led: first attendance non-admitted face to face.

b Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code for fitting of hearing aids and counselling: assessments (Code AS1A).

c Consultant led: first attendance multiprofessional non-admitted face to face.

TABLE 26 Unit and total cost of surgery for adults and paediatrics

Item	Adult unit costs (£)	Paediatric unit costs (£)
BAHA surgery procedure[a]	2004.57	4009.14
Guide drill[b]	15.50	15.50
Countersink[b]	29.50	29.50
Fixture[b]	310.00	310.00
Abutment[b]	520.00	520.00
Cover screw[b]	62.00	62.00
Healing cap[b]	23.50	23.50
Dermatome blade[b]	29.00	29.00
Total	**2994.07**	**4998.64**

Sources:
a Healthcare Resource Group Code CZ26Z bone-anchored hearing aids: day case. *NHS reference costs 2007/08*,[43] inflated to 2008–9 values.
b UHB.

surgeon or a specialist nurse undertaking the assessments. It was assumed that in the base case the surgeon undertook the assessment. This uncertainty was explored in a scenario analysis. Therefore, the cost of each consultation was taken from the NHS reference costs for an ENT outpatient follow-up attendance with a cost of £72.11 for adults and £90.93 for paediatric ENT[43] (*Table 27*).

The first audiological attendance is to fit the BAHA device. This includes adjusting the device settings based on audiometric results and the user's response, and recording the output of the device using electro-acoustic methods. Furthermore, consideration is given to fitting BAHA accessories, discussing the user's expectation and providing information on the management of the device. The second audiological follow-up occurs at around 4 weeks and includes a site and abutment inspection when the tightness of the abutment is checked using a torque driver. A record of the output from the device using electro-acoustic methods is taken and this is compared with the measure taken at the time of fitting. The third audiological follow-up occurs at 3 months and includes a record of the output from the device to further assess performance and outcome measurement. An unaided and aided sound field audiometry test is undertaken and post-operative questionnaires are administered. The costs of the three audiological visits were taken from the NHS reference costs and were assumed to consist of a fitting cost of £64.80 and two follow-up visits of £50.17 each.[43] The breakdown of unit cost of post-operative follow-up for adults and children is given in *Table 27*. The total cost of post-operative follow-up for adults is £381.47 and £437.91 for children.

The cost of the BAHA processor is also included in this phase. The average cost of four of the BAHA processors currently used in the NHS and the cost of their maintenance plans are reported in *Table 28*. The average cost for both the processor and the maintenance plan was used in the model owing to uncertainty over which processors are currently used most in the NHS. A sensitivity analysis of the range of costs of the processor and first year maintenance plan was used to explore this uncertainty.

The total cost of the post-operative follow-up and the processor (and maintenance plan) after surgery for adults is £3308.97 for adults and £3365.41 for children.

The cost per user at each stage of a successful implantation of a BAHA in the first year is given in *Table 29*. This includes the cost of audiological assessment, surgery costs and post-operative

TABLE 27 Unit and total cost of post-operative follow-up

	Unit costs (£)	
Item	**Adults**	**Paediatrics**
ENT outpatient visit to change dressing[a,b]	72.11	90.93
ENT outpatient visit to remove healing disk and stitches[a,b]	72.11	90.93
ENT outpatient visit to ensure osseointegration[a,b]	72.11	90.93
Initial fitting of the BAHA by an audiologist[c]	64.80	64.80
4-week audiological follow-up of BAHA fitting[d]	50.17	50.17
3-month performance of processor by an audiologist[d]	50.17	50.17
Total costs	**381.47**	**437.91**

Source: *NHS reference costs 2007/08*,[43] inflated to 2008–9 values.

a Consultant led: follow-up attendance non-admitted face to face using service codes for ENT (Code 120).

b Consultant led: follow-up attendance non-admitted face to face using paediatric ENT (Code 215).

c Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code: fitting of hearing aids and counselling: fitting (Code AS1FA).

d Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code: fitting of hearing aids and counselling: follow-up (Code AS1FU).

TABLE 28 Unit and average cost of processors and maintenance plans for each BAHA

BAHA model	Cost of processor (£)	Cost of maintenance plan (£)	Total cost (£)
Divino	1820.00	610.00	2430.00
Intenso	1980.00	665.00	2645.00
Cordelle	1970.00	670.00	2640.00
BP 100	2995.00	1000.00	3995.00
Average cost	**2191.25**	**736.25**	**2927.50**

Source: UHB and Southampton University Hospitals Trust (SUHT).

TABLE 29 Total costs per patient for successful BAHA implantation in the first year

Stage of BAHA implantation	Total cost adults (£)	Total cost paediatrics (£)
Total initial audiological assessment cost (see *Table 25*)	315.63	336.53
Total surgery costs (see *Table 26*)	2994.07	4998.64
Total post-operative surgery costs, first year (see *Tables 27* and *28*)	3308.97	3365.41
Total cost for BAHA implantation	**6618.68**	**8700.59**

surgery costs with a total cost estimated at £6618.68 for adults and £8700.59 for children per successful implantation of a unilateral BAHA.

Long-term follow-up costs

Expert opinion and current service standards[109] suggest that BAHA users will have periodic audiological follow-ups. In addition to the annual audiological follow-up, those who continue to use a BAHA will have the sound processor replaced every 3 years under the annual maintenance plan. At the annual audiological assessment a record of output from the device is measured using an electro-acoustic test and this is compared with the measure taken at time of fitting. Performance and outcome measurements are undertaken with unaided and aided sound field audiometry tests. Post-operative questionnaires may also be administered at this stage (*Table 30*).

TABLE 30 Long-term costs of BAHA use

Item	Frequency	Unit costs adults (£)	Unit costs paediatrics (£)
Audiological assessment	Every year	50.17[a]	50.17[a]
BAHA maintenance plan	Every year	736.25[b]	736.25[b]
Total long-term costs every year		**786.42**	**786.42**

Sources:
a *NHS reference costs 2007/08*,[43] inflated to 2008–9 values. Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code: fitting of hearing aids and counselling: follow-up (Code AS1FU).
b UHB and SUHT – see *Table 28*.

Adverse events costs

Two types of adverse events were identified from the review of the literature and confirmed by our clinical experts: skin irritation associated with the implantation of the BAHA and a loss of osseointegration of the fixture. The resource use associated with grade 1 or grade 2 skin irritation (using the Holgers and colleagues grading system[105]) was assumed to consist of an outpatient visit for dermatological care and antibiotics to treat the reaction. The costs of an outpatient dermatology visit was taken from the NHS reference costs, with a cost £118.10 for adults and £134.82 for children.[43] The antibiotic steroid cost of £5.40 was taken from the *British National Formulary* (BNF).[125] The total estimated cost of treating a grade 1 or grade 2 skin reaction is £123.50 for adults and £140.22 for children (*Table 31*). Grade 3 skin reactions were assumed to require surgical revision and were costed as intermediate skin procedures, provided as day cases.[43] Grade 4 skin reactions were assumed to require removal of the skin-penetrating implant and were costed using the reference cost applied for the initial day case surgery (see *Table 26*), excluding the costs of surgical consumables required for the initial implantation.

If loss of bone integration occurs, then there is a cost of re-operation surgery to re-implant the fixture and an associated post-operative surgery cost. This was assumed to be the same as the initial surgical operation and post-operative follow-up, as calculated above (see *Table 29*). The breakdown of costs for skin irritation and loss of osseointegration are reported in *Table 31*. The overall total cost of a loss of osseointegration was £4111.80 for adults and £6172.80 for children.

Costs associated with comparator pathways

BCHA costs People considered eligible for provision of BCHA were assumed to undergo an initial audiological assessment (including assessment of middle and external ear status as well as testing of AC and BC using pure-tone audiometry) similar to that for those considered suitable for BAHAs. People receiving a BCHA were assumed to have a single attendance to fit the BCHA with a follow-up visit at 3 months to assess the performance of the hearing aid. In subsequent years individuals were assumed to attend once a year for follow-up, with replacement of the hearing aid occurring every 5 years.

A range of costs was provided for the BCHA device itself, from a low cost of £117–183 for a body-worn aid (which is attached to the transducer via a cord) to £250 for a more cosmetically appealing option, incorporating a behind-the-ear hearing aid constructed onto the headband to drive the vibrator/transducer (using an NHS digital power aid). A higher cost of £300–350 was provided for proprietary behind-the-ear devices (*Table 32*). These costs were supplied by two NHS providers (see *Table 32*) and reflect current NHS purchasing arrangements for BCHA, rather than manufacturers' list prices for any particular devices.

The total cost of providing a BCHA was estimated as between £289 and £522 for the first year (depending on the type of aid provided), with long-term costs of £50.17 (for an annual

TABLE 31 Cost of adverse events

Skin reaction		Cost, adults (£)	Cost, paediatrics (£)
Grades 1 and 2	Dermatologist[a]	118.10	134.82
	Antibiotic steroids	5.40	5.40
	Total	**123.50**	**140.22**
Grade 3	Revision surgery[b]	663.66	663.66
Grade 4	Day case surgery to remove implant[c]	2004.57	2004.57
Loss of bone integration			
Re-operation[d]		2994.07	4998.64
Post-operative follow-up, year 1[e]		1117.72	1174.16
Total		**4111.80**	**6172.80**

Source: *NHS reference costs 2007/08*,[43] inflated to 2008–9 values.
a Dermatology: consultant led: first attendance non-admitted face to face (Code 330).
b Intermediate skin procedure without complication and comorbidities (Healthcare Resource Group Code JC04C) as day case.
c Assume same cost as initial surgery.
d See *Table 26*.
e See *Tables 27* and *28*. Excludes cost of BAHA sound processor.

TABLE 32 Unit and total costs for provision of BCHAs

Item		Unit costs, adults (£)	Unit costs, paediatrics (£)
Initial audiological assessment costs[a]		57.48	57.48
Fitting costs[b]		64.80	64.80
Cost of BCHA device (£)	Low (body-worn)[c]	117–183	117–183
	Low (behind-ear worn)[c]	250	250
	High[d]	300–350	300–350
Post-fitting, follow-up assessment (3 months) of device by an audiologist[e]		50.17	50.17
Total	Low (body-worn)	289–355	289–355
	Low (behind-ear worn)	422	422
	High	472–522	472–522

Sources:
a Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code for fitting of hearing aids and counselling: assessments (Code AS1A).
b Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code: fitting of hearing aids and counselling: fitting (Code AS1FA).
c SUHT.
d UHB.
e Audiological services: fitting of hearing aids and counselling (including tinnitus); using currency code: fitting of hearing aids and counselling: follow-up (Code AS1FU).

audiological assessment) and a replacement cost (for a new device, fitting and post-fitting assessment) of between £232 and £465 every 5 years.

Perspective and time horizon

The perspective of the cost-effectiveness analysis is that of the NHS and Personal Social Services (PSS). The analysis has adopted a medium-term horizon of 10 years. This is shorter than the lifetime horizon proposed in the protocol for this review. However, it is long enough for differences between the two cohorts to become apparent, but avoids extrapolating too far beyond the available clinical data (for example, clinical data on adverse events report outcomes at 7 years, for implant survival, and a maximum of 14 years for other outcomes).

Discounting

Both costs and outcomes were discounted using a 3.5% discounting rate, as currently recommended by the UK Treasury for public sector appraisal.[126]

Assessment of uncertainty

The purpose of this analysis is to test the robustness of the cost-effectiveness results to variation in structural assumptions and parameter inputs. A deterministic sensitivity analysis (DSA) was used to address particular areas of uncertainty in the model. We investigated the uncertainties around the probability, resource use and cost estimates that were expected, a priori, to have a disproportionate impact on the study results, by applying ranges around the point estimates used in the base-case analysis. Scenario analysis was used to address the uncertainty associated with the choice of data source adopted for parameter values in the base case and the structural assumption that patients who stop using their BAHA switch to an alternative hearing aid or continue unaided.

Parameter uncertainty was addressed using probabilistic sensitivity analysis (PSA). Probability distributions are assigned to the point estimates used in the base-case analysis. Variables included in the PSA, the sampling distribution and the parameterisation of the sampling distributions are reported in *Appendix 13*.

Summary of assumptions and input parameters used in the model

Table 33 summarises the probabilities included as input parameters to the model and is predominantly concerned with adverse events associated with BAHA provision (for full details of potential data sources and selection of parameter inputs see *Clinical effectiveness data*). The table includes 95% CIs, used as upper and lower limits in deterministic analyses.

Table 34 summarises the input parameters used to estimate potential utility gain from aided hearing and usage of BAHA and comparator hearing aids. These assumptions are relevant only to the augmented base case used in the exploratory cost-effectiveness analysis reported in *Exploratory cost-effectiveness analysis*. Full details and the rationale for adopting these input values are presented in *Quality of life*.

Table 35 summarises the costs associated with BAHA provision included as input parameters to the model (for full details of resource use assumptions and unit costs, see *Resource use and cost data*). The majority of unit costs have been taken from *NHS reference costs 2007/08*.[43] However, costs for BAHA sound processors and surgical consumables were supplied by NHS providers, as these data are not routinely reported. The BAHA costs reported in *Table 35* are based on list prices and do not take account of discounts that may be available to individual hospital trusts.

Cost analysis

Base case: cost analysis

Table 36 reports the modelled cost per case for providing a BAHA or BCHA to a cohort of children and adults, using a time horizon of 10 years. In both cases, BAHA is the more costly strategy – increasing costs in children by approximately 94% over BCHA, and increasing costs in adults by 93%. The differences between the modelled costs for paediatric and adult cases principally arise in assumptions regarding the use of two-stage surgery for paediatric cases and also higher unit costs for paediatric outpatient assessments. The same assumptions regarding treatment failure, loss of bone integration and skin reactions were applied to both cohorts. There are slight differences between the paediatric and adult cohorts in terms of the general mortality probabilities applied (based on age-specific death rates for the general population in the UK).

TABLE 33 Summary of input parameters for model: probabilities

Input parameter	Base-case value	95% CI		Source
		Lower limit	Upper limit	
Proportion of cohort that is male				
Children (aged < 16 years)	0.5121	NA	NA	ONS[127]
Adults (aged > 50 years)	0.4639	NA	NA	ONS[127]
Probability of adverse outcome from initial surgery	**0.0970**			**See (a) and (b) below**
(a) Probability of bleeding within 24 hours of surgery	0.0121	0.0015	0.0431	Badran *et al.*[114]
(b) Probability of surgical removal of skin growth or soft tissue thickening around the abutment	0.0848	0.0472	0.1383	Badran *et al.*[114]
Probability of failure of initial surgery	**0.0332**			**See (c) and (d) below**
(c) Probability of intolerable pain requiring removal of abutment and flange fixture	0.0272	0.0075	0.0682	Badran *et al.*[114]
(d) Probability of failure to integrate	0.0060	0.0002	0.0329	Håkansson *et al.*[101]
Probability of ceasing treatment in patient with adverse outcome from initial surgery	0.0000	NA	NA	Assumption
Probability of resolution of adverse outcome of surgery	1.0000	NA	NA	Assumption
Probability of skin reaction (all grades)	**0.1819**			**See (e), (f) and (g) below**
(e) Probability of grade 1 or 2 skin reaction	0.1451	0.1124	0.1842	Håkansson *et al.*[101]
(f) Probability of grade 3 skin reaction	0.0346	0.0198	0.0563	Håkansson *et al.*[101]
(g) Probability of grade 4 skin reaction	0.0022	0.0001	0.0121	Håkansson *et al.*[101]
Probability of losing bone integration	0.0428	0.0188	0.1028	Bonding[100]
Probability of re-operation	0.9474	0.7397	0.9987	Proops[54]
Probability of death from all causes	Age-specific	NA	NA	ONS[128]

NA, not applicable; ONS, Office for National Statistics.
Sources given are for base-case estimates. Exact binomial 95% CIs estimated from reported data.

TABLE 34 Summary of input parameters for model: estimated utility gain from aided hearing and usage of devices

Input parameter	Base-case value	95% CI		Source
		Lower limit	Upper limit	
Utility associated with hearing levels (HUI3)				
Unable to hear at all (level 6)	0.465	NA	NA	Furlong *et al.*[120,121]
Able to hear conversation with one person but not group (level 5)	0.644			
Able to hear conversation with one person and with group (level 3)	0.849			
Proportion of cohort using devices				
BCHA	0.90	0.683	0.988	Hol *et al.*[82]
BAHA	1.00	0.832	1.000	

NA, not applicable.
Sources given are for base-case estimates. Exact binomial 95% CIs estimated from reported data.

Table 37 reports a breakdown of the modelled cost per case for BAHA provision, using the phases of the management pathway identified in *Resource use and cost data*. The most costly phase identified in *Table 37* is long-term maintenance, constituting 36% and 42% of total costs in paediatric and adult cases, respectively. The high cost of the long-term maintenance is primarily

TABLE 35 Summary of input parameters for model: unit costs

Input parameter		Base-case value (£)	Source
Costs of BAHA provision			
Cost of assessments prior to initial surgery			
Adult		315.63	*NHS reference costs 2007/08*[43]
Paediatric		336.53	
Cost of initial surgery			
One-stage		2994.07	*NHS reference costs 2007/08*[43]
Two-stage		4998.64	
Post-surgical costs (in year following surgery)			
Adult		381.47	*NHS reference costs 2007/08*[43]
Paediatric		437.91	
Cost of BAHA processor		2191.25	UHB; SUHT
Long-term costs of BAHA		786.42[a]	
Adverse outcomes			
Bleeding within 24 hours of operation		332.35[b]	*NHS reference costs 2007/08*[43]
Surgical removal of tissue round abutment		663.66[c]	
Removal of abutment due to intolerable pain		2004.57	
Adverse events			
Grade 1 or 2 skin irritation (adult)		123.50	*NHS reference costs 2007/08*,[43] BNF[125]
Grade 1 or 2 skin irritation (paediatric)		140.22	
Grade 3 skin irritation		663.66[c]	*NHS reference costs 2007/08*[43]
Grade 4 skin irritation		2004.57[d]	
Repeat operation owing to loss of bone integration (adult)		4111.80	See *Table 31*
Repeat operation owing to loss of bone integration (paediatric)		6172.80	
Costs of BCHA provision			
Cost of assessments prior to fitting of BCHA		57.48	*NHS reference costs 2007/08*[43]
Cost of fitting BCHA		64.80	
Cost of audiological assessment post-BCHA fitting		50.17	
Cost of BCHA device	Low (body-worn)	117–183	SUHT
	Low (behind-ear worn)	250	
	High	300–350	UHB
Long-term costs of BCHA[e]		75–123	

a Includes annual maintenance cost.
b Assume an overnight stay – excess bed day for BAHAs (Healthcare Resource Group Code CZ26Z).
c Intermediate skin procedure without complications and comorbidities (HRG Code JC04C) as day case.
d Assume same cost as initial surgery.
e Includes an annual audiological assessment and one-fifth of cost of BCHA (on assumption that the device is replaced every 5 years) without maintenance/replacement contract. Range from least to most expensive BCHA device.
All unit costs are expressed in 2009 pound sterling.

the result of costs associated with maintenance plans for the BAHA sound processors and the periodic replacement of the processors.

In the base-case analyses reported in *Tables 36* and *37* it is assumed that individuals modelled as treatment failures with a BAHA, either because of intolerable pain or because they choose not

TABLE 36 Base-case analysis: cost per case

Strategy	Paediatric (£)	Adult (£)
BC hearing aid	1105	1084
BAHA	17,514	14,533
Incremental cost per case	16,409	13,449

TABLE 37 Base-case analysis: breakdown of costs of BAHA provision by phase of management

Cost breakdown	Paediatric (£)	Adult (£)
Initial assessment	337	316
Surgical costs	4999	2994
Post-surgery costs, including fitting of sound processor	3253	3189
Long-term maintenance costs	6241	6114
Adverse event costs	2684	1921

to have a re-operation following loss of bone integration, are not provided with an alternative hearing aid. These constitute approximately 6% of the original cohort and a proportion might be expected to revert to their previous hearing aid on failure with a BAHA. *Table 38* presents an alternative base-case analysis with all individuals who experience treatment failure with a BAHA switching to BCHA in the year following treatment failure. The effect of this is to marginally increase the cost per case for the BAHA cohort, by 0.8% for children and 0.9% for adults.

Table 39 reports a breakdown of the modelled cost per case for BAHA provision, similar to that in *Table 37*, but including the cost of people switching to BCHA on failure with a BAHA. As this change in management occurs as a result of experiencing adverse events, the cost of providing BCHA and continued audiological management is classified under adverse event costs in *Table 39*.

Average cost per case successfully treated with a BAHA

Given that a proportion of participants (up to 6%) are assumed, in the base case, to choose not to continue with their BAHAs, the results in *Table 36* could be presented slightly differently – as cost per case successfully treated with a BAHA. To derive this figure we calculated the proportion of the cohort still using a BAHA at the end of the modelled time horizon by subtracting those who had died (0.1% for children and 1.9% for adults) and those who had chosen not to continue with the BAHA owing to adverse events (6.1% for children and 6% for adults). This equates to 93.8% successfully treated children and 92.1% successfully treated adults. The average cost per successfully treated patient was derived by dividing the average costs in *Table 36* (average cost per patient, assuming treatment failures do not receive an alternative device) by the estimated proportion of cases successfully treated. Under these assumptions the cost per patient successfully treated with a BAHA is £18,681 for paediatric cases and £15,785 for adults.

Cost analysis: deterministic sensitivity analysis

We conducted a series of univariate sensitivity analyses, varying one parameter at a time from its base-case value while leaving all other variables unchanged. Probability parameters were varied between their 95% confidence limits, calculated as exact binomial CIs (see *Table 33*). In the absence of appropriate measures of variability in NHS reference costs,[43] cost parameters were varied by plus or minus 25%, except for the costs of the BAHA sound processor and associated maintenance plans, which were varied between the lowest and highest model costs (as supplied by UHB and SUHT; see *Table 28*).

TABLE 38 Base-case analysis: cost per case. BAHA treatment failures revert to BCHA

Strategy	Paediatric (£)	Adult (£)
BC hearing aid	1105	1084
BAHA	17,649	14,666
Incremental cost per case	16,545	13,582

TABLE 39 Base-case analysis: breakdown of costs of BAHA provision by phase of management. BAHA treatment failures revert to BCHA

Cost breakdown	Paediatric (£)	Adult (£)
Initial assessment	337	316
Surgical costs	4999	2994
Post-surgery costs, including fitting of sound processor	3253	3189
Long-term maintenance costs	6241	6114
Adverse event costs	2820	2054

Table 40 reports the results of a DSA for paediatric cases, assuming that BAHA treatment failures do not switch to an alternative hearing aid. The value of the input parameter for each analysis is shown in the second column of the table. The table contains two rows for each input parameter – the first of which reports the results at the lower limit (either lower 95% confidence limit or the lower limit of the assumed range) and the second of which reports the results at the upper limit (either upper 95% confidence limit or the upper limit of the assumed range). The table also shows the base-case value for each input parameter, following the description of the parameter in the first column of the table. *Table 41* reports similar analyses, assuming that participants switch to using BCHA on treatment failure.

The DSA suggests that the cost results are generally robust to variation in the value of input parameters. The results are most sensitive to variation in the probability of re-operation when implants lose bone integration, the cost of surgical implantation, the cost of the BAHA processor maintenance plan and, to a lesser extent, the initial cost of the BAHA processor and the probability of intolerable pain requiring removal of the BAHA fixture.

Table 42 reports the results of a DSA for adult cases, assuming that BAHA treatment failures do not switch to an alternative hearing aid. *Table 43* reports similar analyses assuming that participants switch to using BCHA on treatment failure. As with the previous analysis for paediatric cases, the DSA suggests that the results are generally robust to variation in the value of input parameters, with costs of BAHA provision being most sensitive to variation in the probability of re-operation (for loss of bone integration), the cost of surgical implantation and the cost of the BAHA processor maintenance plan and, to a lesser extent, the initial cost of the BAHA processor and the probability of intolerable pain requiring removal of the BAHA fixture.

Exploratory cost-effectiveness analysis

An exploratory, augmented base-case analysis was developed to incorporate potential benefits from improved hearing resulting from BAHA provision and improvements in wearability, compared with BCHA. In this analysis we assumed that the QoL benefit from improved hearing could be proxied by levels of the hearing attribute in the HUI3 and that identical gains were potentially achievable through use of BAHA and BCHA. Differences between the utility associated with use of BAHA or BCHA were assumed to be realised through differences in

TABLE 40 Deterministic sensitivity analysis: paediatric cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1105	17,513	16,408
	0.04310	1105	17,518	16,413
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1105	17,509	16,404
	0.13826	1105	17,525	16,420
Probability of failure to integrate (0.006)	0.00015	1105	17,513	16,409
	0.03291	1105	17,501	16,397
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1105	17,840	16,735
	0.06820	1105	17,445	16,341
Probability of re-operation (0.9474)	0.73972	1105	16,668	15,563
	0.99867	1105	17,732	16,627
Cost of initial ENT consultation (£131.69)	98.77	1105	17,481	16,376
	164.61	1105	17,547	16,442
Cost of audiological assessment (£57.48)	43.11	1105	17,499	16,395
	71.85	1105	17,528	16,424
Cost of ENT multiprofessional assessment (£147.36)	110.52	1105	17,477	16,372
	184.21	1105	17,551	16,446
Cost of day case surgery for implantation (£4009.14)	3006.86	1105	16,159	15,055
	5011.43	1105	18,868	17,764
Cost of fixture and abutment (£830.00)	622.50	1105	17,338	16,234
	1037.50	1105	17,689	16,585
Cost of surgical consumables (£159.50)	119.63	1105	17,460	16,355
	199.38	1105	17,568	16,463
Cost of follow-up ENT consultations (£90.93)	68.20	1105	17,424	16,319
	113.66	1105	17,604	16,499
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1105	17,493	16,388
	81.00	1105	17,535	16,431
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1105	17,481	16,376
	62.71	1105	17,547	16,442
Cost of BAHA sound processor (£2191.25)	1820.00	1105	17,147	16,042
	2995.00	1105	18,308	17,204
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1105	16,346	15,242
	1000.00	1105	19,953	18,849
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1105	17,459	16,354
	829.58	1105	17,569	16,464

For each input parameter, the first row reports the results at the lower limit (either the lower 95% confidence limit or the lower limit of the assumed range) and the second reports the results at the upper limit (either the upper 95% confidence limit or the upper limit of the assumed range).

the proportion of time that members of the modelled cohorts used each device, as outlined in *Quality of life*.

The results are reported in terms of total costs and total QALYs for each treatment strategy, incremental costs and benefits and the ICERs. Costs and outcomes are discounted at 3.5%.

The results of the augmented base-case analysis in paediatric cases are reported in *Table 44*. The average costs estimated for each cohort are identical to those reported in *Table 38* (average costs,

TABLE 41 Deterministic sensitivity analysis: paediatric cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1105	17,648	16,544
	0.04310	1105	17,654	16,549
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1105	17,644	16,540
	0.13826	1105	17,661	16,556
Probability of failure to integrate (0.006)	0.00015	1105	17,649	16,545
	0.03291	1105	17,653	16,549
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1105	17,881	16,777
	0.06820	1105	17,601	16,496
Probability of re-operation (0.9474)	0.73972	1105	16,950	15,846
	0.99867	1105	17,830	16,725
Cost of initial ENT consultation (£131.69)	98.77	1105	17,616	16,512
	164.61	1105	17,682	16,578
Cost of audiological assessment (£57.48)	43.11	1105	17,635	16,530
	71.85	1105	17,664	16,559
Cost of ENT multiprofessional assessment (£147.36)	110.52	1105	17,612	16,508
	184.21	1105	17,686	16,582
Cost of day case surgery for implantation (£4009.14)	3006.86	1105	16,295	15,190
	5011.43	1105	19,004	17,899
Cost of fixture and abutment (£830.00)	622.50	1105	17,474	16,369
	1037.50	1105	17,825	16,720
Cost of surgical consumables (£159.50)	119.63	1105	17,595	16,491
	199.38	1105	17,703	16,599
Cost of follow-up ENT consultations (£90.93)	68.20	1105	17,559	16,455
	113.66	1105	17,740	16,635
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1105	17,628	16,524
	81.00	1105	17,671	16,566
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1105	17,616	16,512
	62.71	1105	17,682	16,578
Cost of BAHA sound processor (£2191.25)	1820.00	1105	17,282	16,178
	2995.00	1105	18,444	17,339
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1105	16,482	15,377
	1000.00	1105	20,089	18,984
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1105	17,595	16,490
	829.58	1105	17,704	16,600

For each input parameter, the first row reports the results at the lower limit (either the lower 95% confidence limit or the lower limit of the assumed range) and the second reports the results at the upper limit (either the upper 95% confidence limit or the upper limit of the assumed range).

assuming BAHA treatment failures revert to BCHA). The QALY outcomes for BCHA and BAHA have been estimated on two different potential levels of hearing gain associated with the use of hearing aids (based on items in the HUI3 hearing domain). Under the first assumption (QALY1) the utility gain from aided hearing is 0.178, based on attaining level 5 on the HUI hearing domain ('able, when using hearing aid, to hear conversation with one other person, but unable to hear what is said in a group conversation'; see *Table 21* and *Quality of life* for more details), while under the second assumption (QALY2) the utility gain from aided hearing is 0.384, based on attaining level 3 on the HUI3 hearing domain ('able to hear both conversation with one other person and

TABLE 42 Deterministic sensitivity analysis: adult cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1084	14,532	13,448
	0.04310	1084	14,538	13,453
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1084	14,529	13,444
	0.13826	1084	14,545	13,460
Probability of failure to integrate (0.006)	0.00015	1084	14,533	13,449
	0.03291	1084	14,531	13,447
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1084	14,828	13,744
	0.06820	1084	14,472	13,387
Probability of re-operation (0.9474)	0.73972	1084	13,877	12,792
	0.99867	1084	14,702	13,618
Cost of initial ENT consultation (£110.78)	83.09	1084	14,506	13,422
	138.48	1084	14,561	13,477
Cost of audiological assessment (£57.48)	43.11	1084	14,519	13,435
	71.85	1084	14,548	13,464
Cost of ENT multiprofessional assessment (£147.36)	110.52	1084	14,497	13,412
	184.21	1084	14,570	13,486
Cost of day case surgery for implantation (£2004.57)	1503.43	1084	13,860	12,776
	2505.71	1084	15,207	14,123
Cost of fixture and abutment (£830.00)	622.50	1084	14,359	13,275
	1037.50	1084	14,708	13,624
Cost of surgical consumables (£159.50)	119.63	1084	14,480	13,396
	199.38	1084	14,587	13,503
Cost of follow-up ENT consultations (£72.11)	54.09	1084	14,462	13,378
	90.14	1084	14,604	13,520
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1084	14,512	13,428
	81.00	1084	14,555	13,470
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1084	14,501	13,417
	62.71	1084	14,566	13,482
Cost of BAHA sound processor (£2191.25)	1820.00	1084	14,168	13,084
	2995.00	1084	15,325	14,241
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1084	13,388	12,303
	1000.00	1084	16,927	15,843
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1084	14,480	13,396
	829.58	1084	14,587	13,503

For each input parameter, the first row reports the results at the lower limit (either the lower 95% confidence limit or the lower limit of the assumed range) and the second reports the results at the upper limit (either the upper 95% confidence limit or the upper limit of the assumed range).

what is said in a group conversation, when using hearing aid'; see *Table 21* and *Quality of life* for more details). These assumptions apply equally to BCHA and BAHA. However, different assumptions apply to the proportion of the cohort using the relevant hearing aid (see *Table 34*), which give rise to different QALY estimates for the two modelled cohorts (BAHA vs BCHA).

For paediatric cases, the QALY gain associated with providing BAHA ranges from 0.14 to 0.30 (depending on the assumed level of utility associated with aided hearing), resulting in ICERs of £119,367 and £55,642, respectively.

TABLE 43 Deterministic sensitivity analysis: adult cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1084	14,665	13,580
	0.04310	1084	14,670	13,586
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1084	14,661	13,577
	0.13826	1084	14,677	13,593
Probability of failure to integrate (0.006)	0.00015	1084	14,666	13,582
	0.03291	1084	14,679	13,595
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1084	14,869	13,784
	0.06820	1084	14,623	13,539
Probability of re-operation (0.9474)	0.73972	1084	14,152	13,068
	0.99867	1084	14,798	13,714
Cost of initial ENT consultation (£110.78)	83.09	1084	14,638	13,554
	138.48	1084	14,694	13,609
Cost of audiological assessment (£57.48)	43.11	1084	14,651	13,567
	71.85	1084	14,680	13,596
Cost of ENT multiprofessional assessment (£147.36)	110.52	1084	14,629	13,545
	184.21	1084	14,703	13,618
Cost of day case surgery for implantation (£2004.57)	1503.43	1084	13,992	12,908
	2505.71	1084	15,339	14,255
Cost of fixture and abutment (£830.00)	622.50	1084	14,491	13,407
	1037.50	1084	14,841	13,756
Cost of surgical consumables (£159.50)	119.63	1084	14,612	13,528
	199.38	1084	14,719	13,635
Cost of follow-up ENT consultations (£72.11)	54.09	1084	14,595	13,511
	90.14	1084	14,737	13,653
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1084	14,645	13,561
	81.00	1084	14,687	13,603
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1084	14,633	13,549
	62.71	1084	14,699	13,614
Cost of BAHA sound processor (£2191.25)	1820.00	1084	14,300	13,216
	2995.00	1084	15,458	14,373
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1084	13,520	12,436
	1000.00	1084	17,060	15,976
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1084	14,612	13,528
	829.58	1084	14,719	13,635

For each input parameter, the first row reports the results at the lower limit (either the lower 95% confidence limit or the lower limit of the assumed range) and the second reports the results at the upper limit (either the upper 95% confidence limit or the upper limit of the assumed range).

The results of the augmented base-case analysis in adults, applying the same assumptions on potential utility gain from aided hearing and usage of hearing aids, are reported in *Table 45*. The estimated QALY gain from use of BAHA is similar to that for children, ranging from 0.14 to 0.29 depending on the assumed level of utility associated with aided hearing. However, the ICERs are lower (£100,029 and £46,628, respectively) given the lower incremental costs estimated for adults (resulting from the use of one-stage surgery in adults and lower costs for adult outpatient attendances).

TABLE 44 Augmented base-case analysis: including estimated QALY outcomes (paediatric cases)

	Cost (£)	QALY1[a]	QALY2[b]
BCHA	1105	5.20	6.74
BAHA	17,649	5.34	7.04
Difference	16,545	0.14	0.30
ICER (£)		119,367	55,642

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

TABLE 45 Augmented base-case analysis: including QALY outcomes (adult cases)

	Cost (£)	QALY1[a]	QALY2[b]
BCHA	1084	5.10	6.60
BAHA	14,666	5.23	6.89
Difference	13,582	0.14	0.29
ICER (£)		100,029	46,628

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

Deterministic sensitivity analysis

Table 46 reports the results of a series of univariate sensitivity analyses, indicating the effect of variation in input parameters on the ICER. Results for each parameter are reported on two lines – the first gives the results at the lower limit of the input parameter and the second gives the result at the upper limit. Probabilities are varied between the lower and upper limits of the 95% CI, while unit costs derived from NHS reference costs[43] are varied by plus or minus 25% of their average values. Costs of the BAHA processor and annual maintenance contract are varied from the lowest to highest reported values. As for the base-case analysis, ICERs are reported for two outcome scenarios (QALY1 and QALY2) in which the utility gain from aided hearing is estimated based on levels of the hearing domain of the HUI3. In the final two analyses reported in *Table 46*, two scenarios are considered. Firstly, the proportion of the BCHA cohort using their aid for ≥ 8 hours is varied between the lower and upper limits of the 95% CI for the value adopted in the base case [18/20 (90%)], while keeping the proportion of the BCHA cohort using their aid for ≥ 8 hours at 100%. Secondly, in a bivariate sensitivity analysis in which the proportion of people using their BCHA and the proportion of people using their BAHA are varied simultaneously, the ICERs are estimated at the lower limit of the 95% CI for hearing aid usage in the BCHA and BAHA cohorts (0.683 and 0.832, respectively). No analysis is reported for the upper limit of the CI, as that would simply repeat the results presented in the previous row. The majority of the input variables included in the DSA have minimal impact on the QALY outcomes for both BCHA and BAHA (full results are reported in *Appendix 12*).

The DSA suggests that the results are generally robust to variation in input probabilities and unit costs. In terms of input probabilities, the greatest variation in ICER relates to initial failure of bone integration, failure of BAHA implantation owing to intolerable pain and the probability of re-operation because of loss of bone integration. In contrast to other input probabilities, the ICER reduces as the probability of re-operation increases. This occurs because, although costs

TABLE 46 Deterministic sensitivity analysis (DSA): impact on ICER

Input parameter (base-case value)	Input value	ICER (£ per QALY gained)			
		Paediatric		Adult	
		QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	119,358	55,638	100,020	46,624
	0.04310	119,397	55,656	100,060	46,642
Probability of surgical reduction of skin growth/thickening around abutment (0.0848)	0.04716	119,331	55,626	99,993	46,611
	0.13826	119,448	55,680	100,111	46,667
Probability of failure to integrate (0.006)	0.00015	119,461	55,686	100,110	46,666
	0.03291	124,361	57,970	104,302	48,620
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	117,042	54,559	98,172	45,763
	0.06820	119,872	55,878	100,433	46,816
Probability of re-operation (0.9474)	0.73972	119,907	55,894	100,909	47,038
	0.99867	119,248	55,587	99,825	46,533
Cost of initial ENT consultation [£131.69 (paediatric) and £110.78 (adult)]	98.77 (P), 83.09 (A)	119,129	55,532	99,825	46,533
	164.61 (P), 138.48 (A)	119,604	55,753	100,233	46,723
Cost of audiological assessment (£57.48)	43.11	119,263	55,594	99,923	46,579
	71.85	119,471	55,691	100,135	46,678
Cost of ENT multiprofessional assessment (£147.36)	110.52	119,101	55,518	99,758	46,502
	184.21	119,633	55,766	100,301	46,755
Cost of day case surgery for implantation (£2004.57)	1503.43	109,596	51,088	95,068	44,316
	2505.71	129,138	60,197	104,990	48,941
Cost of fixture and abutment (£830.00)	622.50	118,099	55,052	98,742	46,028
	1037.50	120,634	56,233	101,316	47,228
Cost of surgical consumables (£159.50)	119.63	118,978	55,461	99,634	46,444
	199.38	119,756	55,824	100,424	46,812
Cost of follow-up ENT consultations [£90.93 (paediatric) and £72.11 (adult)]	68.20 (P), 54.09 (A)	118,716	55,339	99,506	46,385
	113.66 (P), 90.14 (A)	120,018	55,946	100,552	46,872
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	119,213	55,571	99,874	46,556
	81.00	119,520	55,714	100,185	46,701
Cost of audiology follow-up in year of surgery (£50.17)	37.62	119,129	55,532	99,789	46,516
	62.71	119,604	55,753	100,270	46,740
Cost of BAHA sound processor (£2191.25)	1820.00	116,719	54,408	97,336	45,373
	2995.00	125,099	58,314	105,861	49,347
Cost of BAHA sound processor maintenance plan (£735.25)	610.00	110,943	51,715	91,589	42,694
	1000.00	136,966	63,846	117,661	54,847
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	118,971	55,458	99,634	46,444
	829.58	119,762	55,827	100,424	46,812
Proportion of cohort using BCHA for >8 hours per day (0.90)	0.683	37,025	17,259	31,027	14,463
	0.988	1,216,561	567,095	1,019,479	475,226
Proportion using BCHA at lower limit of 95% CI, 0.683, and proportion using BAHA at lower limit of 95% CI, 0.832 (BCHA = 0.90; BAHA = 1.00)	0.683/0.832	82,287	38,358	68,948	32,140

A, adult; P, paediatric.

a Moving from level 6 of the HUI hearing attribute to level 5.

b Moving from level 6 of the HUI hearing attribute to level 3.

For each input parameter, the first row reports the results at the lower limit (either the lower 95% confidence limit or the lower limit of the assumed range) and the second reports the results at the upper limit (either the upper 95% confidence limit or the upper limit of the assumed range).

See *Quality of life* for full details.

increase (associated with additional surgical procedures), the QALY gained from BAHA also increases (with the proportionate QALY gain being greater than the proportionate increase in cost). With respect to cost inputs, the greatest variation in ICER relates to the cost of day surgery for implantation and the cost of components of the BAHA system (fixture and abutment, BAHA sound processor and the cost of the maintenance plan), with the cost of the maintenance plan having the greatest impact. The variable that has the greatest influence on the cost-effectiveness results is the proportion of each cohort using their hearing aids. Very high ICER values are associated with high usage of BCHA (98.8% at the upper limit of the 95% CI, resulting in a small difference in usage between BCHA and BAHA), but more acceptable values are associated with lower BCHA usage (than for BAHA). Threshold values for differences in use of hearing aids and the underlying utility gain from aided hearing are explored further in a range of scenario analyses in the following section of this report.

Scenario analysis

Scenario 1: alternative cost assumptions for BCHA

Tables 47 and *48* report a scenario analysis in which the base-case analysis is re-run for alternative assumptions regarding the cost of BCHA. Four possible costs, two related to body-worn hearing aids (ranging from £117 to £183) and two related to behind-the-ear aids (from £250 for a

TABLE 47 Scenario analysis on unit cost for BCHA (paediatric cases)

Unit cost of BCHA (£)	Strategy	Cost (£)	QALY1[a]	ICER1 (£ per QALY gained)	QALY2[b]	ICER2 (£ per QALY gained)
117	BCHA	765	5.20		6.74	
	BAHA	17,587	5.34	121,362	7.04	56,573
183	BCHA	934	5.20		6.74	
	BAHA	17,618	5.34	120,372	7.04	56,111
250	BCHA	1105	5.20		6.74	
	BAHA	17,649	5.34	119,367	7.04	55,642
350	BCHA	1360	5.20		6.74	
	BAHA	17,696	5.34	117,866	7.04	54,943

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

TABLE 48 Scenario analysis on unit cost for BCHA (adult cases)

Unit cost of BCHA (£)	Strategy	Cost (£)	QALY1[a]	ICER1 (£ per QALY gained)	QALY2[b]	ICER2 (£ per QALY gained)
117	BCHA	765	5.20		6.74	
	BAHA	17,587	5.34	121,362	7.04	56,573
183	BCHA	917	5.10		6.60	
	BAHA	14,635	5.23	101,038	6.89	47,098
250	BCHA	1084	5.10		6.60	
	BAHA	14,666	5.23	100,029	6.89	46,628
350	BCHA	1334	5.10		6.60	
	BAHA	14,712	5.23	98,524	6.89	45,927

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

device incorporating an NHS digital aid to £350 for a proprietary device), were identified by NHS providers for conventional BCHA (see *Table 32*). In the base case, the mid-point of £250 was adopted. The scenario analysis examines the robustness of the model results to alternative assumptions regarding the cost of the comparator device. The values reported in *Tables 47* and *48* suggest that the ICERs are robust to alternative assumptions regarding the cost of BCHA. The costs of providing BCHA vary by approximately £600 between the highest and lowest cost options, over the modelled 10-year time horizon. The difference between BCHA and BAHA ranges from £16,337 to £16,821 for paediatric cases and from £13,337 to £13,853 for adults.

Scenario 2: alternative assumptions regarding the proportion of the cohort using a BCHA for more than 8 hours per day

Tables 49 and *50* report scenario analyses in which the proportion of the cohort using a BCHA for more than 8 hours per day is varied from 0% to 100%, while holding the proportion using a BAHA at 100%. As indicated in the DSA, reported above, the QALY gain from aided hearing and hence the ICERs are highly sensitive to assumptions regarding the usage of hearing aids. The ICERs for QALY1 (able to hear one-to-one conversation) are low and below conventional thresholds for acceptable cost-effectiveness for low-to-medium proportions using a BCHA – up to approximately 60% in both paediatric cases and adults. When the assumed utility for aided hearing is greater (i.e. QALY2, in which members of the modelled cohorts can hear group conversation as well as one-to-one conversation), even relatively high proportions using a BCHA

TABLE 49 Scenario analysis on proportion of cohort using BCHA for more than 8 hours per day (paediatric cases)

Proportion using BCHA	Device	Cost (£)	QALY1[a]	ICER1[a] (£ per QALY gained)	QALY2[b]	ICER2[b] (£ per QALY gained)
0.0	BCHA	1105	3.87		3.87	
	BAHA	17,649	5.28	11,675	6.91	5442
0.1	BCHA	1105	4.01		4.18	
	BAHA	17,649	5.29	12,976	6.92	6049
0.2	BCHA	1105	4.16		4.50	
	BAHA	17,649	5.30	14,603	6.93	6807
0.3	BCHA	1105	4.31		4.82	
	BAHA	17,649	5.30	16,697	6.95	7783
0.4	BCHA	1105	4.46		5.14	
	BAHA	17,649	5.31	19,491	6.96	9085
0.5	BCHA	1105	4.61		5.46	
	BAHA	17,649	5.32	23,408	6.98	10,911
0.6	BCHA	1105	4.76		5.78	
	BAHA	17,649	5.32	29,295	6.99	13,656
0.7	BCHA	1105	4.91		6.10	
	BAHA	17,649	5.33	39,140	7.01	18,245
0.8	BCHA	1105	5.06		6.42	
	BAHA	17,649	5.34	58,951	7.02	27,480
0.9	BCHA	1105	5.20		6.74	
	BAHA	17,649	5.34	119,367	7.04	55,642
1.0	BCHA	1105	5.35		7.06	
	BAHA	17,649	5.35	Dominated	7.05	Dominated

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

(80% usage or an assumed difference of 20% compared with BAHA usage) yield ICERs below conventionally adopted thresholds.

Threshold values (of the proportion using a BCHA) for cost-effectiveness, at a willingness to pay (WTP) of £30,000 per QALY gained, were 61% and 67% for QALY1 and 82% and 85% for QALY2 in paediatric and adult cases, respectively. This means that, when the difference in usage between BCHA and BAHA is greater than 39% in paediatric cases and greater than 33% in adults and the utility gain from aided hearing (with BAHA or BCHA) is 0.178, the BAHA may be a cost-effective option. If functional gain (and hence the utility gain) is greater, then the minimum difference is lower (18% in paediatric cases and 15% in adults). Similar threshold values can be calculated for a WTP threshold of £20,000 per QALY gained. These are 42% and 51% for QALY1 and 73% and 77% for QALY2 in paediatric and adult cases, respectively. BAHA usage is assumed to be 100%, as reported by Hol and colleagues,[82] in these scenarios.

Scenario 3: include pain associated with BCHA use

Tables 51 and *52* report scenario analyses in which the base-case analysis is re-run, but including an assumption that use of a BCHA is associated with some discomfort (proxied by level 2 on the HUI3 pain dimension; see *Quality of life* for details). This has the effect of reducing total QALYs for BCHAs from 5.2 to 4.9 (for QALY1) and from 6.74 to 6.37 (for QALY2). Given that the outcome model for BAHAs incorporates some patients having their implants removed owing

TABLE 50 Scenario analysis on proportion of cohort using BCHA for more than 8 hours per day (adult cases)

Proportion using BCHA	Device	Cost (£)	QALY1[a]	ICER1[a] (£ per QALY gained)	QALY2[b]	ICER2[b] (£ per QALY gained)
0.0	BCHA	1084	3.79		3.79	
	BAHA	14,666	5.17	9784	6.76	4561
0.1	BCHA	1084	3.93		4.10	
	BAHA	14,666	5.18	10,874	6.78	5069
0.2	BCHA	1084	4.08		4.41	
	BAHA	14,666	5.19	12,237	6.79	5704
0.3	BCHA	1084	4.22		4.72	
	BAHA	14,666	5.19	13,992	6.81	6522
0.4	BCHA	1084	4.37		5.04	
	BAHA	14,666	5.20	16,333	6.82	7614
0.5	BCHA	1084	4.51		5.35	
	BAHA	14,666	5.21	19,616	6.83	9144
0.6	BCHA	1084	4.66		5.66	
	BAHA	14,666	5.21	24,549	6.85	11,444
0.7	BCHA	1084	4.81		5.97	
	BAHA	14,666	5.22	32,799	6.86	15,289
0.8	BCHA	1084	4.95		6.29	
	BAHA	14,666	5.23	49,400	6.88	23,028
0.9	BCHA	1084	5.10		6.60	
	BAHA	14,666	5.23	100,029	6.89	46,628
1.0	BCHA	1084	5.24		6.91	
	BAHA	14,666	5.24	Dominated	6.90	Dominated

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

TABLE 51 Scenario analysis – include pain associated with use of BCHA (paediatric cases)

	Cost (£)	QALY1[a]	QALY2[b]
BCHA	1105	4.90	6.37
BAHA	17,649	5.33	7.02
Difference	16,545	0.43	0.65
ICER		38,348	25,559

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

TABLE 52 Scenario analysis – include pain associated with use of BCHA (adult cases)

	Cost (£)	QALY1[a]	QALY2[b]
BCHA	1084	4.80	6.24
BAHA	14,666	5.22	6.87
Difference	13,582	0.42	0.63
ICER		32,136	21,419

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

to intolerable pain, we included a utility loss (proxied by level 5 of the HUI3 pain dimension; see *Quality of life* for details) for these participants, in the cycle in which the removal was modelled to occur. Including pain associated with the BAHA implant results in a reduction in the total QALYs for BAHAs from 5.34 to 5.33 (QALY1) and from 7.04 to 7.02 (QALY2). This has the effect of increasing the QALY difference between BAHAs and BCHAs and leads to substantially lower ICER values.

Scenario 4: threshold analysis on utility difference

A final series of threshold analyses were conducted to estimate the utility difference (the utility gain from aided hearing) required to meet conventional cost-effectiveness thresholds, if the difference in use between BCHAs and BAHAs is 10% (as reported by Hol and colleagues[82]). For a WTP threshold of £30,000 per QALY, the required utility difference in adults is 0.597. Using the anchor points adopted in our base-case analysis (see *Table 34*) of 0.644 for being able to hear one-to-one conversation and 0.849 for being able to hear one-to-one and group conversation, a utility difference of 0.597 implies a utility value of 0.047 for deafness (approximately equal to that for death) at the lower anchor point and of 0.252 for the higher anchor point. Repeating this analysis for paediatric cases gives a larger required utility difference of 0.712.

Table 53 reports similar analyses undertaken at varying values for the difference in the proportion using their BCHA (compared with BAHA). As with the earlier DSA, this analysis suggests that the required utility difference (to achieve acceptable cost-effectiveness) reduces as the proportion using BCHA reduces (compared with the proportion using BAHA).

Probabilistic sensitivity analysis

In a PSA in which the difference in proportion of participants using their hearing aid (BAHA–BCHA), hearing aid costs, the probability of adverse events as well as surgical and long-term maintenance costs were sampled probabilistically, mean costs and QALY outcomes were similar

TABLE 53 Utility difference required for cost-effectiveness (at two cost-effectiveness thresholds) for varying differences in the proportion of the cohort using BCHA for ≥ 8 hours per day

Difference in proportion of cohort using BCHA (compared with BAHA)	Threshold WTP (£ per QALY gained)	
	£20,000	£30,000
0.10	NA	0.597
0.15	0.592	0.395
0.20	0.442	0.295
0.25	0.353	0.235
0.30	0.294	0.196
0.35	0.251	0.168
0.40	0.220	0.146
0.45	0.195	0.130
0.50	0.176	0.117

NA, not applicable.
This analysis was conducted for adult patients at the higher anchor point of QoL gain associated with improved hearing (0.849).

to those reported for the deterministic analysis [see *Table 44* (paediatric cases) and *Table 45* (adults)]. Providing BAHAs for children is associated with increased QALYs [with a range from 0.01 to 0.36 for the lower utility gain (QALY1) and from 0.03 to 0.79 for the higher utility gain (QALY2)], but also increased costs (ranging from £13,804 to £21,496) in all simulations, when compared with BCHAs (*Table 54*).

In this analysis, providing BAHAs in place of BCHAs in paediatric cases had a probability of being cost-effective of 0% at a WTP threshold of £20,000 per QALY gained and of 0.1% at a WTP threshold of £30,000 per QALY gained, if the utility gain from aided hearing was assumed to be 0.178 (moving from level 6 to level 5 of the HUI hearing attribute). If the utility gain from aided hearing was assumed to be 0.384 (moving from level 6 to level 3 of the HUI hearing attribute), providing BAHAs in place of BCHA in paediatric cases had a probability of being cost-effective of 2.3% at a WTP threshold of £20,000 per QALY gained and 12.0% at a WTP threshold of £30,000 per QALY gained (*Figure 5*).

Conducting the same analysis for adults yielded similar results, with BAHAs being associated with increased QALYs [with a range from 0.01 to 0.36 for the lower utility gain (QALY1) and from 0.03 to 0.77 for the higher utility gain (QALY2)], but also increased costs (ranging from £11,362 to £17,851) compared with BCHAs (*Table 55*). BAHA costs are higher in children than in adults, owing to the use of two-stage surgery and higher costs for paediatric outpatient consultations.

Providing BAHAs in place of BCHAs for adults had a probability of being cost-effective of 0% at a WTP threshold of £20,000 per QALY gained and of 0.6% at a WTP threshold of £30,000 per QALY gained, for the lower utility gain (QALY1, moving from level 6 to level 5 of the HUI hearing dimension). For the higher utility gain (QALY2, moving from level 6 to level 3 of the HUI hearing dimension), providing BAHAs had a probability of being cost-effective of 5.0% at a WTP threshold of £20,000 per QALY gained and of 19.0% at a WTP threshold of £30,000 per QALY gained (*Figure 6*).

The PSA was re-run for small changes in the difference of the proportion of participants using their BCHA compared with BAHA. In the base-case analysis, this was assumed to be 10%, based on figures reported by Hol and colleagues.[82] In the PSA reported in *Tables 54* and *55*, the mean

TABLE 54 Mean costs and outcomes (percentile-based 95% CIs) for paediatric cases receiving a BAHA, compared with a BCHA

	Lifetime costs (£) (95% CI)	QALY1[a] (95% CI)	QALY2[b] (95% CI)
BCHA	1039 (765 to 1360)	5.20 (4.97 to 5.33)	6.74 (6.22 to 7.01)
BAHA	17,700 (14,895 to 22,496)	5.34 (5.33 to 5.35)	7.03 (7.00 to 7.05)
Incremental	16,661 (13,804 to 21,496)	0.14 (0.01 to 0.36)	0.30 (0.04 to 0.79)

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

TABLE 55 Mean costs and outcomes (percentile-based 95% CIs) for adults receiving a BAHA, compared with a BCHA

	Lifetime costs (£) (95% CI)	QALY1[a] (95% CI)	QALY2[b] (95% CI)
BCHA	1023 (751 to 1334)	5.10 (4.86 to 5.22)	6.60 (6.10 to 6.87)
BAHA	14,722 (12,440 to 18,813)	5.23 (5.22 to 5.24)	6.89 (6.86 to 6.90)
Incremental	13,699 (11,362 to 17,851)	0.14 (0.01 to 0.36)	0.29 (0.03 to 0.77)

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

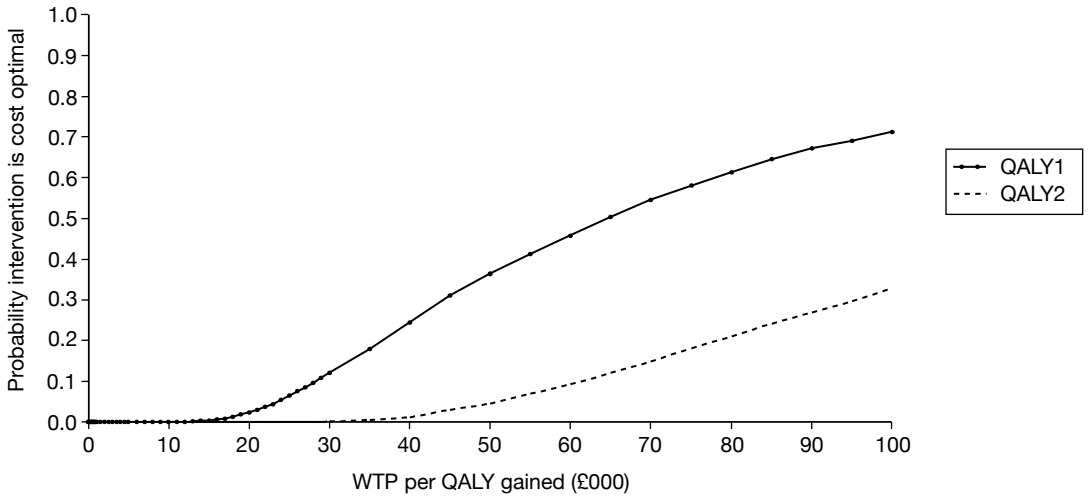

FIGURE 5 Augmented base case: cost-effectiveness acceptability curve for paediatric cases receiving a BAHA.

value for this difference was also 10%, sampled from a beta distribution that was parameterised as having two non-users of their BCHA in a population of 20 (as reported by Hol and colleagues[82]). The PSA was re-run for two alternative scenarios, in which the number of people not using their BCHA was set to three in a population of 20 (yielding an average difference of 15%) and in which the number of people not using their BCHA was set to four in a population of 20 (yielding an average difference of 20%). *Figures 7* and *8* illustrate the cost-effectiveness acceptability curves derived from these PSAs for children, assuming that the QALY gain from aided hearing is 0.178 (QALY1) and 0.384 (QALY2), respectively. *Figures 9* and *10* report the results of the same analysis for adults.

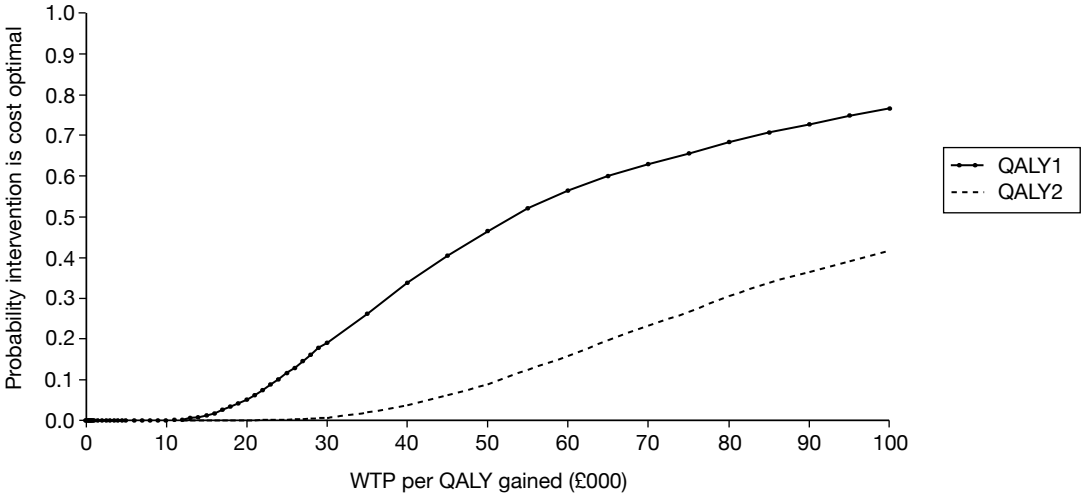

FIGURE 6 Augmented base case: cost-effectiveness acceptability curve for adults receiving a BAHA.

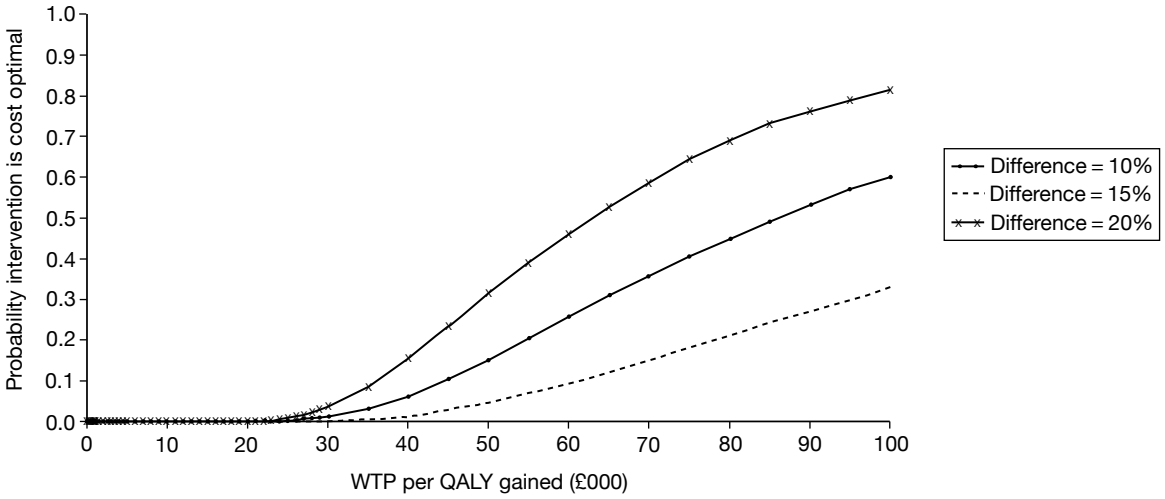

FIGURE 7 Cost-effectiveness acceptability curves for alternative assumptions on the difference in use of BCHA and BAHA in children (QALY1).

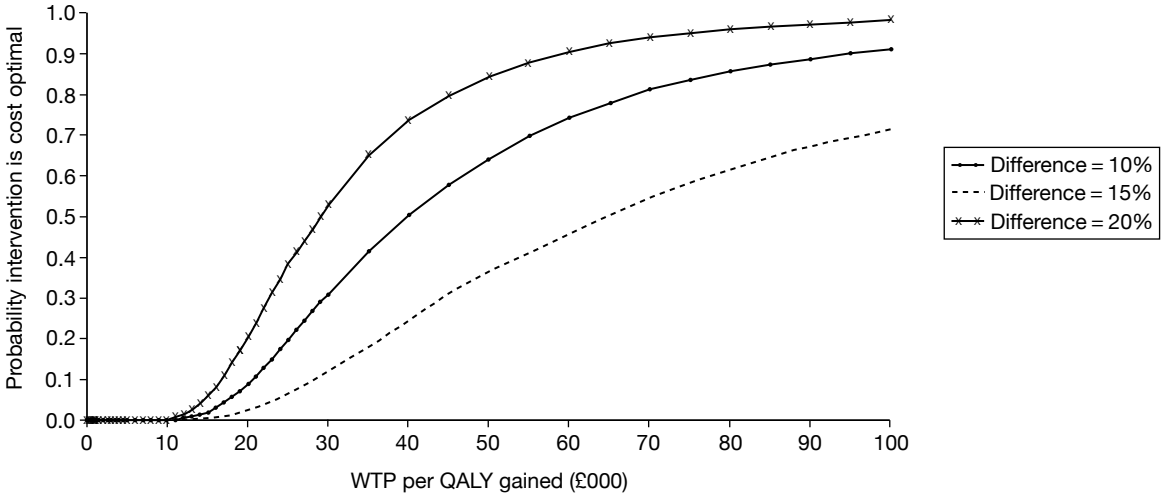

FIGURE 8 Cost-effectiveness acceptability curves for alternative assumptions on the difference in use of BCHA and BAHA in children (QALY2).

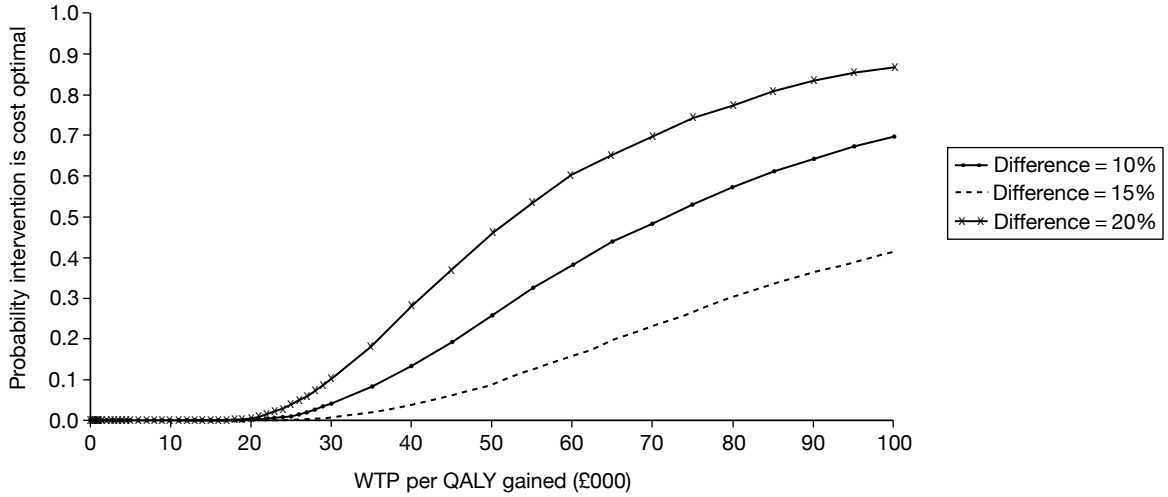

FIGURE 9 Cost-effectiveness acceptability curves for alternative assumptions on the difference in use of BCHAs and BAHAs in adults (QALY1).

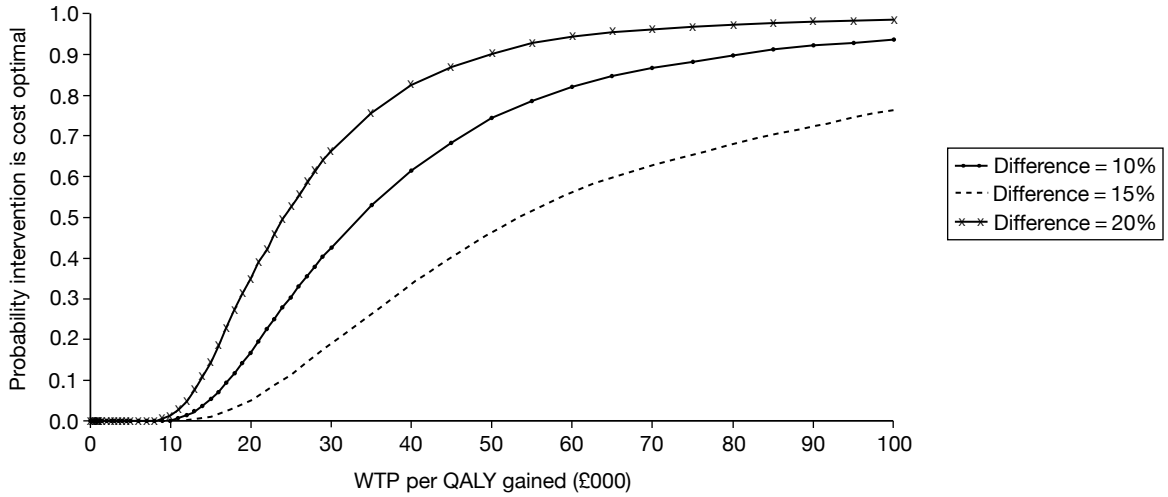

FIGURE 10 Cost-effectiveness acceptability curves for alternative assumptions on the difference in use of BCHAs and BAHAs in adults (QALY2).

Table 56 summarises the probability of BAHAs being a cost-effective option at conventionally adopted thresholds for acceptable cost-effectiveness, from each of the PSAs. These results reinforce the impact of the assumed difference in the proportion of individuals using their comparator hearing aid on the cost-effectiveness of BAHAs.

Summary of cost-effectiveness

The results of our cost analysis demonstrate that BAHAs are a significantly more costly strategy than conventional aids for people with bilateral hearing loss. These additional costs are not restricted to the initial process of surgical implantation, followed by the acquisition and fitting of the BAHA sound processor, but will continue while individuals remain using their BAHAs. Our exploratory cost-effectiveness analysis suggests that, where the benefits (in terms of hearing improvement) are similar for BAHAs and their comparators and where the probability of using

TABLE 56 Probability of a BAHA being cost-effective option (compared with a BCHA)

Patient group	Utility gain from aided hearing	Threshold WTP (£ per QALY gained)	Difference in use		
			10% (2/20)	15% (3/20)	20% (4/20)
Children	QALY1[a]	20,000	0.00	0.00	0.06
		30,000	0.10	1.26	3.66
	QALY2[b]	20,000	2.30	8.58	20.36
		30,000	11.96	30.64	52.80
Adults	QALY1[a]	20,000	0.00	0.14	0.56
		30,000	0.64	4.00	10.18
	QALY2[b]	20,000	5.00	16.68	35.02
		30,000	18.96	42.52	66.20

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Quality of life* for full details.

alternative aids for ≥ 8 hours per day is similar, BAHAs are unlikely to be a cost-effective option. The greater the benefit from BAHA-aided hearing and, in particular, the greater the difference in the proportion of people using the BCHA or BAHA for ≥ 8 hours per day, the more likely BAHAs are to be a cost-effective option. The inclusion of other dimensions of QoL may also increase the likelihood of BAHAs being a cost-effective option.

Chapter 5

Discussion

Statement of principal findings

Clinical effectiveness

The extensive search strategy did not identify any studies with a concurrent control group. The studies included in the systematic review of clinical effectiveness were rated weak overall for methodological quality and quality of reporting, and, thus, they have a high risk of bias and caution is required when interpreting the results.

BAHAs versus BCHAs

Four cohort pre and post studies included a comparison of BAHAs and BCHAs. The people included in these studies (where reported) were described as having inoperable congenital microatresia,[77] either chronic suppurative otitis media or a congenital aetiology,[78] or recurrent otorrhoea or aural atresia.[79–81] The BAHAs used in these studies are no longer manufactured and, although there should be little difference in terms of amplification between older and newer models, the newer models provide greater convenience and more flexible controls and have more benefits for mixed hearing loss. Improvements in the sound field PTA and warble-tone thresholds were found with a BAHA by two studies,[77,78] but statistical analysis was reported by only one study ($p < 0.01$).[78] The third study did not report thresholds averaged across frequencies, but found improved thresholds with the BCHA at 0.25 and 0.50 kHz, and with the BAHA at higher frequencies. A statistically significant improvement in SRT in quiet and speech-to-noise ratio was found in people with a SNHL of less than 30 dB HL,[81] while another study found no statistically significant difference in speech discrimination score in patients with either chronic suppurative otitis media or a congenital aetiology.[78] Statistical analysis was not reported for other results, which included a 23% improvement in 100% speech audiometry discrimination in noise,[77] but little difference in speech recognition threshold.[79]

Two studies reported using a validated measure of QoL,[77,82] although limited data were reported by one of the studies.[77] The second study found no statistically significant differences between BCHAs and BAHAs using the SF-36 and EQ-5D; however, a statistically significant improvement with a large clinical impact was found for handicap and disability with the HHDI.[82] The HHDI is specific to hearing loss, whereas the SF-36 and EQ-5D are generic measures that do not have a hearing dimension, which may explain the difference in outcomes between the different instruments.

In summary, while there appear to be some audiological benefits of BAHAs when compared with BCHAs, the limited evidence base of studies with a high risk of bias does not provide a reliable estimate of the degree of benefit. Improvements in QoL were identified by the hearing-specific instrument but not the generic QoL measures. Other issues such as wearability have not been adequately addressed by the included studies.

BAHAs versus ACHAs

Five cohort pre and post studies[78-81,83,84] and one cross-sectional 'audiological comparison' study[76] included a comparison of BAHAs and ACHAs. The people included in these studies (where reported) were described as having otosclerosis,[83] mixed hearing loss,[76] chronic otitis,[84] either

chronic suppurative otitis media or a congenital aetiology,[78] or recurrent otorrhoea or aural atresia.[79–81] Only one study assessed a BAHA model in current use.[76] The direction of effect for sound field pure-tone or warble-tone thresholds was inconsistent between the studies. One study found a statistically significant improvement in mean warble-tone thresholds (0.5–4.0 kHz) with a BAHA,[78] while in another study data on average warble-tone thresholds (0.2–4.0 kHz) were described as 'comparable' between BAHAs and ACHAs, but no statistical analysis was provided.[83] Three studies presented thresholds at each frequency individually, but there was no clear pattern as to the comparative benefits of ACHAs and BAHAs.[76,79,84]

The direction of the effect was also unclear for speech audiometry, with some studies finding improved outcomes with the ACHA and some with the BAHA. Two studies, which included patients with otosclerosis[83] or recurrent otorrhoea with severe hearing loss,[79] reported better outcomes with the ACHA for speech discrimination scores,[83] maximum phoneme score[79] or speech recognition threshold,[79] although statistical analysis was not conducted. A later publication by the same authors as the latter study, but with a different patient group (less severe hearing loss), found a statistically significant deterioration in SRT in quiet with BAHA ($p < 0.05$), but a statistically significant improvement in speech-to-noise ratio ($p < 0.05$).[81] One study of participants with conductive or mixed hearing loss with chronic otitis found no statistically significant difference in maximum phoneme score, but a statistically significant improvement in speech-to-noise ratio with BAHA.[84] Speech discrimination score was statistically significantly better with the BAHA in the congenital aetiology group, but not the chronic suppurative otitis media group, in one study.[78] The final study reported an improvement in speech-in-noise with the BAHA described as 'large and clinically significant' in participants with mixed hearing loss.[76] Although the ACHA may produce better audiometric results in some situations, it should be noted that the most appropriate hearing aid may not necessarily be the one with the best performance, as other factors such as the ability to wear the aid and reduced susceptibility to infections need to be considered. These issues have not been adequately addressed by the included studies.

One study reported using a validated measure of QoL.[82] A statistically significant increase in anxiety/depression with BAHAs was found by the EQ-5D, but the clinical effect was small. No other statistically significant differences were found between ACHAs and BAHAs by the EQ-5D or the SF-36; however, a statistically significant improvement with a large clinical impact was found for handicap and disability with the HHDI.[82]

One study reported the number of otolaryngology visits over the preceding 6 months for draining ears, and found a reduction with BAHAs compared with ACHAs [mean visits 12.7 (SD 10.5) vs 3.3 (SD 4.8)]; however, statistical analysis was not undertaken.[82]

In summary, the limited evidence base suggests improvements in speech understanding in noise with the BAHA compared with the ACHA; however, the ACHA may produce better audiological results for other outcomes. Improvements in QoL were identified by the hearing-specific instrument but not the generic QoL measures. Other issues such as improvement of discharging ears have not been adequately addressed by the included studies.

BAHAs versus unaided hearing

Four cohort pre and post studies included a comparison of BAHAs with unaided hearing.[66,77,78,83] The people included in these studies were described as having inoperable bilateral congenital microtia atresia,[77] otosclerosis,[83] chronic suppurative otitis media or a congenital aetiology,[78] or CHL with some mild-to-moderate sensorineural loss (details of the aetiology not reported).[66] One of the studies assessed a BAHA model in current use.[60] All four studies found improvements in sound field thresholds with the BAHA compared with unaided hearing, and

these improvements were statistically significant in the two studies that conducted analysis. Improvements were also found in speech discrimination[66,78,83] and speech recognition thresholds in quiet and noise,[66] although statistical analysis was not undertaken. No self-reported measures were reported by these four included studies. In summary, the limited evidence suggests that hearing is improved with BAHAs compared with no hearing aid.

Unilateral versus bilateral BAHAs

Four cross-sectional 'audiological comparison' studies compared unilateral with bilateral BAHAs.[59,60,86,87] The people included in these studies were described as having recurrent otorrhoea,[60] chronic otitis,[60,86,87] congenital atresia,[60,87] otosclerosis,[86,87] congenital syndromes or congenital hearing loss,[86] mastoid cavities,[86] microtia, or[86] symmetrical maximal or near-maximal conductive bilateral hearing loss (details of aetiology not reported).[59] The BAHAs used in these studies are no longer manufactured. The participants in the included studies all underwent sequential (separate operations) implantation of the bilateral BAHAs. The timing of bilateral implantation, whether sequential or simultaneous, is a major issue in cochlear implant research. It has been suggested that simultaneous cochlear implantation enables both ears to adjust to the new form of sound processing simultaneously[129] and leads to better sound localisation[130] in young children, but the timing in adults appears to be less critical.[130] However, as BAHAs do not selectively stimulate each side, these issues may not be so important.

Sound field average tone thresholds were improved with bilateral BAHAs compared with unilateral BAHAs in adults[87] and a small group ($n = 3$) of children[59] with previous experience of bilateral BAHAs, but statistical analysis was not undertaken. Two studies found that speech recognition thresholds in quiet were statistically significantly lower with bilateral BAHAs,[60,87] although another study found similar results between unilateral and bilateral BAHAs.[86] Bilateral BAHAs produced better results than one BAHA when noise was presented from the baffle/best side (the side with the BAHA in the unilateral condition), but not when noise was presented from the shadow side (the side opposite to the BAHA in the unilateral condition);[60,86,87] this is explained by the increase in noise transmitted to the ears with an extra BAHA on the shadow (noise) side. Three studies found that localisation of sound was improved with bilateral BAHAs.[59,60,87] Two studies reported the binaural masking level difference test and suggested that BAHAs give binaural hearing,[60,87] although the validity of the methods is uncertain. One study described self-reported measures using a validated tool (MAIS and MUSS, and IOI-HA), but the sample size was very small ($n = 2$ or $n = 3$ for the bilateral users group). In summary, the limited evidence suggests that there are benefits of bilateral BAHAs in many, but not all, situations, and the presence of binaural hearing with bilateral BAHAs remains uncertain.

Adverse events

As the included studies reported very limited data on adverse events, additional data from prospective case series were described. It should be noted that these studies did not undergo the same process of data extraction and quality assessment. Five prospective case series were discussed and reported loss rates of implants between 6.1% (9–25 months' follow-up)[102] and 19.4% (median 6 years' follow-up).[100] The vast majority of patients in the prospective case series experienced no or minor skin reactions.

Cost-effectiveness

Published economic evaluations

The search strategy did not identify any fully published economic evaluations of BAHAs. Two studies[107,108] reporting resource use or cost data for patients receiving BAHAs were reviewed for their relevance. One cost study,[107] focusing on outpatient BAHA implantation and conducted in the US, was not relevant to the perspective of the current review. The second cost study,[108] a UK retrospective analysis of service use before and after BAHA implantation, was used in conjunction

with current service standards to identify the management pathway for individuals considered eligible for a BAHA, and as a basis for costing the intervention in our economic model.

Unpublished economic evaluations

An unpublished, UK-based, economic evaluation of BAHAs was identified.[110] Although not meeting the inclusion criteria for this review (33.3% of included participants were stated as having bilateral hearing loss, the remainder were unilateral/single-sided or not stated); the methods and data inputs of the model were briefly reviewed. The analysis was based on patient-level data for adult patients undergoing primary BAHA implantation at UHB. Unit costs for surgery were based on PCT charges for BAHAs and for digital (non-BAHA) hearing aids. Health outcomes were assessed using QALYs, based on each patient's age-sex-specific life expectancy with utility values based on responses to the HUI3. Mean HUI3 utility at baseline was reported as 0.59 (95% CI 0.53 to 0.65) and post-BAHA was 0.66 (95% CI 0.60 to 0.72). The incremental cost for BAHA provision was £20,604. The mean discounted QALY gain was 1.17, yielding an ICER of £17,610.

Southampton Health Technology Assessments Centre economic model

We developed a new model to estimate the cost-effectiveness of BAHAs in separate cohorts of eligible adults and children with bilateral deafness. Owing to data limitations identified in the clinical effectiveness section above, the model was limited to comparing BAHAs against BCHAs.

Owing to limitations of the evidence base, the model did not incorporate direct measures of gain in hearing from included studies. As a result, the model reports a cost comparison – of BAHAs compared with BCHAs – and an exploratory cost-effectiveness analysis using estimated potential utility gains based on levels of the hearing dimension of the HUI3 and limited data on the use of BCHAs compared with BAHAs. The analysis assumes that the utility gain from aided hearing is the same for both BAHAs and BCHAs and that differences in outcome for the devices arise from differences in the proportion of individuals using the devices.

Included studies were reviewed for information on the incidence of adverse events, to populate the economic model. Adverse events in the model were limited to perioperative complications (bleeding), failure of initial bone integration, pain leading to removal of the implant, skin reactions and loss of bone integration. We did not identify sources that reported separate incidence of adverse events for children and adults; hence the same event probabilities are used for both populations.

Resource use in the model was estimated based on published reports,[108] current audiology service standards for BAHAs[109] and discussion with clinical experts. We identified a management pathway from an initial consultation with an ENT surgeon, through to surgical implantation and long-term management. Unit costs were derived based on NHS reference costs[43] – where available – and from NHS providers (for costs of components of the BAHA system and comparator hearing aids).

In the cost analysis, the BAHA is the more costly strategy, increasing costs in children by approximately 94% over the BCHA (from £1105 to £17,514) and increasing costs in adults by 93% (from £1084 to £14,533). The higher costs for BAHA provision in children arise from the use of two-stage surgery and from higher outpatient costs for paediatric cases. The single most costly phase of BAHA provision is long-term maintenance (at £6241 for paediatric cases and £6114 for adults), although the majority of BAHA cost is incurred in the first year (for implant surgery and the cost of the BAHA sound processor). An average cost per case successfully treated was estimated, allowing for a proportion of participants choosing not to continue with their BAHA owing to pain from the implant or choosing not to have a re-operation following late failure due

to loss of bone integration or a severe skin reaction leading to implant removal. It was estimated that up to 6% of the initial cohort would have ceased using their BAHA by the end of the 10-year time horizon. Under these assumptions the cost per case successfully treated is £18,681 for children and £15,785 for adults. In a DSA, the results of the cost analysis were generally robust to variation in the value of input parameters. The results were most sensitive to variation in the probability of re-operation when implants lose bone integration, the cost of surgical implantation and the cost of the BAHA processor maintenance plan.

In the absence of usable QoL data for people with bilateral hearing loss or methods to map changes in hearing measures reported in included studies to QoL, we conducted an exploratory cost-effectiveness analysis. This incorporated assumptions regarding the potential gains from aided hearing in people with bilateral hearing loss, who are unable to hear one-to-one or group conversation without a hearing aid, and limited data on use of BCHAs and BAHAs in BAHA-eligible subjects (indicating a 10% increase in the proportion of patients using their BAHA compared with usage of BCHAs). Under these assumptions, provision of BAHAs resulted in a QALY gain of between 0.14 and 0.30 for paediatric cases (depending on the assumed level of utility associated with aided hearing). Combined with the incremental cost estimates described above, these yielded ICERs of £119,367 and £55,642 per QALY gained, respectively. Applying the same analysis to adults yields similar QALY gains from BAHA provision (0.14 and 0.29 QALYs), resulting in lower ICERs (£100,029 and £46,628 per QALY gained, respectively), given the lower incremental costs estimated for adults. These ICERs are high and above conventionally adopted thresholds for acceptable cost-effectiveness in an NHS decision-making context.

Deterministic sensitivity analyses for the exploratory cost-effectiveness analysis suggest that the results are generally robust to variation in input probabilities and unit costs. The variable that has the greatest influence on the cost-effectiveness results is the proportion of each cohort using their hearing aids for 8 or more hours per day. Very high ICER values are associated with a high proportion of people using BCHA for 8 or more hours per day (98.8% at the upper limit of the 95% CI, resulting in a small difference in usage between BCHA and BAHA), but more acceptable values are associated with a lower proportion using BCHA for 8 or more hours per day (compared with BAHA).

Threshold values for differences in use of hearing aids, the presence of pain/discomfort associated with the use of BCHA and the underlying utility gain from aided hearing were explored in a range of scenario analyses. Where the utility gain from aided hearing is related to the ability to hear one-to-one conversation in quiet, the difference in the proportion of people using their hearing aid for 8 hours or more per day needs to be greater than 33% in adults (greater than 39% in children) for BAHAs to be a cost-effective option (at a WTP threshold of £30,000 per QALY). Where the utility gain from aided hearing is related to the ability to hear both one-to-one conversation in quiet and group conversation, the required difference in the proportion of people using their hearing aid for 8 hours or more per day for BAHAs to be a cost-effective option is lower (greater than 15% in adults, 18% in children). Where pain/discomfort is included in the analysis, the ICERs fall substantially, with BAHAs appearing to be a cost-effective option (if the utility gain from aided hearing is related to the ability to hear one-to-one conversation in both quiet and group conversation).

General discussion

The findings of the systematic review of clinical effectiveness are in line with those of a previous systematic review,[11] which assessed the non-acoustic benefits of BAHAs. The earlier review, however, included studies of unilateral as well as bilateral deafness and also included

retrospective studies. The authors concluded that there is limited statistically supported, empirically controlled evidence supporting the non-acoustic benefits of BAHAs relative to more conventional hearing aids or no hearing aids at all. No other systematic reviews of BAHAs for bilateral hearing loss were identified.

The conclusions drawn from the present systematic review of clinical effectiveness are constrained by the limitations of the available evidence. Despite conducting a wide-ranging and systematic search of the literature, no trials with a concurrent control group (either RCTs, controlled clinical trials or prospective cohort analytic studies) were identified. The included studies were rated overall as weak, therefore there is a high risk of bias in the studies. The outcome measures reported by the studies also have limitations, and it is not always clear what is clinically significant or meaningful to the patient. Audiological measures such as hearing threshold levels or speech reception levels in quiet may be too simplistic, and a measure such as 'match to target' may be more meaningful; however, this was not reported by the included studies. Lower hearing thresholds are considered to be better than higher thresholds throughout the review, but it is acknowledged that this is a simplistic approach and may not necessarily be the case if they are below the target value. The review uses the study authors' descriptions such as 'improvement' or 'deterioration' where available.

Only three studies[59,77,82] reported using a validated measure of QoL; thus, it is difficult to make any judgement about the impact of BAHAs on the QoL of a person with bilateral hearing loss. An important issue for the individual is comfort and the ability to wear the hearing aid, especially if he or she cannot wear conventional hearing aids. For example, if the aid cannot be worn owing to discomfort or a discharging ear, then it is not appropriate. These issues are not considered by audiological comparisons of BAHAs with conventional aids. Although some included studies reported patient preference, the tools used were not validated and were likely to be biased, especially considering evidence that suggests that patients report preferring the second hearing aid tested, even if it is in fact an identical aid.[130] Synthesis of the included studies was through narrative review; although 12 studies were included in the review of clinical effectiveness, differences in participants, comparator (ACHA, BCHA or unaided) and outcome measures meant that meta-analysis was inappropriate. No prospective studies comparing BAHAs with ear surgery were identified; thus, no conclusions could be drawn.

There are potentially many benefits of BAHAs for individuals and their families, but these are difficult to quantify and there is little evidence available. In children, improved speech and language development may lead to a reduced need for specialist schooling and involvement of teachers for the deaf. Children may perform better at school, potentially leading to better employment opportunities in the future. For adults, an improvement in the discharging ear may mean attending fewer ENT clinics, less absence from work and again better employment opportunities. There may also be benefits in terms of improved road safety with bilateral BAHAs. These factors could not be addressed in the present report.

Bone-anchored hearing aid technology is continuously evolving and as a result the majority of the evidence in this review is based on BAHA devices that are no longer manufactured. The newer models may have greater benefits for mixed hearing loss than for CHL. They provide more convenience with flexible controls and a directional microphone.

With regards to service provision, BAHAs are likely to remain a specialist service as a part of a comprehensive audiological rehabilitation, with a small number of centres. Children may be more appropriately cared for in specialised children's units because of comorbidities and anaesthetic difficulties. The number of people who meet the existing BAHA criteria and who could potentially benefit from BAHAs is unclear. There is also currently a lack of awareness

about BAHAs, both in primary care and in audiology departments where there is no BAHA programme,[131] so current provision is likely to be below potential. It is thought that growing awareness will lead to increased referrals for BAHA services, which in turn will lead to an escalating number of BAHAs that need repairing, replacing or upgrading.[132] Commissioners of services in the NHS will therefore need to make decisions as to how they support this need. Potential benefits to the NHS may include a decrease in ENT attendance for discharging ears.

Strengths and limitations of the assessment

This review has the following strengths:

- It is independent of any vested interest.
- It has been undertaken following the principles for conducting a systematic review. The methods were set out in a research protocol (see *Appendix 1*), which defined the research question, inclusion criteria, quality criteria, data extraction process and methods to be employed at different stages of the review.
- A multidisciplinary advisory group has informed the review from its initiation. The research protocol was informed by comments received from the advisory group and the advisory group has reviewed and commented on the final report.
- The review brings together the evidence for the clinical effectiveness and cost-effectiveness of BAHAs for people who are bilaterally deaf. This evidence has been critically appraised and presented in a consistent and transparent manner.
- An economic model has been developed de novo following recognised guidelines, and systematic searches have been conducted to identify data for the economic model. The main results have been summarised and presented.

In contrast, this review also has certain limitations:

- Twenty-eight relevant non-English references were identified by the searches, and although titles and English abstracts (where available) were examined, the papers were not translated and screened. However, none of the papers appeared to present higher level evidence and it is unlikely that the inclusion of more low-level evidence would change the conclusions of this report.
- The rigorous methods of the review meant that many of the available data on QoL, patient preference, patient satisfaction, comfort and wearability did not meet the inclusion criteria for the systematic review of clinical effectiveness owing to study design and the use of tools that were not validated.
- Limited data on adverse events were reported in the included studies; thus, data from eligible prospective case series were described. Although data from these studies were extracted by one reviewer and checked by a second reviewer, these studies did not undergo the same process of quality assessment.
- The economic evaluation presented in this report is severely limited by a lack of robust evidence on the outcome of hearing aid provision. This has led to a more restricted analysis than was originally anticipated (limited to a comparison of BAHA with BCHA). In the absence of usable QoL data, the cost-effectiveness analysis is based on potential utility gains from hearing that have been inferred using a QoL instrument rather than measures reported by hearing aid users themselves. As a result, the analysis is regarded as exploratory and the reported results should be interpreted with caution.
- Given the exploratory nature of the economic evaluation (particularly the fact that the utility data used to estimate QALYs), we felt that it was not appropriate to extend this to include a value of information analysis (as we originally proposed).

Chapter 6

Conclusions

Implications for service provision

The findings suggest that hearing is improved with BAHAs compared with no hearing aid and, although there are audiological benefits of BAHAs when compared with conventional BCHAs, the audiological comparisons with ACHAs are more equivocal. However, candidates for BAHAs may not be able to use these conventional aids, or be able to use them for only a limited time, for example owing to discomfort or infections. Limited data suggest an improvement in QoL with BAHAs when compared with conventional aids, but there is an absence of evidence regarding other potential benefits, such as length of time the aid is able to be worn and improvement of discharging ears. The evidence suggests that there are some benefits of bilateral BAHAs compared with unilateral BAHAs. BAHAs are significantly more costly than conventional hearing aids. The additional costs continue while individuals remain using their BAHAs and are not restricted to initial surgical procedures and acquisition of the BAHA sound processor. Our exploratory cost-effectiveness analysis suggests that BAHAs are most likely to be cost-effective in people with the greatest benefit from aided hearing and, in particular, with greater difference in usage of BAHAs compared with conventional aids. Inclusion of other dimensions of QoL (other than hearing) may also increase the likelihood of BAHAs being a cost-effective option. The conclusions are limited by the quality of the available evidence.

Suggested research priorities

- There are many areas of uncertainty surrounding BAHAs, including, but not limited to: the need for BAHA services; resource implications (both costs and potential savings through reduced ENT attendance); type of service provision (e.g. specialist centres); benefits of BAHAs; usage of conventional hearing aids in patients eligible for BAHAs; and adverse events. A national audit of BAHAs should be implemented, which will provide clarity on these issues. The collection of such data would significantly increase the robustness of economic evaluation of BAHAs and would, potentially, broaden the scope of comparators beyond BCHAs.
- Further research is required into the non-audiological benefits of BAHAs, including QoL, improvement of discharging ears and time wearing the aid. While an RCT would be preferable, many patients are referred for BAHAs as a last resort when conventional aids are unsuitable; therefore, a controlled clinical trial or a prospective cohort analytic study (two groups' before and after study) is suggested.
- A randomised crossover study comparing unilateral and bilateral BAHAs is required. The order of BAHA (unilateral or bilateral) should be randomised, and participants should have at least 12 weeks' experience with each aid before assessment.
- The number of people who are potentially eligible for BAHAs is not known. Further research into the incidence and prevalence of hearing loss, and of conductive and mixed hearing loss in particular, is required.
- Empirical studies into the masking level difference with BC in people with normal hearing and CHL are required.

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J Jones (Principal Research Fellow) developed the original research grant application, developed the research protocol, assessed studies for inclusion, extracted data from and quality assessed included studies, synthesised evidence, developed the economic evaluation and drafted the report.

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Appendix 1

Protocol methods

Research methods for synthesis of evidence of clinical effectiveness and cost-effectiveness

A systematic review will be undertaken in accordance with the NHS Centre for Reviews and Dissemination guidelines,[133] published guidelines on meta-analysis[133] and criteria for appraising economic evaluations.[134]

Search strategy

A search strategy will be developed and tested by an experienced information scientist. Literature will be identified from several sources including electronic databases, bibliographies of articles, grey literature sources and hand searching of specialist journals. A comprehensive database of relevant published and unpublished articles will be constructed using Reference Manager (refman) software (Thomson Reuters, London). Searches to identify studies will be carried out via a number of routes:

1. general health and biomedical databases including MEDLINE, EMBASE, Science Citation Index, BIOSIS
2. specialist electronic databases: Database of Abstracts of Reviews of Reviews of Effects, The Cochrane Library
3. grey literature and conference proceedings
4. contact with individuals with an interest in the field
5. checking of reference lists
6. research in progress databases: NIHR Clinical Research Network Portfolio (formerly UK Clinical Research Network website, Current Controlled Trials), Clinical trials.gov.

The draft search strategy for MEDLINE is shown in *Appendix 2*. This will be adapted for other databases. All databases will be searched from inception to the current date with no language restrictions. Hand searching will focus on key meeting abstracts published in the past 2 years identified in consultation with experts and analysis of searches.

Planned inclusion/exclusion criteria

The planned inclusion/exclusion criteria for the systematic review are shown in *Table 57*.

Studies will be selected for inclusion through a two-stage process using the predefined and explicit criteria. The full literature search results will be screened by two reviewers to identify all citations that may meet the inclusion criteria. Full manuscripts of all selected citations will be retrieved and assessed by two reviewers against the inclusion criteria. An inclusion flow chart will be developed and used for each paper assessed. Any disagreements over study inclusion will be resolved by consensus or if necessary by arbitration by a third reviewer.

TABLE 57 Inclusion criteria for systematic reviews

Participants	Adults or children with bilateral deafness
	Papers reporting both bilateral and unilateral hearing loss will be included if the groups are reported separately or if the majority of participants have bilateral hearing loss
	Single-sided deafness will be excluded
Interventions	■ BAHAs (i.e. attached to a surgically implanted titanium fixture)
Comparators	■ Unilateral vs bilateral BAHAs
	■ Conventional hearing aids (AC or BC)
	■ Unaided
	■ Ear surgery: tympanoplasty, myringoplasty, ossiculoplasty, stapedectomy, stapedotomy
Outcomes	■ Validated measures of QoL, patient satisfaction and hearing measures, aided hearing thresholds, speech recognition scores, adverse events, complications
	■ Measures of cost-effectiveness (e.g. cost per QALY, cost per life-year saved), consequences to health service resources
Study design	■ Clinical effectiveness: RCTs, controlled clinical trials, prospective cohort analytic studies (i.e. with control group), prospective cohort (one group pre and post) studies and prospective case series
	■ For each comparator listed above: where evidence from different types of study design is identified, only those studies with the most rigorous designs will be included; where higher level evidence is limited to BAHA models no longer in current use, lower level evidence for models in current use (Divino, Intenso, Cordelle II) will also be considered
	■ Cost-effectiveness studies (including measures of costs and consequences)
	■ Studies published as abstracts or conference presentations will be included only if sufficient details are presented to allow an appraisal of the methodology and the assessment of results to be undertaken

Data extraction and quality assessment

Data extraction and quality assessment will be undertaken by one reviewer and checked by a second reviewer using a predesigned and piloted data extraction form to avoid any errors. The methodological quality of all included studies will be appraised using recognised quality assessment tools[73] and criteria for appraising economic evaluations.[75,134] The tool selected for assessing the quality of primary studies of clinical effectiveness has been recognised as one of the more comprehensive sets of criteria for assessing the quality of different study designs.[135] Where possible, missing information will be obtained from investigators. Any disagreements between reviewers will be resolved by consensus or if necessary by arbitration by a third reviewer.

Data synthesis

Studies will be synthesised through a narrative review with tabulation of results of included studies. Where possible, the results from individual studies will be synthesised through meta-analysis, with causes of heterogeneity of results examined. The specific methods for meta-analysis and for the detection and investigation of heterogeneity will depend upon the summary measure selected.

Southampton Health Technology Assessments Centre economic model

If the systematic review of cost-effectiveness of BAHAs for people who are bilaterally deaf finds any relevant high-quality economic evaluations, the feasibility of adapting and updating these existing models will be investigated. In the absence of relevant high-quality, model-based economic evaluations, a de novo decision analytic model will be developed. The model will be structured using published evidence on the epidemiology and natural history of bilateral

deafness, and will be informed by guidance from clinical advisors, to reflect the natural course of bilateral deafness and the impact of alternative interventions. Accepted guidelines for good practice in decision-analytic modelling and the general principles outlined in the National Institute for Health and Clinical Excellence (NICE) 'reference case'[75,136] will be followed. The model will be used to provide a cost–consequence analysis, reporting the costs of interventions included in the systematic review and their consequences in terms of hearing measures, QoL, complications, and health service resource use for bilaterally deaf patients receiving standard hearing aids (including BC hearing aids), surgery, unilateral BAHAs and bilateral BAHAs. The model will also be used to estimate the longer term consequences in terms of quality-adjusted life expectancy. The model will adopt a UK NHS and PSS perspective. The time horizon for the long-term model will be the patients' lifetime, with health outcomes expressed in terms of QALYs – costs and QALYs will be discounted at an annual rate of 3.5%.

Development of the structure of the model will be informed by several sources including previous models identified in the systematic review of cost-effectiveness, evidence on the epidemiology and natural history of bilateral deafness and guidance from clinical and methodological advisors. The economic model will only include clinically relevant comparators found to be clinically effective by the systematic review. Evidence of effectiveness will originate from the systematic review. Specific targeted literature searches will be required to populate other parameters in the model, including baseline characteristics of the population requiring intervention, age-condition-specific life expectancy and the impact of deafness on patient satisfaction and health-related QoL. Information on adverse events and complications will come from the systematic review of effectiveness.

Resource use and unit costs, including consultations (e.g. ENT surgeon, audiologist), treatments, adverse events and complications will be obtained from published evidence, official sources such as *Unit costs of health and social care*[137] and NHS Reference Costs,[138] and from the Costing Unit at Southampton General Hospital. Costs of hearing aids, both BAHAs and conventional devices, will be taken from published tariff prices for the UK.[139,140] Costs will be inflated to current prices as necessary. If no published data are available, we will consult with expert advisors to obtain estimates for the parameters relating to resource use.

The results of the economic model will be presented as a cost–consequence analysis, clearly specifying the direct costs associated with each intervention and consequences in terms of hearing, QoL and adverse events of each intervention, and as a cost–utility analysis. The results of the cost–utility analysis will be presented as ICERs for the base case and using CEACs to show the probability of each device being cost-effective at different WTP thresholds. Uncertainty will be examined using deterministic sensitivity analysis and PSA. The importance of the underlying model assumptions will be assessed through an analysis of different scenarios. Value of information analysis will be undertaken to help inform payback in terms of reduced parameter uncertainty from additional research, identifying which parameters most contribute to decision uncertainty and should therefore be the focus of future research.[141–143]

The model will be constructed in Microsoft Excel 2003 (Microsoft Corporation, Redmond, WA, USA) to ensure transparency. All stages in the development of the model, analysis of data and interpretation of results will be undertaken by one health economist and checked by a second. All model assumptions and data sources will be clearly specified and their effects on outcomes checked through sensitivity analysis, to ensure model results accurately reflect the inputs used. Internal consistency will also be assessed through the replication of the model in different software to compare results. External consistency will be assessed through comparing results with the previously published analyses.

Appendix 2

Search strategy

All databases searched for the systematic review are presented below.

Database searched	Clinical effectiveness searches	Cost-effectiveness and QoL searches
BIOSIS (Web of Science)	1990–2009	1990–2009
Cochrane Central Register of Controlled Trials (CENTRAL, The Cochrane Library)	All available years	All available years
Cochrane Database of Systematic Reviews (The Cochrane Library)	All available years	All available years
Database of Abstracts of Reviews of Effects (Centre for Reviews and Dissemination)	All available years	All available years
EconLit (American Economic Association's electronic bibliography, Ebsco)	All available years	Searched 11 November 2009
EMBASE	1980–2009	1980–2009
Health Technology Assessment Database (Centre for Reviews and Dissemination)	All available years	Searched 11 November 2009
Health Management Information Consortium	All available years	Searched 11 November 2009
MEDLINE (Ovid)	1950–2009	1950–2009
MEDLINE In-Process & Other Non-Indexed Citations	1950–2009	1950–2009
NHS Economic Evaluation Database (Centre for Reviews and Dissemination)	All available years	Searched 11 November 2009
Web of Science: Proceedings Citation Index (Web of Knowledge)	1970–2009	1970–2009
Web of Science: Science Citation Index (Web of Knowledge)	1970–2009	1970–2009
Searched for ongoing trials		
National Institute for Health Research Clinical Research Network Portfolio (formerly UK Clinical Research Network website)		
Current Controlled Trials		
Clinical trials.gov		
Center Watch		
Computer Retrieval of Information on Scientific Projects		
Health Services Research Projects in Progress		

The MEDLINE search strategy (presented below) for the systematic review of clinical effectiveness was adjusted as necessary for other electronic databases for both clinical effectiveness and cost-effectiveness (including QoL information) searches. Search strategies for the systematic review are available from the authors on request. Citations identified by the searches were added to a Reference Manager database.

MEDLINE search strategy

1. exp Deafness/ (20,783)
2. ((mixed adj5 deaf*) or (mixed adj5 hearing adj loss*)).ti,ab. (392)
3. (sensorineural* adj5 deaf*).ti,ab. (1277)
4. (bilateral* adj5 deaf*).ti,ab. (724)
5. exp Hearing Loss/ (42,445)
6. Hearing Disorders/ (12,599)

7. Hearing Impaired Persons/ (689)
8. (hearing loss* adj5 bilateral*).ti,ab. (1301)
9. (hearing loss* adj5 conductive).ti,ab. (1506)
10. (hearing loss* adj5 sensorineural).ti,ab. (5810)
11. Hearing Loss, Sensorineural/ (9843)
12. Hearing Loss mixed conductive sensorineural/ (55)
13. Hearing Loss, Bilateral/ (1324)
14. Hearing Loss, Conductive/ (2348)
15. (hearing adj2 loss*).ti,ab. (21,232)
16. hearing loss noise induced/ (5132)
17. "Rehabilitation of Hearing Impaired"/ (1203)
18. (hearing adj5 impair*).ti,ab. (8111)
19. **or/1-18 (61,735)**
20. Bone Conduction/ (1998)
21. exp Osseointegration/ (4946)
22. osseointegrat*.ti,ab. (3525)
23. exp hearing aids/ (9717)
24. 23 and (20 or 21 or 22) (320)
25. (divino or intenso or cordelle).ti,ab. (9)
26. (divino or intenso or cordelle).mp. (38)
27. (classic adj1 "300").ti,ab. (6)
28. (HC adj1 "300").ti,ab. (3)
29. (HC adj1 "100").ti,ab. (28)
30. (HC adj1 "200").ti,ab. (13)
31. (HC adj1 "210").ti,ab. (1)
32. (HC adj1 "220").ti,ab. (1)
33. (HC adj1 "300").ti,ab. (3)
34. (HC adj1 "360").ti,ab. (0)
35. (HC adj1 "380").ti,ab. (0)
36. (HC adj1 "400").ti,ab. (2)
37. temporal bone/ (7731)
38. prosthesis implantation/ (4922)
39. bone anchored.mp. (387)
40. 23 and 37 (228)
41. 23 and 38 and 39 (40)
42. or/24–36,40-41 (584)
43. **19 and 42 (323)**
44. (bone anchor* and (hear* or deaf*)).ti,ab. (259)
45. 23 and (BAHA or BAHAs or "BAHA's").ti,ab. (164)
46. 19 and (BAHA or BAHAs or "BAHA's").ti,ab. (138)
47. ((BAHA or BAHAs or "BAHA's") and (hear* or deaf*)).ti,ab. (172)
48. 19 and 38 and 39 (28)
49. (bone anchor* adj5 hearing aid*).ti,ab. (246)
50. **43 or 44 or 45 or 46 or 47 or 48 (470)**
51. (letter or comment or editiorial).pt. (769,498)
52. 50 not 51 (458)
53. limit 52 to humans (453)
54. from 53 keep 1-453 **(453)**

Update searches run on 18 November 2009.

Reference lists

The reference lists of retrieved articles were examined for additional studies.

Other searches

The experts advisory group and BAHA manufacturers were contacted in order to obtain information about additional references and any ongoing studies.

British societies and conferences (sources checked on 25 November 2009)

American Academy of Otolaryngology-Head and neck Surgery.
American Otological Society.
American Society of Pediatric Otolaryngology.
Association for Research in Otolaryngology.
BAHA Professionals Group.
Baha User Group.
British Academy of Audiology.
British Association of Paediatric Otolaryngology.
British Society of Hearing Aid Audiologists.
Canadian Society of Otolaryngology.
Deafness Research.
Ear Foundation.
European Academy of Otorhinolaryngology.
European Academy of Otology & Neuro-Otology.
European Academy of Otorhinolaryngolgy, Head and Neck Surgery.
European archives of Oto-Rhino-Laryngology.
European Federation of Audiology Societies.
European Federation of Oto-Rhino-Laryngological Societies.
European Society of Pediatric Otorhinolaryngology.
Hearing Aid Council.
Institute of Hearing Research.
National Deaf Children's Society.
Royal National Institute for Deaf People.
Scottish Otolaryngological Society.

Appendix 3

List of excluded studies

Albrektsson T, Branemark PI, Jacobsson M, Tjellstrom A. Present clinical applications of osseointegrated percutaneous implants. *Plast Reconstr Surg* 1987;**79**:721–31. Reason for exclusion: design.

Arthur D. The Vibrant Soundbridge™ *Trends Amplif* 2002;**6**:67–72. Reason for exclusion: intervention.

Blackmore KJ, Kernohan MD, Davison T, Johnson IJ. Bone-anchored hearing aid modified with directional microphone: do patients benefit? *J Laryngol Otol* 2007;**121**:822–5. Reasons for exclusion: intervention; comparator.

Bonding P, Nielsen LH, Pedersen U, Brask T. The Danish BAHA file – preliminary results. In Portman M, Boudard P, Portmann D, editors. *Transplants and implants in otology – III*. New York, NY: Kugler Publications; 1996. pp. 297–9. Reason for exclusion: paper not available from British Library (conference proceedings dated 1996).

Bosman AJ, Snik AFM, Mylanus EAM, Cremers CWRJ. Fitting range of the BAHA Intenso. *Int J Audiol* 2009;**48**:346–52. Reason for exclusion: comparator.

Browning GG. The British experience of an implantable, subcutaneous bone conduction hearing aid (Xomed Audiant). *J Laryngol Otol* 1990;**104**:534–8. Reasons for exclusion: intervention; comparator; outcomes; design.

Buratti C, Romagnoli M, Galli A, Parmigiani F. Bone-anchored hearing-aid – our experience in children with congenital external and middle-ear malformations. *Child and the Environment – Present and Future Trends* 1993;**1012**:256–8. Reason for exclusion: outcomes.

Cano FAC, Blass FA. Branemark bone-anchored implants for hearing-aid adaptation. *Otolaryngol Head Neck Surg* 1991;1221–9. Reason for exclusion: design.

Carlsson P, Håkansson B. The bone-anchored hearing-aid. *Child and the Environment – Present and Future Trends* 1993;**1012**:247–50. Reasons for exclusion: outcomes; design.

Davison T, Marley S, Leese D, Johnson I. *Clinical impressions of a new bone anchored hearing aid processor*. Unpublished. 2009. Reason for exclusion: comparator.

Dunham ME, Friedman HI. Audiologic management of bilateral external auditory canal atresia with the bone conducting implantable hearing device. *Cleft Palate J* 1990;**27**:369–73. Reason for exclusion: intervention.

Durvasula VS, Patel H, Mahendran S, Gray RF. Bone anchored hearing aids: a second fixture reduces auditory deprivation in Cambridge. *Eur Arch Otorhinolaryngol* 2007;**264**:991–4. Reason for exclusion: design.

Dutt SN, McDermott AL, Burrell SP, Cooper HR, Reid AP, Proops DW. Patient satisfaction with bilateral bone-anchored hearing aids: the Birmingham experience. *J Laryngol Otol* 2002:37–46. Reason for exclusion: study design.

Flynn MC, Sadeghi A. *Results of the first clinical evaluation of Cochlear TM Baha BP100*. 2009. URL: http://bp100.cochlear.com/sites/default/files/E81511%20Results%20of%20the%20first%20clinical%20evaluation%20of%20Cochlear%20Baha%20BP100%20whitepaper.pdf (accessed 16 September 2009). Reason for exclusion: comparator.

Forton GEJ, Van De Heyning PH. Bone anchored hearing aids (BAHA). *B-ENT* 2007:45–50. Reason for exclusion: design, outcomes.

Gatehouse S, Browning G. An evaluation of the role of bone-anchored hearing aids in the management of hearing impairment. *Clin Otolaryngol* 1992;**17**:462. Reason for exclusion: outcomes.

Goodyear PW, Raine CH, Firth AL, Tucker AG, Hawkins K. The Bradford bone-anchored hearing aid programme: impact of the multidisciplinary team. *J Laryngol Otol* 2006;**120**:543–52. Reason for exclusion: design.

Granstrom G, Bergstrom K, Tjellstrom A. The bone-anchored hearing aid and bone-anchored epithesis for congenital ear malformations. *Otolaryngol Head Neck Surg* 1993;**109**:46–53. Reasons for exclusion: outcomes; design; participants.

Granstrom GPB, Bergstrom KM, Tjellstrom AMR. Some considerations regarding the rehabilitation of patients with congenital ear malformations using the osseointegration concept. In Portman M, Boudard P, Portmann D, editors. *Transplants and implants in otology – III*. New York, NY: Kugler Publications; 1996. pp. 91–8. Reasons for exclusion: outcomes; design.

Håkansson B, Tjellstrom A, Rosenhall U. Hearing thresholds with direct bone conduction versus conventional bone conduction. *Scand Audiol* 1984;**13**:3–13. Reason for exclusion: outcomes.

Håkansson B, Tjellstrom A, Rosenhall U. Acceleration levels at hearing threshold with direct bone conduction versus conventional bone conduction. *Acta Otolaryngol* 1985;**100**:240–52. Reason for exclusion: outcomes.

Holgers KM, Tjellstrom A, Bjursten LM, Erlandsson BE. Soft tissue reactions around percutaneous implants: a clinical study of soft tissue conditions around skin-penetrating titanium implants for bone-anchored hearing aids. *Am J Otol* 1988;**9**:56–9. Reason for exclusion: design.

Johnson RM, Schleuning A. Evaluation of the new behind-the-ear audiant(TM) bone conductor(TM). *Semin Hear* 1992;**13**:325–30. Reason for exclusion: intervention.

Kunst SJ, Hol MK, Cremers CW, Mylanus EA. Bone-anchored hearing aid in patients with moderate mental retardation: impact and benefit assessment. *Otol Neurotol* 2007;**28**:793–7. Reasons for exclusion: comparator; design.

Liepert DR, DiToppa JC. The Nobelpharma auditory system bone-anchored hearing aid: the Edmonton experience. *J Otolaryngol* 1994;**23**:411–18. Reason for exclusion: design.

Negri S, Bernath O, Hausler R. Bone conduction implants: Xomed Audiant bone conductor vs. BAHA. *Ear Nose Throat J* 1997;**76**:394–6. Reasons for exclusion: outcomes; design.

Niehaus HH, Helms J, Muller J. Are implantable hearing devices really necessary? *Ear Nose Throat J* 1900;**74**:271–4. Reasons for exclusion: intervention; comparator; outcomes.

Priwin C, Granstrom G. A long-term evaluation of bone-anchored hearing aid (BAHA) in children. *Cochlear Implants Int* 2005;**6**:81–3. Reason for exclusion: design.

Ringdahl A, Israelsson B, Caprin L. Paired comparisons between the Classic 300 bone-anchored and conventional bone-conduction hearing aids in terms of sound quality and speech intelligibility. *Br J Audiol* 1995;**29**:299–307. Reason for exclusion: outcomes.

Roper A, Hobson J, Green K. Combined bone anchored hearing aid and mastoidectomy. *Mediterr J Otol* 2008;**4**:138–42. Reason for exclusion: paper not available from British Library (reports four cases only of BAHA and mastoidectomy combined).

Rosenbom T, Specht Petersen A. *Clinical study of a direct bone conductor*. Askim, Sweden: Oticon Medical; 2010. Reason for exclusion: comparator.

Snik AFM, Mylanus EAM, Cremers CWRJ. The bone-anchored hearing aid (BAHA) versus air-conduction hearing aids. In Portman M, Boudard P, Portmann D, editors. *Transplants and implants in otology – III*. New York, NY: Kugler Publications; 1996. pp. 309–12. Reason for exclusion: paper not available from British Library (conference proceedings dated 1996).

van der Pouw CT, Carlsson P, Cremers CW, Snik AF. A new more powerful bone-anchored hearing aid: first results. *Scand Audiol* 1998;**27**:179–82. Reason for exclusion: comparator.

Wade PS, Halik JJ, Chasin M. Bone conduction implants: transcutaneous vs. percutaneous. *Otolaryngol Head Neck Surg* 1992;**106**:68–74. Reasons for exclusion: comparator; design.

Appendix 4

List of relevant non-English-language publications identified by searches

Aguado BF, ntoli Candela CF. [Branemarck bone-anchored implants for the adaptation of conductive hearing aids.] [Spanish.] *Acta Otorrinolaringologica Espanola* 1990;**41**:169–72.

Belus JF, Sarabian A, Triglia JM, Zanaret M. [Bone anchored auditory prosthesis. Indications, clinical and audiometric results.] [French.] *Annales d Oto-Laryngologie et de Chirurgie Cervico-Faciale* 1996;**113**:79–85.

Bonding P, Jonsson MH, Salomon G. [Bone-anchored hearing aids. Preliminary results.] [Danish.] *Ugeskrift for Laeger* 1990;**152**:667–70.

Bonding P. [Permanent, percutaneous osseointegrated titanium implants. A review and preliminary results.] [Danish.] *Ugeskrift for Laeger* 1990;**152**:664–7.

Bonding P, Jonsson MH, Salomon G, Ahlgren P. [The bone-anchored hearing aid. Host-reaction and audiological effect.] [Danish.] *Ugeskrift for Laeger* 1993;**155**:1183–5.

Candela Cano FA, Aguado BF, Sada Garcia-Lomas J. [Branemark-type osteo-integrated implants for the adaptation of endosseous hearing aids.] [Spanish.] *Acta Otorrinolaringologica Espanola* 1990;**41**:61–4.

Cremers CWRJ, Snik AFM, Beynon AJ. [A hearing aid anchored in the cranial bone to amplify bone conduction.] [Dutch.] *Nederlands Tijdschrift voor Geneeskunde* 1991;**135**:468–71.

Federspil P, Kurt P, Koch A. [Bone-anchored epitheses and audioprostheses: 4 years' experience with the Branemark system in Germany.] [French.] *Revue de Laryngologie Otologie Rhinologie* 1992;**113**:431–7.

Federspil PA, Plinkert PK. [Bone-anchored hearing aids: always bilateral!.] [German.] *HNO* 2002;**50**:405.

Grunder I, Seidl RO, Ernst A, Todt I. [Relative value of BAHA testing for the postoperative audiological outcome.] [German.] *HNO* 2008;**56**:1020–4.

Hamann C, Manach Y, Roulleau P. [Bone anchored hearing aid. Results of bilateral applications.] [French.] *Revue de Laryngologie Otologie Rhinologie* 1991;**112**:297–300.

Healthcare Insurance Board.]Evaluation of bone-anchored hearing aids – primary research.] [Dutch.] URL: www.cvz.nl (accessed March 2008).

Hoelzl M, Caffer P, Jungk J, Scherer H, Schrom T. [The Ti-Epiplating system in bone anchored hearing aids.] [German.] *Laryngo-Rhino-Otologie* 2007;**86**:193–9.

Jankowski R, Pialoux R, Labaeye P, Simon C. [Bone anchored hearing aid (BAHA): clinical evaluation.] [French.] *Annales d Oto-Laryngologie et de Chirurgie Cervico-Faciale* 1998;**115**:315–20.

Kitamura K, Tokano H. Bone-Anchored Hearing Aid: BAHA. [Japanese.] *Oto-Rhino-Laryngology Tokyo* 2004;**47**:8–16.

Klaiber S, Weerda H. [BAHA (bone-anchored hearing aid) in bilateral external ear dysplasia and congenital ear atresia.] [German.] *HNO* 2002;**50**:949–59.

Kondoh K, Matsushiro N, Satoh T, Kuramasu T, Kubo T. [Audiological effect of bone-anchored hearing aid.] [Japanese.] *Nippon Jibiinkoka Gakkai Kaiho* 2005;**108**:1144–51.

Lyberg T, Tjellstrom A. [Craniofacial prostheses. Clinical application of titanium implants for retention of facial prostheses and bone-anchored hearing aids.] [Norwegian.] *Tidsskrift for Den Norske Laegeforening* 1988;**108**:2009–12.

Machida S, Shimakura Y, Okamoto M, Nonomura E. [Fitting of bone-conduction hearing aids for persons suffering from mixed hearing loss.] [Japanese.] *Audiology Japan* 1983;**26**:27–33.

Negri S, Bernath O, Hausler R. [Implantable bone conduction hearing aids: Audiant(TM) vs. BAHA(TM).] [German.] *Oto-Rhino-Laryngologia Nova* 1996;**6**:82–8.

Nystrand A. [Bone-anchored hearing aids and implants in the cochlea improve hearing.] [Swedish.] *Lakartidningen* 143;**88**:137–8.

Portmann D, Boudard P, Vdovytsya O. [Bone-anchored hearing aids BAHA: 10 years' experience.] [French.] *Revue de Stomatologie et de Chirurgie Maxillo-Faciale* 2001;**102**:274–7.

Portmann D, Bourdin M. [Bone anchored hearing aid: the Bordeaux experience.] [French.] *Revue de Laryngologie Otologie Rhinologie* 1995;**116**:299–300.

Portmann D, Dutkiewicz J, Boudard P. [The use of osseointegration of hearing aids.] [Polish.] *Otolaryngologia Polska* 1995;**49**:543–8.

Sanchez-Camon I, Lassaletta L, Castro A, Gavilan J. [Quality of life of patients with BAHA.] [Spanish.] *Acta Otorrinolaringologica Espanola* 2007;**58**:316–20.

Shrom T, Siegert R. [Problems with the BAHA abutment.] [German.] *Laryngo- Rhino- Otologie* 2008;**87**:764–7.

Schupbach J, Kompis M, Hausler R. [Bone anchored hearing aids (B.A.H.A.).] [German.] *Therapeutische Umschau* 2004;**61**:41–6.

Zhang Q, Gao X. [Bone anchored hearing aid.] [Chinese.] *Chin J Clin Rehabil* 2006;**10**:124–8.

Appendix 5

List of potentially eligible studies but lower level of evidence

Abramson M, Fay TH, Kelly JP, Wazen JJ, Liden G, Tjellstrom A. Clinical results with a percutaneous bone-anchored hearing aid. *Laryngoscope* 1989;**99**:707–10. Design: audiological comparison study.

Bance M, Abel SM, Papsin BC, Wade P, Vendramini J. A comparison of the audiometric performance of bone anchored hearing aids and air conduction hearing aids. *Otol Neurotol* 2002;**23**:912–19. Design: audiological comparison study.

Bonding P, Jonsson MH, Salomon G, Ahlgren P. The bone-anchored hearing aid. Osseointegration and audiological effect. *Acta Otolaryngol* 1992;**492**:42–5. Design: audiological comparison study.

Bonding P. Titanium implants for bone-anchored hearing aids – host reaction. *Acta Otolaryngol* 2000;**543**:105–7. Design: prospective case series.

Bosman AJ, Snik AF, Mylanus EA, Cremers CW. Fitting range of the BAHA Cordelle. *Int J Audiol* 2006;**45**:429–37. Design: prospective case series.

Browning GG, Gatehouse S. Estimation of the benefit of bone-anchored hearing aids. *Ann Otol Rhinol Laryngol* 1994;**103**:872–8. Design: audiological comparison study.

Carlsson P, Håkansson B, Rosenhall U, Tjellstrom A. A speech-to-noise ratio test with the bone-anchored hearing aid: a comparative study. *Otolaryngol Head Neck Surg* 1986;**94**:421–6. Design: audiological comparison study.

Cremers CW, Snik FM, Beynon AJ. Hearing with the bone-anchored hearing aid (BAHA, HC 200) compared to a conventional bone-conduction hearing aid. *Clin Otolaryngol* 1992;**17**:275–9. Design: audiological comparison study.

Cremers CWRJ, Snik AFM, Beynon AJ. The bone anchored hearing-aid versus the previous conventional bone conduction hearing-aid – a preliminary-report of a clinical-trial. In Charachan R, Garcaibanez E, editors. *Long-term results and indications in otology and otoneurosurgery*. Amsterdam: Kugler Publications; 1991. pp. 461–4. Design: audiological comparison study.

Håkansson B, Liden G, Tjellstrom A, Ringdahl A, Jacobsson M, Carlsson P, *et al.* Ten years of experience with the Swedish bone-anchored hearing system. *Ann Otol Rhinol Laryngol* 1990;**151**:1–16. Design: audiological comparison study.

Håkansson B, Tjellstrom A, Rosenhall U, Carlsson P. The bone-anchored hearing aid. Principal design and a psychoacoustical evaluation. *Acta Otolaryngol* 1985;**100**:229–39. Design: audiological comparison study.

Håkansson BE, Carlsson PU, Tjellstrom A, Liden G. The bone-anchored hearing aid: principal design and audiometric results. *Ear Nose Throat J* 1994;**73**:670–5. Design: audiological comparison study.

Hartland SH, Proops D. Bone anchored hearing aid wearers with significant sensorineural hearing losses (borderline candidates): patients' results and opinions. *J Laryngol Otol* 1996;**21**:41–6. Design: audiological comparison study.

Hickson L, Mackenzie D, Gordon J, Neall V, Wu D, Wu J. The outcomes of bone anchored hearing aid (BAHA) fitting in a paediatric cohort. *ANZJA* 2006;**28**:75–89. Design: audiological comparison study.

Hol MK, Snik AF, Mylanus EA, Cremers CW. Long-term results of bone-anchored hearing aid recipients who had previously used air-conduction hearing aids. *Arch Otolaryngol Head Neck Surg* 2005;**131**:321–5. Design: audiological comparison study.

Jacobsson M, Albrektsson T, Tjellstrom A. Tissue-integrated implants in children. *Int J Pediatr Otorhinolaryngol* 1992;**24**:235–43. Design: prospective case series.

Lindeman P, Tengstrand T. Clinical experience with the bone-anchored hearing aid. *Scand Audiol* 1987;**16**:37–41. Design: audiological comparison study.

Mylanus EA, Cremers CW. A one-stage surgical procedure for placement of percutaneous implants for the bone-anchored hearing aid. *J Laryngol Otol* 1994;**108**:1031–5. Design: prospective case series.

Mylanus EA, Snik AF, Jorritsma FF, Cremers CW. Audiologic results for the bone-anchored hearing aid HC220. *Ear Hear* 1994;**15**:87–92. Design: audiological comparison study.

Mylanus EA, Snik FM, Cremers CW, Jorritsma FF, Verschuure H. Audiological results of the bone-anchored hearing aid HC200: multicenter results. *Ann Otol Rhinol Laryngol* 1994;**103**:368–74. Design: audiological comparison study.

Mylanus EA, Beynon AJ, Snik AF, Cremers CW. Percutaneous titanium implantation in the skull for the bone-anchored hearing aid. *J Invest Surg* 1994;**7**:327–32. Design: audiological comparison study.

Portmann D, Boudard P, Herman D. Anatomical results with titanium implants in the mastoid region. *Ear Nose Throat J* 1997;**76**:231–4. Design: prospective case series.

Powell RH, Burrell SP, Cooper HR, Proops DW. The Birmingham bone anchored hearing aid programme: paediatric experience and results. *J Laryngol Otol* 1996;**21**:21–9. Design: audiological comparison study.

Priwin C, Stenfelt S, Edensvard A, Granstrom G, Tjellstrom A, Kansson H. Unilateral versus bilateral bone-anchored hearing aids (BAHAs). *Cochlear Implants Int* 2005;**6**:79–81. Design: audiological comparison study.

Soo G, Tong MC, Tsang WS, Wong TK, To KF, Leung SF, *et al.* The BAHA hearing system for hearing-impaired postirradiated nasopharyngeal cancer patients: a new indication. *Otol Neurotol* 2009;**30**:496–501. Design: prospective case series.

Stenfelt S, Håkansson B, Jonsson R, Granstrom G. A bone-anchored hearing aid for patients with pure sensorineural hearing impairment: a pilot study. *Scand Audiol* 2000;**29**:175–85. Design: audiological comparison study.

Appendix 6

Data extraction: BAHA versus BCHA

Reference and design	Intervention	Participants	Outcome measures
Béjar-Solar *et al.* 2000[77] Mexico *Design:* cohort (one group pre and post) *Study setting:* tertiary referral centre for patients of low socioeconomic status *Number of centres:* single centre *Funding:* grant from Hospital's Board of Patrons	1. Unaided (pre-surgery) 2. BCHA (evaluated pre-BAHA surgery) 3. BAHA Classic 300 (evaluated after 6 months) *Other interventions used:* none	*Indication for treatment:* inoperable bilateral congenital microtia atresia *Number of participants:* 11 *Sample attrition/dropout*: implant was rejected in one patient *Inclusion criteria for study entry:* BC PTA 45 dB HL or better with 100% speech discrimination; high resolution CT demonstrating inoperable bilateral congenital microtia atresia; age at least 5 years; current use of a conventional BCHA *Exclusion criteria:* sensineural hearing loss with a BC PTA < 45 dB HL; lack of hygiene facilities to properly clean the skin around the implant; insufficient score on the psychological evaluation (minimal standards for intelligence and family support); economic capability to purchase batteries (approximately cost US$1); accessibility to hospital for follow-up visits	*Primary and secondary outcome:* audiological benefit; complications; patient satisfaction (not validated); QoL;Coop/Dartmouth test (validated) *Method of assessing outcomes:* pre-operative audiologic evaluation: PTA (125–3000 Hz) in both air and BC; free-field PTA with and without BCHA; 100% speech audiometry discrimination in dB HL; 100% speech audiometry discrimination with background noise at 65 dB HL with BCHA Test adapted to their group, using sentences with a high degree of difficulty, using colloquial language common to Mexico City Patient, wearing BCHA, asked to pinpoint the position of natural vocal speech in an area with high background noise, at a distance of 3m Tests repeated with BAHA at 6 months' follow-up. PTA (125–3000 Hz) in both AC and BC measured for use as a control Subjective patient satisfaction questionnaire, no further details reported QoL psychological test evaluating emotional condition, impact on daily chores and social activities, social support and pain (Coop/Dartmouth test) Condition of skin evaluated on 0–4 scale (0 = normal, 1 = slight erythema, 2 = red and moist, 3 = red and moist with granulation tissue, 4 = infection leading to loss or removal of implant) *Length of follow-up:* states that follow-up was 24 months. Visits scheduled at 1, 2, 4, 6, 12, 18 and 24 months Audiological tests were scheduled at 6 months

Characteristics of participants

Age, years	5–17
Sex (M : F)	7 : 4
No. with mandibulofacial dysotosis (Treacher Collins syndrome)	4/11
Mean age at implantation, years	10

Results

Attained successful use of BAHA at 2-year follow-up	10/11

Audiologic results, dB HL	Before surgery (unaided)	After surgery (unaided)	Difference in threshold
AC PTA, RE	69	71	± 2
AC PTA, LE	69	68	± 1
BC PTA, RE	20	18	± 2
BC PTA, LE	14	15	± 1
Free-field PTA, dB HL (1.25–3.00 kHz)	64	63	± 1

Level at which right ear 100% speech audiometry discrimination was achieved	87	90	±3
Level at which left ear 100% speech audiometry discrimination was achieved	84	83	±1
	BCHA (before surgery)	**BAHA**	
Free-field PTA dB HL (1.25–3.00 kHz)	30	19	–11 (37% improvement)
Free-field 100% speech audiometry, background noise at 65 dB	62	48	–14 (23% improvement)
	BCHA (before surgery)	**BAHA**	
Accurate directional identification of location of a sound source (% of cases)	0	80%	
Patient satisfaction questionnaire	Data not presented. States that compared with the BCHA 'all patients preferred the BAHA, believed there had been an excellent improvement in aesthetic appearance, and would choose to have it done again (scale: excellent, good, fair, poor, very poor)'		
QoL	Data not presented. States that 'tests results uniformly showed the response "hardly could have done better" (options: hardly could have done better, pretty good, indifferent, pretty bad, hardly could have done worse). Physical and emotional condition was reported as very improved. Positive family support was confirmed in all cases'		

Adverse effects

Unable to obtain osseointegration (following impact to mastoid area 24 hours after discharge from first stage)	1/11
Major complications	0/11

Types of skin reactions, n *of observations (%)*

■ No irritation	71/82 (87)
■ Slight erythema	7/82 (9)
■ Erythema and moisture	3/82 (4)
■ Red and moist with granulation tissue	1/82 (1)
■ Infection leading to loss of implant	0
■ Total number of observations	82 (71 at scheduled visits, 11 at unscheduled visits)

Methodological comments

- Allocation to treatment groups: one group. All patients evaluated with BCHA before BAHA surgery
- Blinding: NR
- Comparability of treatment groups: there were no differences between PTA BC or AC thresholds before and after implantation
- Method of data analysis: statistical analysis not presented
- Sample size/power calculation: NR
- Attrition/dropout: one patient experienced lack of ossiointegration leading to rejection of implant. It is not clear if this patient is included in the assessment of skin reactions or the pre-surgery audiological results

General comments

- Generalisability: patients were carefully selected (psychological assessment, establish realistic expectations of the BAHA established, evaluated by social worker), all information was reviewed by the authors and the decision had to be unanimous for patients to be accepted into study. Participants were from Mexico and of low socioeconomic status
- Outcome measures: outcome measures were relevant and measured appropriately. Limited details given about the patient satisfaction questionnaire and QoL psychological test, and no data presented
- Conflict of interests: paper states that 'Nobel Biocare AB did not sponsor this research, and does not have a commercial interest with any of the authors'

F, female; L, left; M, male; NR, not reported; R, right.

Quality assessment for primary studies

A. Selection bias					
1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100%	60–79%	< 60%	Not applicable	Can't tell x
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT Controlled clinical trial Cohort analytic (two group pre and post) Case–control Cohort [one group pre and post (before and after)] Interrupted time series Other – specify Can't tell			x reviewer's opinion	
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below.					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?' ■ If there is one group of participants in the study: 'are potential confounding variables reported?'	Yes	No x	Can't tell		
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropouts				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No x	Can't tell	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No x	Can't tell	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes	No x	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Appendix 7

Data extraction: BAHA versus ACHA

Reference and design	Intervention	Participants	Outcome measures
Burrell *et al.* 1996[83] UK *Design:* cohort pre–post *Study setting:* secondary care *Number of centres:* single centre *Funding:* NR	1. BAHA (model not stated) 2. ACHA (states 'old aid' in paper. Information from author) 3. Unaided *Other interventions used:* NR	*Indication for treatment:* otosclerosis *Number of participants:* 1. 32 assessed over 5-year period 2. 19 suitable for BAHA 3. 10 fitted with BAHAs (nine waiting for surgery) 4. data available for nine *Sample attrition/dropout:* missing data for two patients for one outcome *Inclusion criteria for study entry:* audiological criteria: average BC thresholds (0.5–4 kHz) < 40 dB HL (ear level BAHA); average BC thresholds < 60 dB HL (body-worn Superbass); speech discrimination > 60% (AB wordlists via headphones); realistic expectations; good support. Final decision to proceed with BAHA taken by a multidisciplinary team including ENT surgeons, audiologists and a specialist speech therapist	*Primary and secondary outcomes:* audiological performance; free-field warble-tone audiometry; free-field speech audiometry; subjective evaluation of sound quality and comfort *Method of assessing outcomes:* pre-operative audiologic evaluation unaided and with any existing hearing aids. Free-field speech audiometry using Boothroyd list, aided and unaided Post-operative evaluation unaided and with BAHA (duration not stated) Sound quality and comfort rated pre-operatively for their old aid, and post-operatively for BAHA, on a scale of 1–10 (sound quality: 1 = distorted, 10 = clear and natural; comfort: 1 = so uncomfortable the aid cannot be worn, 10 = so comfortable you are unaware of its presence) *Length of follow-up:* NR

Characteristics of participants

Patients suitable for BAHA (n = 19)	
Age, years (mean)	45.7, range 25.0–76.0
Sex (M : F)	4 : 15
Average BC thresholds (0.5–4.0 kHz)	24 dB HL
Patients unsuitable for BAHA (n = 13)	
Reasons for being unsuitable:	Hearing too bad = 8 Hearing too good = 1 (unilateral otosclerois) Declined = 4

Results (n = 9)

Outcomes	Unaided	ACHA	BAHA
Average free-field warble-tone thresholds (0.5–4.0 kHz), mean (SD), range[a]	49.4 (11.9), 40–78 dB(A)	33.0 (5.4), 28–40 dB(A)	30.6 (8.1), 22–43 dB(A)
Comments: all patients gained improvements in threshold using their ACHA compared with unaided. BAHA results were comparable with the ACHA, but 'significantly' better in only one case			
Free-field speech discrimination at 63 dB(A), mean (SD), range[a]	74.0 (19.5), 50–98 dB(A)[b]	91.6 (14.7), 60–100 dB(A)	84.0 (22.3), 30–100 dB(A)
Comments: improvements were observed from unaided to ACHA, however, comparisons between ACHA and BAHA showed no improvement			

Subjective assessment	ACHA	BAHA	*p*–value
Patients' rating of sound quality, mean (SD), range[a]	4.6 (2.1), 2–8	7.9 (2.4), 2–10	
Patients' rating of comfort, mean (SD), range[a]	4.1 (2.7), 1–10	9.4 (1.0), 7–10	
Cosmetic preference	BAHA: 8/9 (89%)		
	No difference: 1/9 (11%)		
Patient preference in background noise	BAHA: 55%		
	ACHA: 11%		
	No difference: 34%		

Methodological comments

Allocation to treatment groups: one group tested pre- and post-operatively, aided and unaided

Blinding: none

- Comparability of treatment groups: no baseline characteristics for the nine included participants
- Method of data analysis: statistical analysis not undertaken. Individual patient data presented graphically. Data estimated from figure, and means and SDs calculated by reviewer
- Sample size/power calculation: NR
- Attrition/dropout: data missing for two patients for speech discrimination, but reasons not given. Of 10 patients fitted with a BAHA, a full set of post-operative data was available for nine patients; reasons not given why data not available for one of the patients

General comments

- Generalisability: patients with otosclerosis who had declined stapedectomy, or stapedectomy was not indicated or had experienced previous failed surgery
- Outcome measures: audiologic measures appropriate, but timing of assessments not given (i.e. duration of use with ACHA and BAHA NR). Subjective assessments were made using a questionnaire that was not a validated measure. For patient preference, it is not clear with the questionnaire was administered pre-operatively as well as post-operatively. It is not clear whether the unaided data presented were assessed pre-operatively or post-operatively (as it appears the unaided condition was assessed on both occasions)
- Conflict of interests: NR
- Other: no description of 'old aid' is given in paper. Information requested and received from author. Model of BAHA used NR

F, female; M, male; NR, not reported.
a Individual patient data estimated from figure, and means and SDs calculated by reviewer.
b Data missing for two patients.

Quality assessment for primary studies

A. Selection bias

1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	<60%	Not applicable	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate* x	*Weak*		

B. Study design

1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT Controlled clinical trial Cohort analytic (two group pre and post) Case-control Cohort [one group pre and post (before and after)] Interrupted time series Other – specify Can't tell			x reviewer's opinion	
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below.					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

C. Confounders					
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?'	Yes	No x	Can't tell		
■ If there is one group of participants in the study: 'are potential confounding variables reported?'					
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
E. Data collection methods					
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell		
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell		
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*		
F. Withdrawals and dropouts					
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No x	Can't tell		
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100% x	60–79%	< 60%	Can't tell	
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
G. Intervention integrity					
1. Was the consistency of the intervention measured?	Yes	No x	Can't tell		
H. Analysis					
1. Are the statistical methods appropriate for the study design?	Yes	No x	Can't tell		
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell		Not applicable
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**		

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and design	Intervention	Participants	Outcome measures
Flynn *et al.* 2009[76] Sweden *Design:* audiology comparison study *Study setting:* unclear *Number of centres:* unclear *Funding:* Cochlear Bone Anchored Solutions	1. BAHA Intenso 2. ACHA Oticon Sumo DM (digital superpower hearing aid)	*Indication for treatment:* mixed hearing loss *Number of participants:* 10 *Sample attrition/dropout:* NR *Inclusion/exclusion criteria for study entry:* had worn BAHA for at least 1 year at time of study and had previous experience of ACHA. Mixed hearing loss defined as an average sensorineural component > 25 dB HL in addition to an air-bone gap ≥ 30 dB	*Primary and secondary outcomes:* aided warble-tone free-field thresholds Speech understanding in noise (speech-to-noise ratio) *Method of assessing outcomes:* aided free-field thresholds (warble tones) measured as described by Morgan *et al.*[144] Speech understanding in noise measured using adaptive procedures from Swedish version of Hearing In Noise Test (Hällgren *et al.* 2006[94]). Two loud speakers used at 1 m from subject. Speech presented from 0° and noise from 180° *Length of follow-up:* appears that assessments were undertaken at same session

Characteristics of participants

Mean age, years (range)	59 (32–75)
Sex (M : F)	5 : 5
Unaided PTA AC thresholds (0.5, 1.0 and 2.0 kHz), mean	77 (range 55–80) dB HL
Unaided PTA BC thresholds (0.5, 1.0 and 2.0 kHz), mean	41 (range 25–66) dB HL

Results

Outcomes	BAHA Intenso ($n = 10$)	ACHA ($n = 10$)	p-value
Average aided thresholds (dB SPL)[a]			
250 Hz	47	39	
500 Hz	39[b]	42	
1 kHz	30[b]	37	
2 kHz	31[b]	43	
3 kHz	39[b]	46	
4 kHz	41[b]	50	
6 kHz	53	75	
8 k Hz	55	68	
Speech-to-noise ratio, dB (SNR)	**0.88**	**3.44**	**Difference 2.56**

Methodological comments

- Allocation to treatment groups: not clear how participants were recruited. All participants tested with BAHA and ACHA, probably at same session
- Blinding: NR
- Comparability of treatment groups: not applicable
- Method of data analysis: p-value reported for aided thresholds, but no details given on method of analysis
- Sample size/power calculation: NR
- Attrition/dropout: NR, not clear how many people invited to participated in study

General comments

- Generalisability: participants with mixed hearing loss who had at least 1 year's experience with BAHAs, and had previous experience with ACHAs
- Outcome measures: appear valid and reliable
- Inter-centre variability: NR
- Conflict of interests: authors are employees of Cochlear Bone Anchored Solutions

F, female; M, male; NR, not reported.
a Data estimated from figure by reviewer.
b States that clinically, audibility improved by 5–15 dB at these frequencies (although the figure does not seem to show that for 500 Hz).
At 250 Hz, the audibility was better with ACHA.
States that BAHA provided a significant improvement ($p < 0.01$) in audibility, as measured though the sound field audiogram.
States that the improvement in speech understanding of 2.56 dB (SNR) with BAHA is 'large and clinically significant'.

Quality assessment for primary studies

A. Selection bias					
1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely	Not likely	Can't tell x	
2. What percentage of selected individuals agreed to participate?	80–100%	60–79%	< 60%	Not applicable	Can't tell x
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT				
	Controlled clinical trial				
	Cohort analytic (two group pre and post)				
	Case–control				
	Cohort [one group pre and post (before and after)]				
	Interrupted time series				
	Other – specify: audiology comparison study			x reviewer's opinion	
	Can't tell				
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below.					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?' ■ If there is one group of participants in the study: 'are potential confounding variables reported?'	Yes	No x	Can't tell		
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Data collection methods					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropouts				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No x	Can't tell	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes	No	Can't tell x	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No	Can't tell x	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and design	Intervention	Participants	Outcome measures
Mylanus *et al.* 1998[84] Netherlands *Design*: cohort, pre and post *Study setting*: outpatient (secondary care) *Number of centres*: one *Funding*: not stated	1. BAHA monaurally BAHA model NR 2. ACHA (tested pre-operatively) *Other interventions used*: none	*Indication for treatment*: bilateral conductive or mixed hearing loss and chronic ear problems. Blockage of the ear canal with the ear mould of ACHA had caused or exacerbated chronic 'otitis' (assume otitis media) *Number of participants*: 34 *Sample attrition/dropout*: no attrition reported, one participant did not complete the questionnaire *Inclusion/exclusion criteria for study entry*: not stated explicitly. Previously fitted ACHA	*Primary outcomes*: not defined as primary or secondary outcomes; free-field aided thresholds, speech recognition in quiet, speech recognition in noise, MPS, speech-to-noise ratio, subjective questionnaire *Secondary outcomes*: also functional gain (not data extracted) *Method of assessing outcomes*: before testing aids checked for normal functioning and adjusted to the patients preferred setting The MPS was calculated from the free-field speech recognition-intensity function (speech audiogram) Speech-to-noise ratio was determined according to criteria of Plomp and Mimpen[89] at a fixed noise level of 65 dB The subjective questionnaire concerned ear infection, frequency of visits to outpatient clinic, 'handling and feedback', also speech recognition in quiet and noise, quality of sound, cosmetic appearance, patient preference advantages and disadvantages. Sent after BAHA use between 9 months and 7 years (mean 32 months) and compared with a questionnaire sent out after 5 months of BAHA fitting (this aspect not data extracted) *Length of follow-up*: 4–6 weeks after fitting of BAHA (for objective outcomes)

Characteristics of participants

	BAHA	*p*-value
Age, years	Average age 48 years, range 26–72 years	
Sex (M : F)	12 : 22	
PTA for AC at 0.5, 1.0, 2.0 and 4.0 kHz[a]	12 with linear, medium-power ACHA: between 25 and 65 dB HL 22 with linear, high-power ACHA: between 40 and 90 dB HL Overall group mean: 60 dB HL (range 25–90 dB HL)	
PTA for BC at 0.5, 1.0, 2.0 and 4.0 kHz[a]	Mean: 26 dB HL (range 6–46 dB HL)	
Air-bone gap[a]	Mean 34 dB HL (range 11–54 dB HL); 15 (44%) had one totally deaf ear	

Results

Outcomes	BAHA (4–6 weeks post fitting)	ACHA (pre-op)	Difference and *p*-value
Mean free-field threshold, dB HL (SD)	0.25 kHz: 39	0.25 kHz: 40	$p = NS$
	0.50 kHz: 36	0.50 kHz: 36	$p = NS$
	1.00 kHz: 22 (8.3)	1.00 kHz: 28	$p < 0.01$
	2.00 kHz: 25	2.00 kHz: 22 (11.9)	$p = NS$
	4.00 kHz: 33	4.00 kHz: 37	$p = NS$
	8.00 kHz: 43 (22.3)	8.00 kHz: 55 (21.3)	$p < 0.001$
Comments: estimated from figure by reviewer			
SDs for each frequency NR. States that the SD varied between 11.9 dB at 2 kHz and 21.3 dB at 8 kHz for ACHA, and between 8.3 dB at 1 kHz and 22.3 dB at 8 kHz for BAHA			
MPS (mean ± SD)	Data NR	Data NR	1.0% ± 5.4%, $p = NS$
Comments: states 16 participants obtained an MPS of 100% with both types of hearing aid. Individual participant data of improvement in MPS by air-bone gap reported in a figure but not data extracted			
Speech-to-noise ratio improvement	Data NR	Data NR	1.1 ± 2.1 dB, $p < 0.01$
Comments: when a 1.4 dB criterion for significance was used on an individual basis, the speech-on-noise ratio with the BAHA improved significantly in 15 patients, it did not change in 14 patients and it deteriorated significantly in five patients			
Individual participant data presented for speech-to-noise ratio by air-bone gap in a figure but not data extracted. A significant correlation was found between the change in speech-to-noise ratio and the width of the air-bone gap ($r = 0.59$, $p < 0.01$)			
Speech recognition in quiet			
Comments: despite this being an outcome measure no mention is made of this in the results			
Preference of device based on ($n = 33$)	**BAHA (%)**	**ACHA (%)**	**No preference (%)**
Ear infections	32 (97)	0 (0)	1 (3)
Speech-in-quiet	20 (61)	5 (15)	8 (24)
Speech-in-noise	10 (30)	9 (27)	14 (42)
Quality of sound	20 (61)	6 (19)	7 (21)
Visibility	15 (45)	8 (24)	10 (30)
Handling	13 (39)	5 (15)	15 (45)
Feedback	25 (76)	4 (12)	4 (12)
ENT visits	21 (64)	4 (12)	8 (24)
Overall preference	27 (82)	5 (15)	1 (3)
Comments: all estimated from figure by reviewer. One participant did not complete the questionnaire. No statistical significance testing undertaken. Also reports most important advantage and disadvantage of BAHA but data not extracted			

Adverse effects

Comments: states surgery was uneventful in all patients. Two stopped using their BAHA after 3 months and 2.5 years respectively, owing to pain – no explanation for this found

Methodological comments

- Allocation to treatment groups: consecutive BAHA users reported
- Blinding: NR
- Comparability of treatment groups: not applicable
- Method of data analysis: states that the speech-to-noise ratio is statistically significant if the change exceeds 2 dB (2 × SD), where the SD of the test is known to be 1 dB. In this study a difference in excess of 1.4 dB between two speech-to-noise ratios was regarded as significant as the values used were the average values from two successive measurements, it is unclear how valid this is. *t*-tests applied to differences between ACHA and BAHA with statistical significance set at 0.05
- Sample size/power calculation: NR
- Attrition/dropout: unclear, one participant did not complete the questionnaire

General comments

- Generalisability: previous users of ACHAs, who had been advised to discontinue use. All are described as 'BAHA users'. Also states all used their BAHAs daily, although two stopped using their BAHA after 3 months and 2.5 years respectively, owing to pain. Difficult to establish if this is before or after the period described in the study. No detail on the type of BAHA used was reported
- Outcome measures: valid and appropriate measures except subjective questionnaire which was not validated and hence data need to be interpreted cautiously. Questions relating to previous aid are subject to recall bias
- Inter-centre variability: not applicable
- Conflict of interests: NR

f, female; m, male; MPS, maximum phoneme score; NR, not reported; NS, not stated.
a Data refer to the ear ipsilateral to the side of implantation, always ear with best cochlear reserve.

Quality assessment for primary studies

A. Selection bias

1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	< 60%	Not applicable	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate* x	*Weak*		

B. Study design

1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT			
	Controlled clinical trial			
	Cohort analytic (two group pre and post)			
	Case–control			
	Cohort [one group pre and post (before and after)]			x reviewer's opinion
	Interrupted time series			
	Other – specify			
	Can't tell			
2. Was the study described as randomised?	Yes	No x		
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below.				
3. If answer was yes, was the method of randomisation described?	Yes	No		
4. If answer was yes, was the method appropriate?	Yes	No		
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	

C. Confounders				
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?'	Yes	No x	Can't tell	
■ If there is one group of participants in the study: 'are potential confounding variables reported?				
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
D. Blinding				
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell	
2. Were the study participants aware of the research question?	Yes x	No	Can't tell	
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropout				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No	Can't tell x	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes x	No	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	
Global rating for study[a]	**Strong**	**Moderate**	**Weak**	
(Overall methodological strength of study – based on sections A–F)			**x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Appendix 8

Data extraction: BAHA versus BCHA/ACHA

Reference and design	Intervention	Participants	Outcome measures
Cooper *et al.* 1996[78] UK *Design*: cohort (one group pre and post) *Study setting*: secondary care *Number of centres*: single *Funding*: NR	1. Previous aid: AC/BC 2. BAHA *BAHA model*: Nobel Biocare HC200/300/220 *Other interventions used*: candidates discuss surgical procedure, pros/cons of BAHAs and other options. Patients must meet another BAHA patient in order to have realistic expectations If existing aid was old or inadequate, a new appropriate aid/s was fitted prior to testing, so testing was against previous optimal aiding	*Indication for treatment*: adults with bilateral hearing loss *Number of participants*: 68 Subgroups (aetiology and previous aid): 1. CSOM/ACHA, $n=24$ 2. CSOM/BCHA, $n=19$ 3. CON/ACHA, $n=9$ 4. CON/BCHA, $n=16$ *Sample attrition/dropout*: 68/106 successfully follow up *Inclusion/exclusion criteria for study entry*: minimum age 17 years. Audiological criteria: 1. average BC thresholds (0.5–4 kHz) < 40 dB HL (ear level), < 60 dB HL (body-worn) 2. speech discrimination score > 60% 3. realistic expectations 4. reasonable social support. Final decision about suitability for BAHA made by multidisciplinary team	*Outcomes*: PTA, free-field speech results, free-field warble-tone threshold and questionnaire *Method of assessing outcomes*: PTAs are calculated from thresholds at 500, 1000, 2000 and 4000 Hz. Free-field speech results (%) discrimination at 63 dB(A) and obtained under three conditions (without aid, with existing aid, with BAHA) and frequencies are the same as for PTA Average free-field warble-tone threshold at same frequencies and conditions Questionnaire: 11 questions on usage and satisfaction, scored pre- and post-BAHA fitting ■ Question 7: included seven questions, but only the three identified as being most important to patients when using their BAHA were reported [(1) listening to radio or TV; (2) listening in quiet surroundings with friends and family at home; (3) listening in noisy surroundings with a group of people] and scored from 1 to 5 (very satisfactory to very unsatisfactory). Number of patients that showed a worse score, improved by one point, and improved by more than one point reported ■ Question 9 (feelings about old aid and BAHA): results were scored by allocating a positive point to any positive comments and a negative point to any negative comments made by patients. An overall score was obtained for the old aid and for the BAHA. BAHA performance was compared with the old aid by counting the number in each group showing a worse, same or better score on this question ■ Question 10: patients rated their feelings regarding sound quality of their old aid and the BAHA against 12 descriptions of sound presented to them. Patients ticked the best descriptor describing their experience. Descriptions one and three were positive sound quality attributes and the remainder negative quality attributes ■ Overall satisfaction with the BAHA compared with the previous aid was scored worse, same or better by patients *Length of follow-up*: pre-op assessment and 6 months post-BAHA fitting assessment

Characteristics of participants[a]

	CSOM/ACHA, $n=24$	CSOM/BCHA, $n=19$	CON/ACHA, $n=9$	CON/BCHA, $n=16$	*p*-value
Age, mean years	~58	~61	~30	~24	$p<0.01$[b]
Mean PTA threshold, AC 500–4000 Hz (db HL)	~58	~65	~70	~60	$p>0.05$[b]
Mean PTA threshold, BC 500–4000 Hz (db HL)	~24	~30	~20	~13	$p<0.01$[b]

Air-bone gap (500–4000 Hz) (db HL)	~33		~32	~52		~49	

CON overall group mean for BC threshold = 17.2 dB HL – just inside the normal range, both CSOM groups are outside the normal range

Results

Questionnaire (BAHA compared with old aid):	**CSOM/ACHA, *n* = 24**			**CSOM/BCHA, *n* = 19**			***p*-value**
	Worst	**Same**	**Better**	**Worst**	**Same**	**Better**	
Hearing in quiet, *n*	3	9	9	0	7	9	CSOM/BCHA $p<0.01$[c]
Hearing in noise, *n*	2	5	12	1	4	11	CSOM/ACHA $p<0.01$,[c] CSOM/BCHA $p<0.01$[c]
Hearing TV/radio, *n*	4	9	9	1	4	11	CSOM/BCHA $p<0.01$[c]
Feelings about BAHAs, *n*	3	3	15	1	5	10	CSOM/ACHA $p<0.01$,[c] CSOM/BCHA $p<0.01$[c]
Overall satisfaction, *n*	5	5	10	2	3	12	CSOM/ACHA p = NS, CSOM/BCHA $p<0.01$[c]
Questionnaire (BAHA compared with old aid):	**CON/ACHA, *n* = 9**			**CON/BCHA, *n* = 16**			***p*-value**
	Worst	**Same**	**Better**	**Worst**	**Same**	**Better**	
Hearing in quiet, *n*	0	3	3	0	6	6	CON/BCHA $p<0.05$[c]
Hearing in noise, *n*	1	0	5	3	4	5	
Hearing TV/radio, *n*	0	1	5	0	5	7	CON/ACHA $p<0.01$,[c] CON/BCHA $p<0.05$[c]
Feelings about BAHAs, *n*	0	0	8	2	1	9	CON/ACHA $p<0.05$,[c] CON/BCHA $p<0.01$[c]
Overall satisfaction, *n*	0	2	5	0	5	9	CON/ACHA $p<0.05$,[c] CON/BCHA $p<0.01$[c]
Questionnaire: sound quality (BAHA compared with old aid),% of patients with:	**Old aid**			**BAHA**			***p*-value**
Positive responses	44			67			
Negative responses	63			50			

Comments: 95.5% of patients used BAHA for >8 hours a day, 89.7% of these reporting sufficiently amplified sound

Hearing measures	**CSOM/ACHA, *n* = 24**			**CSOM/BCHA, *n* = 19**			***p*-value**
	No aid	**Old aid**	**BAHA**	**No aid**	**Old aid**	**BAHA**	
Mean free-field warble-tone thresholds [dB(A), 500–4000 Hz]	~60	~40	~33	~63	~42	~35	CSOM/ACHA $p<0.01$,[d] CSOM/BCHA $p<0.01$[d]
Mean free-field speech discrimination score (at 63 dB),% correct	~19	~69	~72	~17	~65	~72	CSOM/ACHA p = NS,[e] CSOM/BCHA p = NS[e]
	Worst	**Same**	**Better**	**Worst**	**Same**	**Better**	
Mean free-field warble-tone threshold (BAHA compared with previous aid), number of patients	~6	0	~18	~1	~1	~15	
Speech discrimination scores at 63 dB (BAHA compared with old aid), number of patients	~12	~2	~9	~5	~1	~12	
	CON/ACHA, *n* = 9			**CON/BCHA, *n* = 16**			***p*-value**
	No aid	**Old aid**	**BAHA**	**No aid**	**Old aid**	**BAHA**	
Mean free-field warble-tone thresholds, dB (500–4000 Hz)	~68	~41	~28	~62	~31	~26	CON/ACHA $p<0.01$,[d] CON/BCHA $p<0.01$[d]
Mean free-field speech discrimination score (at 63 dB),% correct	~17	~57	~82	~3	~86	~85	CON/ACHA $p<0.05$,[f] CON/BCHA p = NS[f]

	Worst	Same	Better	Worst	Same	Better
Mean free-field warble-tone threshold (BAHA compared with previous aid), number of patients	0	0	~9	~3	0	~11
Speech discrimination scores at 63 dB (BAHA compared with old aid), number of patients	~0	~3	~5	~3	~5	~5

Methodological comments

- Allocation to treatment groups: patients divided into four subgroups based on aetiology and previous hearing aid, pre-op data with previous aid compared with post BAHA fitting
- Blinding: none
- Comparability of treatment groups: CON groups significantly younger than CSOM group, with a mean age of approximately half
- Method of data analysis: students unpaired *t*-test between CSOM groups and CON groups for age and PTA thresholds. Students paired *t*-test for free-field warble-tone thresholds and free-field speech discrimination scores (at 63 dB). For each group, results of BAHAs were compared with old aid. Questionnaire was analysed using sign test for paired samples, with each group separately considered. Owing to low number of patients, the better +1 and better >1 scores were combined
- Sample size/power calculation: NR
- Attrition/dropout: of 106 patients wearing BAHAs, 68 were followed up. The data for number for patients whose warble-tone thresholds/speech discrimination scores were worse, same or better with BAHA appear to have some missing patients (CSOM/BCHA = 11%; CON/BCHA = 12%). There also appear to be missing data for the questionnaire

General comments

- Generalisability: adults with bilateral conductive hearing loss from CSOM or congenital causes
- Outcome measures: outcomes appear appropriate, but no SD etc. has been reported. It is unclear if the questionnaire used has been validated. Out of 15 questions in the questionnaire, only 1–11 were included, as the others were related to hardware usage and views on service provision. However, only questions 7 (3 out of 7 questions), 9 and 10, plus overall satisfaction, were actually reported in the results section. Authors state that data were gathered at 1, 6 and 12 months post-fitting of BAHA and annually thereafter. The 6-month data set was 'chosen' for analyses because 'patients had achieved the main benefits with the BAHA by this stage'. Data were mostly supplied in graph format only and had to be calculated by the researcher
- Inter-centre variability: not applicable
- Conflict of interests: NR

CON, congenital hearing loss; CSOM, chronic suppurative otitis media; NR, not reported; NS, not significant.
a Data estimated from figure by reviewer for patient characteristics, mean free-field warble-tone thresholds, mean free-field speech discrimination scores, reported hearing in quiet, noise or with TV/radio and for overall satisfaction with BAHA.
b Between CSOM and CON group.
c Significant improvement in hearing with BAHA compared with old aid.
d Results significantly better with BAHA than with the old aid, average improvement approximately 10 dB.
e The differences in speech discrimination scores between the old aid and BAHA were NS.
f The differences in speech discrimination scores between the old aid and BAHA were significantly improved with the BAHA for the CON/ACHA group, but not the CON/BCHA group.

Quality assessment for primary studies

A. Selection bias

1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100%	60–79%	<60%	Not applicable	Can't tell x
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT				
	Controlled clinical trial				
	Cohort analytic (two group pre and post)				
	Case–control				
	Cohort [one group pre and post (before and after)]			x reviewer's opinion	
	Interrupted time series				
	Other – specify				
	Can't tell				
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below.					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
Two groups: are confounders reported AND controlled for in the analysis? OR if one group: are potential confounding variables reported?	Yes	No x	Can't tell		Not applicable
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		*Not applicable*
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
E. Data collection methods					
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell		
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell		
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*		
F. Withdrawals and dropouts					
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No x	Can't tell		
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79% x	< 60%	Can't tell	
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes x	No	Can't tell	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes x	No	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	N/A
Global rating for study[a]	**Strong**	**Moderate**	**Weak**	
(Overall methodological strength of study – based on sections A–F)			**x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and Design	Intervention	Participants	Outcome measures
Hol *et al.* 2004[82] Netherlands *Design*: cohort pre and post *Study setting*: otorhino-laryngology department *Number of centres*: one *Funding*: NR	1. Previous aid (ACHA/BCHA) 2. BAHA Classic (51), BAHA Cordelle (5)	*Indication for treatment*: acquired conductive or mixed hearing loss *Number of participants*: $n = 56$ (ACHA, $n = 36$; BCHA, $n = 20$) *Sample attrition/dropout*: NR *Inclusion/exclusion criteria for study entry*: consecutive adult patients with acquired conductive or mixed hearing loss and listed for BAHA surgery	*Primary outcomes*: quality of life *Secondary outcomes*: number of hours of daily BAHA use; number of visits to otolaryngologist due to otorrhoea or skin irritations; frequency of episodes of otorrhoea; prevalence of skin irritations *Method of assessing outcomes*: SF-36; EQ-5D: 5 domains (1 = no problems to 3 = severe problems); utility index (0 = worse than death to 1 = perfect health); EQ-5D visual analogue scale (0 = worst imaginable state of health to 100 = best imaginable state of health); HHDI Note: data for whole group also reported but not extracted *Length of follow-up*: questionnaires completed with previous aid (ACHA or BCHA) before surgery and after 6 months experience with BAHA

Characteristics of participants

	Total ($n = 56$)	ACHA ($n = 36$)	BCHA ($n = 20$)
Age, years, mean (range)	52.9 (24–82)	47.9 (24–73)	62.0 (42–82)
Sex,% male	39	33	45
Hearing loss at 0.5, 1.0 and 2.0 kHz, dB HL, mean (range):			
AC	68.1 (30–107)	63.2 (30–103)	76.5 (40–107)
BC	31.8 (9–63)	26.8 (9–51)	43.4 (17–63)
Air-bone-gap, dB, mean (range)	36.3 (13–60)	36.4 (16–60)	36.1 (13–53)

Results: previous aid ACHA

SF-36 mean (SD)	ACHA ($n = 36$)	BAHA ($n = 36$)	Mean difference	Effect size
Physical functioning	80.3 (21.8)	79.8 (22.4)	–0.5 (p = NS)	0.02
Role limitations (physical)	71.5 (39.7)	68.9 (40.5)	–2.6 (p = NS)	0.06
Role limitations (emotional)	76.2 (40.1)	73.2 (38.1)	–3.0 (p = NS)	0.07
Vitality	60.4 (20.0)	59.9 (19.9)	–0.5 (p = NS)	0.02
Mental health	62.4 (18.0)	67.9 (21.3)	5.5 (p = NS)	–0.28
Social functioning	69.8 (28.3)	75.0 (27.8)	5.2 (p = NS)	–0.19

Pain	74.7 (25.2)	79.2 (25.0)	4.5 (p=NS)	–0.18
General health	63.2 (21.4)	63.6 (21.2)	–0.4 (p=NS)	–0.18
No statistically significant changes in any domain (better functioning leads to a higher score on a specific item). Mental health improved but not statistically significantly and effect size was small (–0.28)				
EQ-5D mean (SD)	**ACHA (n=36)**	**BAHA (n=36)**	**Mean difference**	**Effect size**
5 domains (score 1–3):				
Mobility	1.29 (0.46)	1.31 (0.47)	0.02	–0.04
Self-care	1.03 (0.17)	1.03 (0.17)	0.00	0.0
Usual activities	1.47 (0.66)	1.44 (0.50)	–0.03 (p>0.05)	0.05
Pain/discomfort	1.49 (0.51)	1.47 (0.51)	–0.02 (p>0.05)	0.04
Anxiety/depression	1.26 (0.44)	1.42 (0.60)	0.16 (p<0.01)	–0.30
Utility (score 0–1)	0.78 (0.17)	0.77 (0.17)	–0.01	0.06
Visual analogue scale (score 0–100)	76.1 (14.1)	73.4 (17.1)	–2.7	0.17
Anxiety/depression increased (p<0.01), but the clinical effect was small (–0.3)				
HHDI mean (SD)	**ACHA (n=36)**	**BAHA (n=36)**	**Mean difference**	**Effect size**
Disability	25.8 (6.5)	20.9 (6.2)	–5.0 (p<0.01)	0.79
Handicap	25.0 (5.9)	19.6 (6.7)	–5.4 (p<0.01)	0.86
Statistically significant improvements in disability and handicap, large clinical impact				
	ACHA (n=36)	**BAHA (n=36)**		
Number of otolaryngology visits over preceding 6 months for draining ears, mean (SD)	32 patients, 12.7 (10.5) visits, range 0–30	33 patients, 3.3 (4.8), range 0–25		
Patient preference in regard to:				
Otorrhoea	1 (3%)	17 (47%)		
Skin irritation	6 (17%)	14 (39%)		
Proportion using aid >8 hours per day	78%	100%		

Results: previous aid BCHA

SF-36 mean (SD)	**BCHA (n=20)**	**BAHA (n=20)**	**Mean difference**	**Effect size**
Physical functioning	69.2 (25.4)	70.8 (24.6)	1.4 (p=NS)	–0.06
Role limitations (physical)	61.3 (40.1)	57.5 (45.2)	–3.8 (p=NS)	0.09
Role limitations (emotional)	76.7 (39.1)	63.3 (41.8)	–13.4 (p=0.19)	0.33
Vitality	60.8 (16.6)	61.0 (21.9)	0.2 (p=NS)	–0.01
Mental health	68.4 (17.6)	74.2 (14.2)	5.8 (p=NS)	–0.36
Social functioning	80.6 (17.9)	82.2 (18.3)	1.6 (p=NS)	–0.09
Pain	73.8 (20.0)	67.9 (27.9)	–5.9 (p=0.30)	0.24
General health	61.0 (19.8)	59.5 (20.3)	–1.5 (p=NS)	0.07
No statistically significant changes in any domain. With BAHA, role limitations (emotional) deteriorated (meaning increased emotional problems) and pain scores were lower (meaning more pain experienced), but not statistically significant. Mental health improved, but not statistically significant. The clinical effect was small (–0.36)				
EQ-5D mean (SD)	**BCHA (n=20)**	**BAHA (n=20)**	**Mean difference**	**Effect size**
5 domains (score 1–3):				
Mobility	1.35 (0.49)	1.50 (0.51)	0.15 (p=0.26)	–0.3
Self-care	1.2 (0.41)	1.10 (0.31)	–0.10 (p=ns)	0.28
Usual activities	1.60 (0.68)	1.55 (0.60)	–0.05 (p=ns)	0.08
Pain/discomfort	1.7 (0.57)	1.85 (0.49)	0.15 (p=0.26)	–0.28
Anxiety/depression	1.26 (0.45)	1.20 (0.41)	–0.06 (p=ns)	0.13
Utility (score 0–1)	0.71 (0.23)	0.70 (0.19)	–0.01	0.05
Visual analogue scale (score 0–100)	74.0 (16.0)	72.4 (17.4)	–1.6	0.10

Scores on mobility and pain/discomfort increased, meaning patients were slightly less mobile and experienced more pain/discomfort. Effect size small

HHDI mean (SD)	BCHA ($n=20$)	BAHA ($n=20$)	Mean difference	Effect size
Disability	31.0 (6.0)	20.8 (8.2)	–10.2 ($p<0.01$)	1.42
Handicap	27.4 (6.2)	21.8 (8.0)	–5.6 ($p<0.01$)	0.79

Statistically significant improvements in disability and handicap, large clinical impact

	BCHA ($n=20$)	BAHA ($n=20$)
Number of otolaryngology visits over preceding 6 months for draining ears, mean (SD)	19 patients, 5.4 (4.9) visits, range 0–20	20 patients, 1.5 (2.1), range 0–6
Patient preference in regard to:		
Otorrhoea (%)	0 (0)	5 (25)
Skin irritation (%)	2 (10)	10 (50)
Per cent using aid >8 hours per day	90	100

Methodological comments

- Allocation to treatment groups: two subgroups according to previous hearing aid (ACHA or BCHA). All patients then fitted with BAHA. Assessments made pre- and post-surgery
- Blinding: NR
- Comparability of treatment groups: hearing loss was more profound and participants were older in the BCHA subgroup
- Method of data analysis: difference scores were used to compare pre-surgery and post-fitting results and were analysed with the *t*-test, $p<0.05$ chosen as level of significance. Effect size calculated, which shows the absolute clinical effect of the difference between the previous aid and the BAHA on a certain question, irrespective of the number of patients. Effect size 0.2–0.5 = small effect; 0.5–0.8 = moderate effect; >0.8 = large effect
- Sample size/power calculation: NR
- Attrition/dropout: NR, although states that 'in the case of missing data, aggregated domains were not computed (at most, this reduced the overall number of patients from 56 to 51)'

General comments

- Generalisability: adults with acquired conductive or mixed hearing loss. Five participants given the more powerful BAHA Cordelle owing to their sensorineural component
- Outcome measures: postal-based questionnaire completed before BAHA surgery and after 6 months BAHA experience. SF-36, EQ-5D and HHDI are valid and appropriate measures. Data on frequency of episodes of otorrhoea and prevalence of skin irritations do not appear to be reported in paper. Data on preference with regard to otorrhoea and skin irritations presented, but validity unclear
- Inter-centre variability: not applicable
- Conflict of interests: states that the authors have no relevant financial interest in the article

NR, not reported; NS, not significant.

Quality assessment for primary studies (modified for BAHAs)

A. Selection bias					
1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	< 60%	N/A	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate* x	*Weak*		
B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT Controlled clinical trial Cohort analytic (two group pre and post) Case–control Cohort [one group pre and post (before and after)] Interrupted time series Other – specify Can't tell				x reviewers opinion
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below.					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?' ■ If there is one group of participants in the study: 'are potential confounding variables reported?'	Yes	No x	Can't tell		
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropout				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes x	No	Can't tell	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes x	No	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes x	No	Can't tell	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and design	Intervention	Participants	Outcome measures
Snik *et al.* Three linked studies: 1998,[81] 1994[80] and 1992[79] Netherlands *Design*: cohort (one group pre and post) *Study setting*: hospital otorhinolaryngology department *Number of centres*: one;[80,81] two[79] Data on TBS from a second centre presented but not extracted[79] *Funding*: grants from the Fund of the Investigative Medicine of the Ziekenfondsraad;[81] NR[79,80]	1. BCHA 2. ACHA 3. BAHA HC 200,[81] 4. HC 200 and HC 220,[80] 5. HC 220[79] One patient did not want to use a body-level hearing aid so a behind-the-ear combined with the HC 200 was used instead[79]	**1998:**[81] *Indication for treatment*: conductive or mixed type binaural hearing loss, with SNHL of ≤ 30 dB HL *Number of participants*: $n=41$ (BCHA, $n=33$; ACHA: $n=8$) *Sample attrition/dropout*: $n=2$ (15%) *Sample dropout/attrition*: two unrelated deaths – long-term follow-up $n=39$ *Inclusion/exclusion criteria for study entry*: ■ binaural hearing loss of conductive or mixed type ■ SNHL component of at most 25–30 dB HL ■ no surgical options for hearing improvement ■ rejection of a conventional BCHA due to pain or skin irritation caused by the pressure of the BC vibrator Note: these participants appear to be the same as those in the HC 200 group of the 1994 study[80] **1994:**[80] *Indication for treatment*: chronic otitis media, chronic otitis externa, aural atresia *Number of participants*: $n=58$ (HC200, $n=42$; HC220, $n=16$) *Sample attrition/dropout*: five did not complete speech-in-noise test *Inclusion/exclusion criteria for study entry*: all patients at the Nijmegen clinic who were fitted with a BAHA between 1988 and 1992. No other details reported Note: six of the participants from the HC 220 group are reported in the 1992 study[79] The participants in the HC 200 group appear to be the same as those in the 1998 study[81]	**1998:**[81] *Primary outcome*: SRT in quiet and noise *Secondary outcome*: subjective opinion questionnaire on device use and speech recognition in quiet and noise *Method of assessing outcomes*: Speech Recognition in Noise Test[91] consisted of 13 sentences and a steady-state, speech-shaped noise presented at a fixed level. SRT of the sentences established with an adaptive procedure. The critical speech-to-noise ratio (difference between SRT and noise level in decibels) was determined Speech-to-noise ratio is independent of the volume setting of the hearing aid, as long as the speech level is above the patient's threshold. The difference in speech-to-noise ratio between the old and new device was expressed as a change in the percentage of correctly repeated sentences (i.e. change of 1 dB in the speech-to-noise ratio equals 17% change in sentence recognition) SRT also determined in quiet SRT results calculated by deducting the new device from the old device and averaging it for each subgroup The questionnaire was marked on a 1–10 scale (impossible to excellent), administered pre- and post-surgery (details previously described in Mylanus *et al.*[145]). Questions on recognition of speech in relatively quiet surrounding (five subquestions) and noisy situations (nine subquestions) were considered. An average score on both sets of subquestions was calculated for each patient. During follow-up, patients were regularly asked about actual use of the BAHA, and any relevant medical and technical problems were documented *Length of follow-up*: ACHA and BCHA data obtained before surgery. BAHA evaluated after at least 6 weeks. Questionnaire pre surgery and 3–5 months post BAHA fitting. Long-term evaluation exceeded 4.5 years **1994:**[80] *Primary and secondary outcomes*: percentage of patients whose SQ score (speech recognition in quiet) and speech-to-noise ratio (speech-to-noise ratio) improved or deteriorated significantly Questionnaire on speech recognition in quiet and noisy situations *Method of assessing outcomes*: SQ: free-field phoneme recognition score obtained using standard phonetically balanced lists of monosyllables, presented at 60 dB. If phoneme score < 100%, phoneme scores also obtained at 70 dB and 80 dB. SQ value is the maximum phoneme score obtained Speech recognition in noise: used test by Plomp and Mimpen[89] (see above). Noise presented at 65 dB(A). SRT and speech-to-noise ratio determined as above Questionnaire answers rated on a scale 1–10. Five subquestions for speech in quiet, nine subquestions for speech-in-noise *Length of follow-up*: tests on previous aids performed 1–8 weeks prior to fitting BAHA Tests on BAHA after at least a 4-week period of daily use Questionnaire completed before surgery and 5 months after BAHA fitted

	1992:[79] *Indication for treatment*: severe mixed hearing loss *Number of participants*: 12 Previous aid: BCHA, 7; ACHA, 5 *Sample attrition/dropout*: speech recognition thresholds could not be obtained for some patients *Inclusion/exclusion criteria for study entry*: all had recurrent otorrhoea preventing use of occluding ear moulds Note: six of these participants are also reported in the 1994 study[80]	**1992:**[79] *Primary and secondary outcomes*: maximum phoneme score; speech recognition threshold; average difference between free-field warble thresholds; patient's opinions (questionnaire, not validated) *Method of assessing outcomes*: warble-tones use to obtain free-field thresholds, generated by the standard audiometer, with a frequency modulation of 5%. Sound was presented from the front via loudspeaker Free-field speech audiogram used standard Dutch PB word lists consisting of 10 monosyllables. The level of the (fluctuating) signal was read at the slow speed, using the 'A' filter. The readings for 40 words were averaged and the free-field speech levels are presented in dB(A) Phoneme scores as a function of the presentation levels were recorded separately for the BAHA and ACHA, and the maximum phoneme score and speech recognition threshold were determined, the later being the presentation level in dB(A) at which 50% of the presented phonemes were repeated properly by the patient Average difference between the free-field warble thresholds obtained by subtracting the thresholds obtained with the BAHA from those obtained with the previous aid Questionnaire: patient's opinions rated on scale 1 to 10. Three scores were calculated from this: speech recognition in quiet, speech recognition in noise, and comfort. The scores are the average of the rated scores of the questions involved per topic. Positive scores indicate better score with BAHA *Length of follow-up*: tests performed after at least 4 weeks of daily use Questionnaire after at least 4 months of daily use with BAHA

1998[81] characteristics of participants

	BAHA ($n=41$)	**p-value**
Age, mean, years (range)	43 (10–70)	
PTA AC, dB HL (range)	55 (30–90)	
PTA BC, dB HL (range)	16 (0–28)	

Comments: PTA indicates average hearing loss at 0.5, 1.0 and 2.0 kHz

1998[81] results

	Previous aid BCHA ($n=33$)	**Previous aid ACHA ($n=8$)**	**p-value**
Change in SRT in quiet (previous aid minus BAHA), mean dB (SD)	2.7 (4.4)[a]	–6.4 (3.7)[b]	
Improvement in speech-to-noise ratio, mean dB (SD)	2.5 (2.2)[a]	1.6 (1.0)[a]	
Questionnaire	**Previous aid BCHA ($n=33$)**	**Previous aid ACHA ($n=8$)**	**p-value**
Speech recognition in quiet surroundings, median change in questionnaire score (range)	1.4 (–0.6–5.6)	0.2 (–1.4–3.3)	
Speech recognition in noisy surroundings, median change in questionnaire score (range)	1.6 (–0.8–7.0)	0 (–1.5–4.0)	

Note: change in score from previous aid NS for both subgroups

	Previous aid BCHA ($n=33$)			**Previous aid ACHA ($n=8$)**		
Proportion of patients who preferred each device with regard to:	**New device**	**No preference**	**Old device**	**New device**	**No preference**	**Old device**
Speech recognition in noisy surroundings,% of patients[c]	~76	~12	~12	~37	~26	~37
Speech recognition in quiet surroundings,% of patients[c]	~70	~20	~12	~50	~24	~24

Comments: after the trial, all patients chose to use the BAHA, not their previous aid

Adverse events	*BAHA (n = 39)*[d]	p-*value*
Lost implant due to inflammation after 2 years of use	1 (implant not replaced)	
Requested implant removal due to pain after 3 years	1	
Implants loss owing to inflammation	1 (implant replaced)	
Lost implant due to trauma	2 (implant replaced)	
Reduction of thickness of the subcutaneous layer around implant to minimise risk for inflammation	2	
Total re-operations	6	
Rejections of BAHA due to insufficient amplification	0	
Severe deterioration in sensorineural hearing (25–65 dB HL) after surgery for cholesteatoma in the cerebellopontine angle and refitted with a more powerful BAHA (NBC-HC-220). However, result was poor owing to severe deterioration of cochlear function	1	
Non-users after at least 4.5 years (all others using BAHA on daily basis)	2/39 (5%)	

1994[78] *characteristics of participants*

Age, years	Range 10–77
Average hearing loss at 0.5, 1.0 and 2.0 kHz in best ear	Range 30–100 dB HL
Average bone-conduction thresholds at 0.5, 1.0 and 2.0 kHz (PTA[b,c])	HC200, 0–44 dB HL; HC220, 33–63 dB HL
History of patients	Chronic otitis media, 86%; chronic otitis externa, 5%; aural atresia, 9%
Previous hearing aid	BC 44/58 (76%); AC 14/58 (24%)

1994[78] *results*

Outcomes	**Previously used BC**		
Percentage of patients with a statistically significant improvement or deterioration in:	**HC 200 (*n* = 33)**	**HC220 (*n* = 11)**	***p*-value**
SQ score	Improved, 12; deteriorated, 0	Improved, 54; deteriorated, 0	
Speech-in-noise score	Improved, 60; deteriorated, 0	Improved, 44; deteriorated, 0	
Speech recognition in quiet (questionnaire)	Improved, 63; deteriorated, 9	Improved, 91; deteriorated, 0	
Speech recognition in noise (questionnaire)	Improved 75; deteriorated, 12	Improved, 91; deteriorated, 0	
Comments: in the total group of patients who previously used BC, the average subjective improvement with the BAHA on the speech recognition-in-quiet and in-noise was > 1.3 points			
	Previously used AC		***p*-value**
Percentage of patients with a statistically significant improvement or deterioration in:	**HC 200 (*n* = 9)**	**HC220 (*n* = 5)**	
SQ score	Improved, 0; deteriorated, 11	Improved, 40; deteriorated, 20	
Speech-in-noise score	Improved, 55; deteriorated, 11	Improved, no results; deteriorated, no results	
Speech recognition in quiet (questionnaire)	Improved, 22; deteriorated, 44	Improved, 80; deteriorated, 20	
Speech recognition in noise (questionnaire)	Improved, 11; deteriorated, 44	Improved, 80; deteriorated, 20	

1992[79] characteristics of participants

	Previous aid BCHA ($n=7$)	Previous aid ACHA ($n=5$)
Age, years mean (SD), range[e]	60.6 (18.8), 34–84	62 (13.9), 46–78
PTA for BC, dB HL, mean (SD) range[e]	46.2 (12.6), 28 to >62	49.6 (7.3), 40–57
PTA for AC, dB HL, mean (SD) range[e]	91.1 (14.3), 70–108	84.8 (12.3), 72–100
Hearing aid use (years)	23 (range 7–40)	

1992[79] results

Previous aid BCHA ($n=7$)

	BCHA	BAHA	*p*-value
Maximum phoneme score,%, mean (SD) range[e]	36.1 (28.9), 0–85	48.7 (31.7), 0–100	
Speech recognition threshold, dB(A), mean (SD) range[e]	($n=2$); 40 (7.1), 35–45	($n=4$); 38.8 (11.1), 25–50	
Average difference between the free-field warble thresholds, dB[c]	(BCHA minus BAHA)		
	250 Hz: 2		
	500 Hz: –3		
	1000 Hz: –2		
	2000 Hz: –10		
	4000 Hz: –14		
	8000 Hz: NR		

Comments: the maximum phoneme score with the BAHA was equal to the BCHA in three patients and better in four patients (range of improvement 15–28%)

Speech recognition threshold values could be compared only in two patients: in one patient the value was 10 dB better with the BAHA; in one patient the values were equal

At higher frequencies, the average difference in warble-tone thresholds was negative, indicating that the hearing in this region was, on average, better with the BAHA than BCHA

Change scores from questionnaire, mean (SD), range:[e]	**BAHA minus BCHA**	***p*-value**
Speech recognition in quiet	0.7 (2.0), –1.2–4.4	
Speech recognition in noise	0.4 (2.0), –3.0–2.8	
Comfort	1.0 (1.0), 0.0–2.7	

Previous aid ACHA ($n=5$)

	ACHA	BAHA
Maximum phoneme score,%, mean (SD) range[e]	81.6 (8.7), 70–90	67.6 (22.2), 43–90
Speech recognition threshold, dB(A), mean (SD) range[e]	39 (10.8), 20–45	($n=3$); 45 (5), 40–50
Average difference between the free-field warble thresholds, dB[c]	(ACHA minus BAHA)	
	250 Hz: –6	
	500 Hz: –5	
	1000 Hz: 3	
	2000 Hz: 4	
	4000 Hz: 15	
	8000 Hz: 0	

Comments: maximum phoneme scores with the BAHA were better in one patient (+10%), equal in one patient and worse in three patients (–13–40%)

Speech recognition threshold values could be compared in three patients: values obtained with the ACHA and BAHA were equal within 5 dB

At higher frequencies, the average difference in warble-tone thresholds was positive, indicating that the hearing in this region was, on average, better with the ACHA than the BAHA

Change scores from questionnaire, mean (SD), range:[e]	BAHA minus ACHA
Speech recognition in quiet	–1.0 (4.6), –5.8–3.5
Speech recognition in noise	0.1 (3.3), –4.2–3.7
Comfort	0.6 (3.2), –3.2–5.4

Methodological comments

- Allocation to treatment groups: 1998:[81] patients divided into two groups based on previous hearing aid (BCHA or ACHA). 1994:[80] all patients at the clinic who had been fitted with a BAHA were included, and allocated to subgroups according to the type of BAHA fitted and the type of previous aid used. 1992:[79] the paper states that the tests on the BAHA and previous aids were conducted separately and after 4 weeks of continuous use. However, the order of the tests is not stated. The reviewers have made an assumption that the study is a cohort pre–post design; however, it is possible that this is not the case
- Blinding: NR[79–81]
- Comparability of treatment groups: 1998:[81] no subgroup baseline characteristics. 1994:[80] SNHL was more severe in patients fitted with an HC220, but otherwise baseline data NR. 1992: not clear[79]
- Method of data analysis: 1998:[81] no details of data analysis provided. Pre- and post-data NR, only change in outcomes reported. Only five subquestions on speech recognition in quiet surroundings and nine subquestions on noise situations were used from the questionnaire. 1994:[80] difference in SQ values tested using critical difference according to Thornton and Raffin.[146] Difference in speech-to-noise ratios was compared with the 95% CI calculated from the known intra-individual SD. 1992:[79] statistical analysis not undertaken. Presented correlation of questionnaire outcomes with MPS, but not data extracted
- Sample size/power calculation: none reported[79–81]
- Attrition/dropout: 1998:[81] two unrelated deaths are reported. 1994:[80] five with more severe inner ear impairment (HC 220 group) found the speech recognition in noise test too difficult so it was discontinued. 1992:[79] the speech recognition threshold could not be determined in five patients with their previous hearing aid and five patients with a BAHA because the maximum score was < 50%. The speech recognition threshold values of both hearing aids could be compared in only five patients

General comments

- Generalisability: 1998:[81] adults with conductive or mixed type binaural hearing loss, with SNHL of ≤ 30 dB HL. 1994:[80] includes patients with chronic otitic media, chronic otitis external and aural atresia, but limited baseline data presented. Patients with normal to moderate SNHL and more severe SNHL included. 1992:[79] patients with severe mixed hearing loss, with sensorineural components of 45–60 dB HL
- Outcome measures: it is unclear if the questionnaires on subjective opinions are validated,[79–81] and the clinical meaning of the change scores is not clear.[79,81] 1994:[80] Speech recognition in quiet and in noise assessed pre-operatively and post-operatively, but data not presented. Instead, the proportion of patients who improved or deteriorated is derived from the data and presented. The validity of this is not clear. 1998 and 1992: hearing measures appropriate[79,81]
- Inter-centre variability: not applicable.[80,81] NR[79]
- Conflict of interests: NR[79–81]

NR, not reported; NS, not significant; SQ, speech recognition-in-quiet; TBS, temporal bone stimulator.
a $p < 0.05$ significant improvement.
b $p < 0.05$ significant deterioration.
c Data estimated from figure by reviewer. Note that the legend on this figure appears to be incorrectly labelled.
d 2/41 deaths due to causes not related to hearing problems.
e Individual patient data presented in paper, means and SDs calculated by reviewer.

Quality assessment for primary studies (Snik *et al*. 1998[81])

A. Selection bias					
1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	< 60%	Not applicable	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate* x	*Weak*		
B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT Controlled clinical trial Cohort analytic (two group pre and post) Case–control Cohort [one group pre and post (before and after)] Interrupted time series Other – specify Can't tell			x reviewer's opinion	
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
Two groups: are confounders reported AND controlled for in the analysis? OR if one group: are potential confounding variables reported?	Yes	No x	Can't tell		
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of binding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
E. Data collection methods					
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell		
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell		
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*		

F. Withdrawals and dropouts				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes x	No	Can't tell	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100% x	60–79%	< 60%	Can't tell
Summary of withdrawals and dropouts (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No	Can't tell x	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes	No	Can't tell x	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No X	Can't tell	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Appendix 9

Data extraction: BAHA versus unaided hearing

Reference and design	Intervention	Participants	Outcome measures
Kompis *et al.* 2007[66] Switzerland *Design*: cohort pre–post *Study setting*: secondary care *Number of centres*: single centre *Funding*: Divinos provided by Entific Medical Systems	1. BAHA Divino 2. Unaided *Other interventions used*: none Note: BAHA Compact also assessed but not extracted. See *General comments*	*Indication for treatment*: substantial bilateral CHL, some combined with mild-to-moderate SNHL *Number of participants*: seven *Sample attrition/dropout*: one did not complete questionnaires *Inclusion/exclusion criteria for study entry*: NR	*Primary and secondary outcomes*: sound field thresholds using narrow-band noise; speech audiometry in quiet and noise; APHAB questionnaire (data not presented in paper) *Method of assessing outcomes*: All speech materials were pre-recorded in German, presented to the participant who was sat in between two loudspeakers, placed just off one diagonal axis Speech audiometry in quiet: speech recognition thresholds in quiet (levels required for 50% speech understanding) measured using Freiburger two-digit numbers; the percentage of correctly repeated words at 50, 65 and 80 dB SPL measured using Freiburger monosyllabic words Speech audiometry in noise used Basler sentence test, speech was presented at 70 dB and the SNR in dB, at which 50% of the key words were understood correctly, was measured. The speech signal was emitted from a loudspeaker in front of the listener and noise was emitted either from the same direction or from the back (180°) *Length of follow-up*: unaided and Compact assessed at month 0. Then 3 months use with Divino. Divino and Compact assessed at 3 months

Characteristics of participants

Age, years, mean (range)	48.6 (19–66)
Sex (M : F)	3 : 4

No other hearing aid used in contralateral ear

Comments: five used a Compact, two had experience with a Compact but were regular users of a Classic 300. At least 2 years use with BAHA Compact or Classic 300 prior to study

AC and BC thresholds in both ears presented for individual patients in a figure (not data extracted). PTA 'yielded essentially the same results at 0 months and 3 months' [average of the differences: AC 0.3 dB (SD 5.0); BC –1.2 dB (SD 4.2)]

Results

Outcomes	Unaided (0 months)	Divino (3 months)	*p*-value
Average improvement in sound field thresholds over all frequencies compared with unaided, dB		28.0	$p < 0.0001$ vs unaided
Speech recognition thresholds in quiet using two-digit numbers, dB[a]	54	23	NR

Speech recognition thresholds in quiet using two-digit numbers: average improvement between unaided and Compact at 0 months, and between unaided and Compact or Divino at 3 months = 29.0–30.3 dB, $p = 0.016$

	Unaided (0 months)	Divino (3 months)	*p*-value
Speech recognition scores for monosyllabic words in quiet, % correct[a]	50 dB SPL: 5	50 dB SPL: 45	
	65 dB SPL: 15	65 dB SPL: 90	
	80 dB SPL: 50	80 dB SPL: 95	

Comments: the average gain in speech understanding over all presentation levels (50–80 dB) is 52%. The improvement is statistically significant ($p < 0.001$) for each of the three aided conditions (Compact 0 months, Compact 3 months, Divino 3 months)

	Unaided (0 months)	Divino (3 months)	*p*-value
Speech recognition threshold in noise (noise presented from front or back), dB[a]	Front: 12 Back: 9	Omnidirectional mode Front: 3 Back: 3 Directional mode Front: 4 Back: 1	

Comments: Compact and Divino (omnidirectional and directional mode) had significantly better speech recognition threshold in noise than unaided, when noise arrives from both the front and rear (average improvement of 8.7–12.0 dB, $p = 0.03$ for all comparisons)

Methodological comments

- Allocation to treatment groups: one group experienced all conditions
- Blinding: none
- Comparability of treatment groups: PTA gave similar results at 0 months and at 3 months, average of the difference + 0.3 dB (SD 5.0) for AC and –1.2 dB (4.2) for BC
- Method of data analysis: Wilcoxon matched-pairs signed rank test used to compared unaided with Compact at 0 months, Compact at 3 months and Divino at 3 months. Most data presented in figures only
- Sample size/power calculation: NR
- Attrition/dropout: one patient was unable to complete the questionnaires

General comments

- Generalisability: not clear. All participants had a substantial bilateral CHL; some had an additional, predominately mild SNHL
- Outcome measures: a description is given of how the tests were conducted, including the acoustic chamber, audiometer, position of speakers, position of participants, calibration of equipment. Order of testing as well as the order of test lists was varied systematically between participants to avoid bias due to training or fatigue. The APHAB appears to be a validated measure, but data are not presented. Only selected items from the 'custom-made' questionnaire were presented
- Inter-centre variability: not applicable
- Conflict of interests: not stated. Divinos provided by Entific Medical Systems
- Other: after 3 months with Divino, PTA, unaided and aided sound field thresholds, and speech audiometry in quiet were repeated with the Compact to ensure that no significant change in hearing had occurred
- The aim of the study was to compare the BAHA Compact and BAHA Divino, but this comparison is not eligible for inclusion in the systematic review. The BAHA Compact was assessed at baseline and at 3 months' follow-up, but had not been worn for the 3 months prior to the follow-up assessment. Therefore the BAHA Compact data were not extracted

APHAB, abbreviated profile of hearing aid benefit; F, female; M, male; NR, not reported.
a Data estimated from figure by reviewer, assume that it is the mean.

Quality assessment for primary studies

A. Selection bias					
1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100%	60–79%	< 60%	Not applicable	Can't tell x
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT				
	Controlled clinical trial				
	Cohort analytic (two group pre and post)				
	Case–control				
	Cohort [one group pre and post (before and after)]			x reviewer's opinion	
	Interrupted time series				
	Other – specify				
	Can't tell				
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?' ■ If there is one group of participants in the study: 'are potential confounding variables reported?	Yes	No x	Can't tell		
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
E. Data collection methods					
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell		
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell		
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*		

F. Withdrawals and dropouts				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No x	Can't tell	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes x	No	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Appendix 10

Data extraction: unilateral versus bilateral BAHAs

Reference and design	Intervention	Participants and outcome measures
Bosman *et al.* 2001[60,85] Netherlands *Design*: audiological comparison study *Study setting*: clinic *Number of centres*: one *Funding*: Entific Audiological comparison study	1. BAHA – bilateral. HC 200 or Classic 300 2. BAHA – unilateral (first implant side). HC 200 or Classic 300	***Participants*** *Indication for treatment*: contraindication to ACHA due to either recurrent otorrhoea or otitis externa, or to congential aural atresia *Number of participants*: 25 Only nine participants undertook the binaural masking level difference (BMLD) assessments *Sample attrition/dropout*: NR *Inclusion/exclusion criteria for study entry*: all bilaterally fitted patients from the clinic; at least 3 months' experience with two BAHAs. Initial series of participants had to have symmetry of BC thresholds (average BC thresholds across 0.5, 1.0, 2.0 and 4.0 kHz to not differ by more than 10 dB; thresholds at individual frequencies should lie within 15 dB) but criterion of symmetry relaxed slightly for latter phase of the study (no details of the new criterion or the numbers of participants) ***Outcome measures*** *Primary outcomes*: directional hearing, speech reception in quiet and noise, SRT, BMLD *Method of assessing outcomes*: directional hearing with either seven or nine loudspeakers arranged in a circle with a 1-metre radius, distributed with 30° intervals, spanning 180° or 240°. One placed to the front and three (or four) to the left and right. Stimuli consisted of 1-second noise bursts. The BAHA volume control was kept in the position used by the patient in everyday life. All stimuli were presented two or three times to estimate test–retest reliability. To obtain equivalent results for unilateral left- and right-sided fittings, data from right sided fitting were mirrored to left-sided fittings before pooling results Speech recognition was measured with the sentence material of Plomp and Mimpen[91] with a female speaker and Smoorenburg[96] with a male speaker. Each sentence contained eight or nine syllables and was representative of everyday speech In quiet, the speech material was presented in front of the participant, in noise, speech presented at the front, with masking noise at + 90° or –90° at baffle side (side participant used unilateral BAHA) or shadow side (opposite side) Sounds were presented at 65 dB(A) [except for one patient, 60 dB(A) was used as 65 dB(A) was 'too loud'] SRT used the adaptive one-up one-down procedure of Plomp and Mimpen to determine the presentation level, to provide a whole-sentence correct score of 50%. Each list contained 13 sentences; first three to obtain an initial estimate of the SRT, the next 10 were averaged to produce the SRT for the condition. Participants had to repeat the sentences as accurately as possible. Effects of quiet and noise also tested. SNR = value relative to the noise level, where better performance equals a more negative SNR. A 1 dB change in SRT corresponds to a change in score of about 15% for normally hearing listeners. Both stimulus conditions and sentence lists were varied to a counterbalanced design. In the quiet conditions, four lists uttered by the male speaker were used, and in the noise conditions, eight lists by the female speaker were used. Noise conditions were measured twice to allow for test–retest estimation BMLD stimuli were generated by an audiometer, the output of which was connected to the external input of the BAHA 300 (two 'matched' devices were used which had been checked by Entific for phase and amplitude characteristics). BMLDs were measured with pure-tones of 125, 250, 500 and 1000 Hz masked by 1/3 octave bands of filtered white noise centred at the stimulus frequency. Thresholds obtained for (1) in-phase tone stimuli and noise bands (S_0N_0); (2) 180° out-of-phase tone stimuli and in-phase noise bands ($S_\pi N_0$); and (3) in-phase tone stimuli and 180° out-of-phase noise bands (S_0N_π). Noise bands were presented at the participants most comfortable level. The tone stimulus had a rhythmic pattern with symmetric on and off intervals of 0.5 seconds. Detection thresholds were measured with manual procedures employing 1 dB steps and measured twice for test–retest reliability. The level difference between S_0N_0 and $S_\pi N_0$ was taken as the BMLD estimate. The BAHA volume controls were kept at their maximum value *Length of follow-up*: same day. Two 45-minute testing sessions with a break in between

Characteristics of participants

	BAHA
Age, years mean (standard deviation, SD), range[a]	44.3 (16.3) 12–74
Sex (M : F)	14 : 11
Diagnosis	Six congenital atresia (four with Treacher Collins syndrome) of which five had bilateral aural atresia. Four of these have been published previously[85] Nineteen recurrent otorrhoea [eight with cholesteatoma; 10 with chronic otitis (externa); one with cheilognato-palato schisis]. Three of these have been published previously[147] All of the cholesteatoma patients had a previous radical mastoidectomy
BAHA experience, unilateral, mean (SD) months, range[a]	49.1 (26.2), 18–105
BAHA experience, bilateral, mean (SD) months, range[a]	13.6 (9.2), 3–39
PTA (PTA) at 0.5, 1.0 and 2.0 kHz, dB HL, first side, air conduction (AC), mean (SD), range[a]	59.5 (13.7), 32–82
PTA at 0.5, 1.0 and 2.0 kHz, dB HL, first side, BC, mean (SD), range[b]	21.0 (10.7), –5–36
PTA at 0.5, 1.0 and 2.0 kHz, dB HL, second side, AC, mean (SD), range[a]	63.6 (10.9), 38–82
PTA at 0.5, 1.0 and 2.0 kHz, dB HL, second side, BC, mean (SD), range[b]	21.9 (12.4), –8–48

Results

Directional hearing at 500 Hz, %[c] $n=25$	**Unilateral**	**Bilateral**	***p*-value unilateral vs bilateral**
Correct localisation	23	42[d]	Across all observations $p<0.001$
Localisation within 30°	56	90[d]	
Lateralisation	54	85[d]	
Directional hearing at 2 kHz, %[c] $n=25$			
Correct localisation	24	45[d]	
Localisation within 30°	58	89[d]	
Lateralisation	64	87[d]	

Comments: the effect of stimulus frequency was not statistically significant, $p>0.1$

Paper also states that many participants had difficulty localising sound with one BAHA, that all sounds appeared to come from the fitted side. The bias of responding to the fitted (baffle) side is shown only when aggregating individual response matrices to a group matrix. With unilateral fittings, 75.3% and 70.3% of the responses (for the 500 Hz and 2 kHz stimuli, respectively) correspond to the fitted side. With bilateral fittings, the response patterns are more symmetrical (45.7% and 48.8% of responses corresponded to the fitted side, respectively)

Also reports data for a subgroup of participants with congenital atresia ($n=6$) but not extracted here

Speech recognition $n=25$	**Unilateral**	**Bilateral**	***p*-value**
SRT in quiet [dB(A)]	41.5	37.5	$p<0.001$
Speech-to-noise ratio [dB(A), noise from the baffle side	–0.7	–3.2	$p<0.001$
Speech-to-noise ratio [dB(A)], noise from shadow side	–3.4	–4.0	$p>0.05$

Comments: standard error based on test–retest data also presented in a figure but not estimated here

Also reports data for a subgroup of participants with congenital atresia ($n=6$) but not extracted here

BMLD SNR, ($n=9$)[c]	**Condition: S_0N_0**	**Condition: $S_\pi N_0$**	**Condition: S_0N_π**	***p*-value**
125 Hz	2.2	–3.8	–3.7	$p<0.001$ at 125, 250 and 500 Hz. Not significant ($p>0.05$) at 1 kHz
250 Hz	0.1	–6.0	–5.1	
500 Hz	0.4	–5.9	–3.9	
1 kHz	0.4	–3.3	–4.9	

Comments: text states that in the $S_\pi N_0$ condition the SNR's ('release from masking') were 6.1, 6.0, 6.6 and 4.1 for the frequencies 125, 250, 500 and 1000 Hz respectively; however, the figure does not appear to support this

Methodological comments

- Allocation to treatment groups: one series of patients from the clinic who had two BAHAs for at least 3 months. Report suggests that this is the complete group from the clinic
- Blinding: NR
- Comparability of treatment groups: not applicable
- Method of data analysis: reports that repeated measure ANOVAs used for testing significance, but no further details. Chance levels calculated for directional hearing measurements based on 1/7, 3/7 and 1/2 for the correct localisation, localisation within 30° and lateralisation scores, respectively. States that the 95% CIs were based on twice the SD of a binomial distribution based on 19 items. Subgroup of nine participants undertook the BMLD assessment/analysis
- Sample size/power calculation: NR
- Attrition/dropout: NR

General comments

- Generalisability: consecutive cases from the Netherlands with experience of bilateral BAHAs. The six patients with congenital atresia had lifelong experience with unilateral BCHA
- Outcome measures: appear valid
- Inter-centre variability: not applicable
- Conflict of interests: not noted by authors. Financial assistance and technical cooperation from Entific, the manufacturer of BAHAs

ANOVA, analysis of variance; BMLD, binaural masking level difference; F, female; M, male; NR, not reported.
a Mean (SD) calculated by reviewer.
b SD calculated by reviewer.
c Estimated by reviewer.
d $p < 0.05$ vs chance level. Chance levels (and 95% CI) were set by the study authors and were 14.3% (CI 32[c]) for a correct score, 42.9% (CI 64[c]) for localisation within 30° and 50% (CI 71[c]) for lateralisation.

Quality assessment for primary studies (modified for BAHAs)

A. Selection bias

1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	< 60%	Not applicable	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate* x	*Weak*		

B. Study design

1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT				
	Controlled clinical trial				
	Cohort analytic (two group pre and post)				
	Case–control				
	Cohort [one group pre and post (before and after)]				
	Interrupted time series				
	Other – specify: audiological comparison study			x reviewer's opinion	
	Can't tell				
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

C. Confounders				
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?'	Yes	No x	Can't tell	
■ If there is one group of participants in the study: 'are potential confounding variables reported?'				
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* *x*	
D. Blinding				
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell	
2. Were the study participants aware of the research question?	Yes x	No	Can't tell	
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropouts				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No	Can't tell x	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes	No x	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and design	Intervention	Participants	Outcome measures
Dutt *et al.* 2002[86] UK *Design*: audiology comparison study *Study setting* outpatient: *Number of centres*: one *Funding*: NR	1. Unilateral BAHA 2. Bilateral BAHA BAHA model: Compact (information from author)	*Indication for treatment*: unilateral BAHA users whose professional needs warranted binaural hearing *Number of participants*: $n=15$ with bilateral BAHAs; $n=12$ eligible; $n=11$ participated *Sample attrition/dropout*: one chose not to participate *Inclusion/exclusion criteria for study entry*: used second-side BAHA for at least 12 months. Paper states criteria not stringent: previous knowledge and experience with binaural hearing (conventionally aided or unaided); bilaterally symmetrical hearing loss (interaural threshold difference of < 15 dB four-time average); professional needs of users, e.g. businessmen, teachers, nurses; motivation – voluntarily applied for a second-side BAHA; age (limited to adults)	*Primary outcomes*: speech recognition in quiet and in noise; modified Plomp test *Method of assessing outcomes*: right, left and bilateral BAHAs evaluated Unaided sound field levels [dB(A)] and aided thresholds Sound field speech used Arthur Boothroyd word lists Speech-in-quiet and speech-in-noise evaluated with BKB sentences, at SNRs of + 10 dB, 0 dB and –10 dB Modified Plomb Multitalker Noise Test used to evaluate speech-in-noise with open-set speech recognition. Three speakers used. Speech presented from front speaker at 70 dB(A). Speech babble noise (20 talkers/cocktail party noise) presented from left or right at a SNR of 0 dB *Length of follow-up*: audiological evaluations undertaken at same session

Characteristics of participants (n = 12)

Age, years, mean (SD), range[a]	42.3 (10), 22–54
Sex (M : F)	3 : 9
Duration with one BAHA, years, mean (SD), range[a]	6.3 (3.2), 3–12
Duration with two BAHAs, years, mean (SD), range[a]	2.2 (1.1), 1–5
Diagnosis	Treacher Collins syndrome (2)
	Bilateral mastoid cavities (3)
	Bilateral CON
	Bilateral chronic otitis media (3)
	Goldenhar syndrome
	Bilateral microtia
	Bilateral acquired otosclerosis

Results (n = 11)

Outcomes	**Best-unilateral response**	**Bilateral**	***p*-value**
Speech-in-quiet – cumulative Arthur Boothroyd word (30 words) list scores[b] at:			
30 dB intensity levels	1	5	
40 dB intensity levels	13	19	
50 dB intensity levels	20	24	
60 dB intensity levels	25	28	
70 dB intensity levels	27	29	
80 dB intensity levels	30	30	
Speech-in-quiet (BKB sentences)	Data not presented. All 11 patients scored 100% with right, left and bilateral BAHAs		
Speech-in-noise – cumulative BKB sentence scores[b] at:	**Best-unilateral response**	**Bilateral**	***p*-value**
+ 10 SNR	99	100	
Zero SNR	80	81	
– 10 SNR	0	1	

Plomp test,% correct score [mean (SD), range]:[a]	**Left BAHA**	**Right BAHA**	**Bilateral BAHA**
Sound front, noise front	76 (11.7), 56–93	77.3 (11.7), 58–90	82.4 (13.3), 60–97
Sound front, noise left	40.1 (25.3), 2–71	84.1 (11.2), 55–97	71.1 (14.9), 44–95
Sound front, noise right	88.2 (9.0), 72–100	45.8 (22.1), 13–88	79.5 (11.6), 58–93

Methodological comments

- Allocation to treatment groups: one group of participants who had used bilateral BAHAs for at least 12 months underwent audiological evaluation with unilateral and bilateral BAHAs. No details on order of tests
- Blinding: NR
- Comparability of treatment groups: not applicable
- Method of data analysis: states that no statistics applied as number of patients is small. Descriptive data in form of bar charts, cumulative scores and percentages are presented
- Sample size/power calculation: NR, but states small number of patients would make the power of any analysis insignificant
- Attrition/dropout: total of 15 patients implanted with bilateral BAHA at that centre, 12 of these who had used both BAHAs for at least 12 months were included, one chose not to participate (this patient used second BAHA for special situations including social gatherings and supermarkets)

General comments

- Generalisability: the participants were adult professionals such as businessmen, teachers and nurses. Also self-selected, as they voluntarily applied for the second BAHA. All had previous experience of binaural hearing
- Outcome measures: appear valid and reliable
- Inter-centre variability: not applicable
- Conflict of interests: states 'none declared'

BKB, Bamford–Koval–Bench; CON, congenital hearing loss; F, female; M, male; NR, not reported.
a Mean SD calculated by researcher.
b Data estimated from figure by reviewer.

Quality assessment for primary studies

A. Selection bias

1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely	Not likely x	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	<60%	Not applicable	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		

B. Study design

1. What was the study design?	RCT			
(Please tick appropriate and specify design in No. 7)	Controlled clinical trial			
	Cohort analytic (two group pre and post)			
	Case–control			
	Cohort [one group pre and post (before and after)]			
	Interrupted time series			
	Other – specify: audiology comparison study			x reviewer's opinion
	Can't tell			
2. Was the study described as randomised?	Yes	No x		
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below				
3. If answer was yes, was the method of randomisation described?	Yes	No		
4. If answer was yes, was the method appropriate?	Yes	No		
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	

C. Confounders				
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?' ■ If there is one group of participants in the study: 'are potential confounding variables reported?'	Yes	No x	Can't tell	
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
D. Blinding				
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell	
2. Were the study participants aware of the research question?	Yes x	No	Can't tell	
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes X	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropout				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes x	No	Can't tell	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100% x (11/12)	60–79%	< 60%	Can't tell
Summary of withdrawals and dropouts (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No	Can't tell x	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes	No x	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and design	Intervention	Participants	Outcome measures
Priwin *et al.* 2004[87] Sweden *Design*: audiological comparison study *Study setting*: secondary care *Number of centres*: one *Funding*: in part by Swedish Research Council for Enginerring Sciences (TFR 299–2000–576)	1. Unilateral BAHA 2. Bilateral BAHA Compact and Classic 300	*Indication for treatment*: combined symmetric or slight asymmetric sensorineural and conductive hearing level, and primarily conductive hearing level *Number of participants*: 12 *Sample attrition/ dropout*: assume none *Inclusion/exclusion criteria for study entry*: all patients fitted with bilateral BAHAs at ENT clinic at least 1 year before study	*Primary outcomes*: directional hearing; SRT; binaural hearing *Method of assessing outcomes*: BAHAs were electronically controlled by research personnel without patient's knowledge. Tests randomised and patients blinded to unilateral or bilateral use of BAHAs Pure AC (using earphones) and BC tone thresholds (using bone transducer) were measured using standard audiometric procedures and equipment. Details reported. 12 loudspeakers spaced at 30° intervals placed in a circle with a 1-m radius from patient and at height equivalent to head of sitting patient. Free-field warble tones recorded and presented using the Békésy sweep method, thresholds tested at four directions (front, left, right and behind) Directional hearing: same speaker set up as tone thresholds. Narrow-band (1/3 octave) noise centred at 0.5 or 2.0 kHz presented at 65 dB HL for 1-second duration. Three BAHA options tested: unilateral on best side (usually the aid first implanted), unilateral on shadow side (opposite side) and bilateral. Stimuli presented three times from each speaker and for each option, according to a randomised sequence of three presentations from each speaker. Data presented visually in figure (not possible to extract data) and also presented as correct score or within 30° of stimulation angle SRTs: measured in quiet and noise with phonetically balanced three-word sentences extracted from Hagerman.[97] Each test list comprised of 10 three-word sentences, presented by female voice. Aim was to find the noise level giving a 50% correct score. Three lists (one practice and two test lists) presented for two BAHA options: unilateral on best side and bilateral. Test list randomised among patients. Speech presented at 0° For SRT in noise, noise was speech weighted, presentation at either +90°, –90° or from all 11 remaining loudspeakers simultaneously (with practice and test lists and for unilateral and bilateral, as noted above) BMLD test: sensitive to proving existence of binaural hearing, carried out with bilateral BAHAs. A pure-tone signal is presented in noise and the task is to detect tone. Details reported. Test conducted at 0.25, 5.00 and 1.00 kHz, combined with a narrow and noise centred on the corresponding signal frequency at 65 dB hearing level *Length of follow-up*: unilateral BAHAs and bilateral BAHAs tested at same session	
Characteristics of participants			
Age, mean (SD), range, years[a]			51.7 (13.3), 27–68
Sex (M : F)			3 : 9
Chronic otitis, further underlying etiology unknown, number of patients			8/12
Recurrent external otitis and otosclerosis, number of patients			1/12
Congenital ear canal atresia, number of patients			3/12 (one part of Treacher Collins syndrome)
Duration with unilateral BAHAs at time of study, mean (SD), range, years[a]			14.3 (4.1), 5.8–21.0
Duration with bilateral BAHAs at time of study, mean (SD), range, years[a]			6.8 (6.0), 1.0–19.6
Use of bilateral BAHAs, number of patients			Daily: 11/12 Occasionally: 1/12
BAHA model, number of patients			Compact in both ears: 4 Classic 300 in both ears: 7 Compact and Classic 300: 1
Pure-tone average thresholds of frequencies 0.5, 1.0 and 2.0 kHz [mean (SD)],[a] range, dB HL for:			
AC (first fitted side)			58.3 (15.3), 38–87
BC (first fitted side)			29.8 (15.2), 8–53 (in three patients, at one or more frequency no fixed value was attained, highest measurable value used for mean)
AC (second fitted side)			59 (20.7), 27–102

BC (second fitted side)	30.9 (13.4), 7–50
Symmetric BC thresholds (difference ≤ 10 dB), number of patients	10/12
Asymmetric BC thresholds, number of patients	2/12 (although did not differ by > 20 dB)

Results

Improvement of free sound field tone thresholds (*n* = 12)

Comments: data presented in figure for sound presented at front, best side, shadow side and behind, for frequencies 0.25, 1.00, 1.50, 2.00, 3.00, 4.00, 6.00 and 8.00 kHz. Data not estimated and extracted by reviewer

When sound presented in front, at best side and from behind patient, the average improvement with BAHAs fitting was between 2 and 7 dB (at 0.25–8.00 kHz)

When sound presented at shadow side, the average improvement with bilateral BAHAs was between 5 and 15 dB (at 0.25–8.00 kHz)

States that the results differed greatly among the patients, that the SD of the improvement (data not presented) was almost as great as the average improvement, and that consequently the SD of the improvement when sound was at the shadow side was greater than the other three sides

Directional hearing	**Unilateral at best side**	**Unilateral at shadow side**	**Bilateral**	***p*-value**
Score (% of correct answers)[b]				
0.5 kHz	12	11	25	
2.0 kHz	8	10	23	
(Chance level for correct answer 8.3%)				
Score (% of answers within 30° of correct response)[b]				
0.5 kHz	23	30	53	
2.0 kHz	28	27	51	
(Chance level for correct answer 25%)				

Comments: states that with a unilateral BAHA, results are close to change level, but 'there is a significant increase in the ability to localise the sound source' with bilateral BAHAs

	Unilateral at best side	**Bilateral**	***p*-value**
Speech recognition in quiet, average threshold, dB HL	38.7	33.3	$p = 0.001$
Speech-in-noise	**Difference between bilateral and unilateral BAHAs**		
Masking noise presented:			
At best side	3.1 dB improvement in SNR with bilateral		
At shadow side	1.0 dB deterioration in SNR with bilateral		
As surrounding noise (remaining 11 speakers)	2.8 dB improvement in SNR with bilateral		
BMLD (relative threshold change in dB from the condition 'signal and noise in phase at both sides')	**Bilateral BAHAs**		
0.25 kHz:			
Signal 180° out of phase and noise in phase	Threshold changes within 3 dB except for two patients.		
Signal in phase and noise 180° out of phase	Threshold changes between –18 and 3 dB, mean –5 dB.		
0.5 kHz:			
Signal 180° out of phase and noise in phase	Average threshold change 2 dB		
Signal in phase and noise 180° out of phase	Average threshold change –4 dB		
1 kHz:			
Signal 180° out of phase and noise in phase	Average threshold change 3 dB		
Signal in phase and noise 180° out of phase	Average threshold change –3 dB		

Methodological comments

- Allocation to treatment groups: one group of patients (all those at ENT clinic fitted with bilateral BAHAs) underwent testing with unilateral and bilateral BAHAs at same session
- Blinding: patients were blinded to unilateral or bilateral use of BAHAs and the test order was randomised
- Comparability of treatment groups: not applicable
- Method of data analysis: paired *t*-test for speech reception scores
- Sample size/power calculation: NR
- Attrition/dropout: assume that all participants completed all tests, but not clearly stated

General comments

- Generalisability: adults with conductive or mixed hearing loss, who had experience with unilateral and bilateral BAHAs
- Outcome measures: valid and reliable. The authors note the BMLD was derived for AC testing and that with BC testing there is a significant amount of transcranial transmission
- Inter-centre variability: not applicable
- Conflict of interests: NR

BMLD, binaural masking level difference; F, female; M, male; NR, not reported.
a Means and SDs calculated by reviewer.
b Data estimated from figure by reviewer.

Quality assessment for primary studies

A. Selection bias

1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100% x	60–79%	< 60%	Not applicable	Can't tell
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate* x	*Weak*		

B. Study design

1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT			
	Controlled clinical trial			
	Cohort analytic (two group pre and post)			
	Case–control			
	Cohort [one group pre and post (before and after)]			
	Interrupted time series			
	Other – specify: audiological comparison study			x reviewer's opinion
	Can't tell			
2. Was the study described as randomised?	Yes	No x		
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below				
3. If answer was yes, was the method of randomisation described?	Yes	No		
4. If answer was yes, was the method appropriate?	Yes	No		
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	

C. Confounders				
■ If there are two groups included in the study: 'are confounders reported AND controlled for in the analysis?' ■ If there is one group of participants in the study: 'are potential confounding variables reported?'	Yes	No x	Can't tell	
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
D. Blinding				
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell	
2. Were the study participants aware of the research question?	Yes x	No	Can't tell	
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
E. Data collection methods				
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell	
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell	
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*	
F. Withdrawals and dropout				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No	Can't tell x	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes x	No	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No	Can't tell x	
Global rating for study[a] **(Overall methodological strength of study – based on sections A–F)**	**Strong**	**Moderate**	**Weak** **x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Reference and design	Intervention	Participants	Outcome measures
Priwin *et al.* 2007[59] Sweden *Design*: audiological comparison study *Study setting*: ENT clinic and audiologic units *Number of centres*: two *Funding*: Acta Oto-laryngologica foundation	1. Bilateral BAHA 2. (unaided, one BAHA, two BAHAs) 3. Unilateral BAHA (unaided, one BAHA) BAHA model: BAHA classic and BAHA compact (information provided by author)	*Indication for treatment*: children with bilateral CHL *Number of participants*: $n=9$[a] (bilateral BAHA, $n=6$; unilateral BAHA, $n=3$) *Sample attrition/ dropout*: none reported. One patient did not complete questionnaire *Inclusion/ exclusion criteria for study entry*: patients aged 6–18 years with bilateral CHL and fitted unilaterally or bilaterally with BAHAs. Appropriate school attendance and proficiency in Swedish for age	*Outcomes*: tone thresholds; speech recognition in noise; localisation of sound; questionnaires [MAIS and MUSS (validated), IOI-HA (validated)] *Method of assessing outcomes:* all participants were tested in a soundproof booth, with stimuli of sound presented through headphones, bone conductors, and/or loudspeakers. Five speakers were placed at 45° intervals in a frontal semicircle at head-height, with patients facing frontal speaker Tone thresholds were presented as PTA (M4) for the frequencies 0.5, 1.0, 2.0 and 4.0 kHz. Patients with one aid were tested with and without it; patients with two aids were tested without aid, with both BAHAs and unilaterally on their best side. Sound field thresholds were considered clinically normal at 20 dB HL Speech recognition in noise was measured with phonemically balanced Swedish three-word sentences (both speech and noise were presented from front speaker). Sentences were extracted from five-word sentence test[97] with first two words removed. After a practice list, test included two lists of 10 three-word sentences (60 words) for three different speech/noise ratios (0, 4 and 6 dB). Speech material was CD pre-recorded (female voice) and had a with fixed SNRs and modulated noise, speech presentation level set at 60 dB SPL. Noise test was presented at 60, 56 and 54 dB SPL for all patients with one to three hearing options depending on hearing aid use; (1) no amplification, (2) unilateral BAHA and (3) bilateral BAHA Localisation of sound (five loudspeakers set up): all loudspeakers were numbered and marked in different colours. A narrowband (1/3 octave) noise centred at 0.5 or 3.0 kHz was presented at 50 and 60 dB SPL for 1 second. Stimuli was presented three times from each speaker at each presentation level for each hearing option in a randomised sequence and patients had to identify the speaker presenting the sound IOI-HA to assess hearing aid outcome: seven domains (daily use, benefit, residual activity limitations, satisfaction, residual participation restrictions, impact on others and quality of life, QoL) scored 1–5 (worst to best outcome). Youngest patients allowed some assistance from parent. When comparing group data, mean scores <3.5 indicate poor habilitation outcome in patients with mild-to-moderate hearing impairment unaided and <3.6 in patients with moderate-to-severe hearing impairment MAIS and MUSS: 21 questions dealing with hearing and communication (scored 1–5, never to always) completed by children's guardian and their usual classroom teacher independently. Outcomes were then thematically grouped into hearing aid use, reaction to sounds, sound discrimination, verbal communication and speech intelligibility *Length of follow-up*: none reported, tests appear to have been undertaken at same session

Characteristics of participants

	Unilateral BAHA ($n=6$)	Bilateral BAHA ($n=3$)	*p*-value
Age, mean years (SD, range)[b]	11.3 (4.0, 6–16)	11.3 (5.5, 6–17)	
Sex: M : F	3 : 3	3 : 0	
Syndrome, number of patients:			
Suspected syndrome	1		
Branchio-oto-renal syndrome	1		
Goldenhar syndrome		1	
Crouzon	1		
Treacher Collins	1	1	
CHARGE (not defined)			
Type of malformation/surgery number of patients:			
Microtia and ear canal atresia	4[c]	2	
Ear canal atresia	1		
Bilateral modified radical		1	
Mastoidectomy			

Diagnosis, number of patients:		
Ear malformation	5	2
Chronic otitis media	1	1
Pure-tone thresholds, mean (SD) dB HL:	**All bilateral hearing loss patients ($n=9$)**	
AC PTA (M4) in better ear	61.3 (15.5)	
BC PTA (M4) in better ear	14.1 (12.7)	
AC PTA (M4) in worse ear	72.1 (12.1)	
BC PTA (M4) in worse ear	13.8 (10.7)	

All experienced BAHA users with a minimum use of 3 months

The majority of children with bilateral hearing loss had symmetrical maximal or near-maximal CHL

Results

Sound field average-tone thresholds, dB HL[b]	**Unaided**	**One BAHA**	**Two BAHAs**	***p*-value**
Unilateral BAHA ($n=6$), mean (SD, range)	53 (15, 25–68)	24 (5, 20–32)	N/A	$p=0.046$
Bilateral BAHAs ($n=3$), mean (SD, range)	62 (8, 55–70)	30 (5, 25–35)	25 (5, 20–30)	

Comments: in the unilateral BAHA, thresholds were significantly improved with one BAHA compared with none, but thresholds with hearing amplification still significantly differed from the norm of 20 dB HL ($p=0.028$). Trend similar in bilateral BAHA group, where fitting of a unilateral BAHA improved sound field threshold to almost normal. No extra gain was found with additional BAHA

Speech recognition in noise, median score (%)[d]	**Unaided**			**One BAHA**			**Two BAHAs**		
	S/N 0 dB	**S/N 4 dB**	**S/N 6 dB**	**S/N 0 dB**	**S/N 4 dB**	**S/N 6 dB**	**S/N 0 dB**	**S/N 4 dB**	**S/N 6 dB**
Unilateral BAHA ($n=6$)	0	0	0	87	92	98	N/A	N/A	N/A
Bilateral BAHA ($n=3$)	0	0	0	69	79	97	88	93	90

Comments: without hearing amplification all patients lacked open speech recognition. A trend towards lower test performance in children fitted with bilateral BAHAs when tested with one BAHA compared with two BAHAs was noted. Speech recognition ability diminished with increasing noise level, and with speech-in-noise approaching 0 dB

Localisation of sound[d]	**Unilateral BAHA ($n=6$)**				**Bilateral BAHAs ($n=3$)**			
	0.5 kHz		**3 kHz**		**0.5 kHz**		**3 kHz**	
	50 dB	**60 dB**	**50 dB**	**60 dB**	**50 dB**	**60 dB**	**50 dB**	**60 dB**
Correct score, mean % (chance level 20%)								
One BAHA	~20	~28	~28	~37	~20	~20	~16	~18
Two BAHAs	N/A	N/A	N/A	N/A	~50	~50	~50	~57
Lateralisations score, mean % (chance level 68%)								
One BAHA	~68	~70	~60	~72	~60	~68	~68	~56
Two BAHAs	N/A	N/A	N/A	N/A	~86	~94	~80	~96

Comments: authors note that scores could be obtained only with aids as the levels of stimulus presented in the speech recognition test were around 50 or 60 dB SPL, meaning that children with bilateral moderate-to-severe hearing loss received cues at subthreshold levels without their hearing amplification

Sound localisation ability was poor and close to chance level with one BAHA, but there was a trend towards improved sound localisation ability in children with bilaterally fitted BAHAs. Bilateral BAHAs improved sound lateralisation ability to near normal

IOI-HA, mean (SD)	**Unilateral BAHA ($n=6$)**	**Bilateral BAHAs ($n=2$[e])**	***p*-value**
Use	5.0 (0)	5.0	
Benefit	5.0 (1.0)	5.0	
Residual activity	4.2 (0.5)	4.0	
Limitation:			
Satisfaction	4.3 (1.0)	5.0	
Residual participation	4.2 (1.3)	3.0	
Impact on others	4.8 (0.4)	2.5	
QoL	4.8 (0.4)	5.0	

MAIS and MUSS, mean (SD)	Unilateral BAHA ($n=6$)	Bilateral BAHAs ($n=3$)[f]	*p*-value
Hearing-aid use	3.4 (1.3)	4.0	
Reaction to sounds	3.1 (0.9)	3.5	
Sound discrimination	3.5 (0.8)	3.8	
Verbal communication	3.8 (0.6)	3.7	
Speech intelligibility	3.1 (1.2)	3.3	

Methodological comments

- Allocation to treatment groups: two groups of participants (unilateral BAHAs and bilateral BAHAs) were each tested unaided and with one BAHA, and with two BAHAs for the bilateral BAHA group. Authors note that there may be some selection bias as patients fitted with bilateral BAHAs had actively acquired a second BAHA. Testing of unaided, with one aid and with two aids (in the bilateral BAHA group), appears to have occurred at the same session
- Blinding: NR
- Comparability of treatment groups: no females in bilateral BAHA group and small numbers
- Method of data analysis: results were analysed using descriptive methods and statistical analysis, including Kruskal-Wallis ANOVA, Friedman ANOVA, Sign test and Wilcoxon matched pairs test. Results of no hearing aid, one hearing aid or two hearing aids were analysed separately. No intention to treat, means and SD reported
- Sample size/power calculation: authors reported that a sample size of six participants was needed in each group for 80.0% power to detect a difference in means of 6.0% (speech recognition) and around 20.0% (sound localisation) between groups, assuming that the common SDs are 3.0% and 10.0% respectively, using a two-group *t*-test with a two-sided significance level of 0.05. To reach the same power within groups, a sample size of five is reported to be required, using a paired *t*-test with a two-sided significant level of 0.05. However, there are only three participants in the bilateral BAHA group, which is therefore underpowered
- Attrition/dropout: response rate for questionnaires is 80% and 87%

General comments

- Generalisability: children with moderate-to-severe hearing loss
- Outcome measures: the Swedish speech material is not validated for paediatric use, while it is noted that the MAIS, MUSS and IOI-HA are validated questionnaires
- Inter-centre variability: NR
- Conflict of interests: NR

ANOVA, analysis of variance; F, female; M, male; N/A, not applicable; NR, not reported.
a Data on normal controls and unilateral hearing loss presented but not extracted.
b Means and SD calculated by reviewer.
c One patient had microtia and ear canal atresia (r) and ear canal stenosis (l).
d Data are estimated from figures by reviewer.
e No SD as on 2/3 individuals responded. Scores > 3 indicate success of the hearing aid fitting compared with the unaided situation; 87% response rate to questionnaire, results were in accordance with success of treatment in all groups compared with reference group data.[45]
f No SD owing to small participant number. Eighty per cent response rate to questionnaire. States that since there was a good congruence between parents and teachers, the results are presented together. It is unclear how this was done.

Quality assessment for primary studies

A. Selection bias					
1. Are the individuals selected to participate in the study likely to be representative of the target population?	Very likely	Somewhat likely x	Not likely	Can't tell	
2. What percentage of selected individuals agreed to participate?	80–100%	60–79%	< 60%	Not applicable	Can't tell x
Summary of selection bias (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
B. Study design					
1. What was the study design? (Please tick appropriate and specify design in No. 7)	RCT				
	Controlled clinical trial				
	Cohort analytic (two group pre and post)				
	Case–control				
	Cohort [one group pre and post (before and after)]				
	Interrupted time series				
	Other – specify: audiological comparison study			x reviewer's opinion	
	Can't tell				
2. Was the study described as randomised?	Yes	No x			
If answer to No. 2 is no, go to section on Section C Confounders. If answer yes, answer No. 3 and No. 4 below					
3. If answer was yes, was the method of randomisation described?	Yes	No			
4. If answer was yes, was the method appropriate?	Yes	No			
Summary of study design (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
C. Confounders					
Two groups: are confounders reported AND controlled for in the analysis? OR if one group: are potential confounding variables reported?	Yes	No x	Can't tell		
Summary of confounders (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
D. Blinding					
1. Was the outcome assessor aware of the intervention or exposure status of participants?	Yes x	No	Can't tell		
2. Were the study participants aware of the research question?	Yes x	No	Can't tell		
Summary of blinding (methodological strength of study)	*Strong*	*Moderate*	*Weak* x		
E. Data collection methods					
1. Were data collection tools shown to be valid?	Yes x	No	Can't tell		
2. Were data collection tools shown to be reliable?	Yes x	No	Can't tell		
Summary of data collection (methodological strength of study)	*Strong* x	*Moderate*	*Weak*		

F. Withdrawals and dropouts				
1. Were withdrawals and dropouts reported in terms of numbers and reasons per group?	Yes	No	Can't tell x	
2. Indicate the percentage of participants completing the study (if the percentage differs by groups, record the lowest)	80–100%	60–79%	< 60%	Can't tell x
Summary of withdrawals and dropouts (methodological strength of study)	*Strong*	*Moderate*	*Weak* x	
G. Intervention integrity				
1. Was the consistency of the intervention measured?	Yes	No x	Can't tell	
H. Analysis				
1. Are the statistical methods appropriate for the study design?	Yes x	No	Can't tell	
2. Does the study report how missing data are dealt with in the analysis?	Yes	No x	Can't tell	
Global rating for study[a] (Overall methodological strength of study – based on sections A–F)	**Strong**	**Moderate**	**Weak x**	

a Strong = four strong ratings with no weak ratings; moderate = less than four strong ratings and one weak rating; weak = two or more weak ratings.

Appendix 11

Inclusion/exclusion criteria for systematic review of quality of life

	Yes	Unclear	No
1. Before and after study, or randomised comparison			Exclude
(a) reporting health-state utilities			
(b) quality of life QoL/health-related QoL			
(c) reviews 1(a), 1(b) or both			
2. Intervention			
■ BAHA			
3. Population/indication			
■ adults or children with a conductive or mixed hearing loss			
Retrieve?			
If 1(a) or 1(b) = yes; 2 = yes; and 3 = yes THEN include in review			

Appendix 12

Deterministic sensitivity analyses: full results

Results for each parameter are provided on two lines – the first gives the results at the lower limit of the output parameter and the second gives the results at the upper limit.

Cost analysis

BCHA device cost = £117

DSA – paediatric cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	765	17,513	16,747
	0.04310	765	17,518	16,753
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	765	17,509	16,743
	0.13826	765	17,525	16,760
Probability of failure to integrate (0.006)	0.00015	765	17,513	16,748
	0.03291	765	17,501	16,736
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	765	17,840	17,075
	0.06820	765	17,445	16,680
Probability of re-operation (0.9474)	0.73972	765	16,668	15,902
	0.99867	765	17,732	16,966
Cost of initial ENT consultation (£131.69)	98.77	765	17,481	16,715
	164.61	765	17,547	16,781
Cost of audiological assessment (£57.48)	43.11	765	17,499	16,734
	71.85	765	17,528	16,763
Cost of ENT multiprofessional assessment (£147.36)	110.52	765	17,477	16,711
	184.21	765	17,551	16,785
Cost of day-case surgery for implantation (£4009.14)	3006.86	765	16,141	15,376
	5011.43	765	18,887	18,121
Cost of fixture and abutment (£830.00)	622.50	765	17,338	16,573
	1037.50	765	17,689	16,924
Cost of surgical consumables (£159.50)	119.63	765	17,460	16,694
	199.38	765	17,568	16,802
Cost of follow-up ENT consultations (£90.93)	68.20	765	17,424	16,658
	113.66	765	17,604	16,839
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	765	17,493	16,727
	81.00	765	17,535	16,770
Cost of audiology follow-up in year of surgery (£50.17)	37.62	765	17,481	16,715
	62.71	765	17,547	16,781
Cost of BAHA sound processor (£2191.25)	1820.00	765	17,147	16,381
	2995.00	765	18,308	17,543

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	765	16,346	15,581
	1000.00	765	19,953	19,188
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	765	17,459	16,694
	829.58	765	17,569	16,803

DSA – paediatric cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	765	17,586	16,820
	0.04310	765	17,591	16,826
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	765	17,582	16,816
	0.13826	765	17,598	16,833
Probability of failure to integrate (0.006)	0.00015	765	17,586	16,821
	0.03291	765	17,583	16,818
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	765	17,862	17,097
	0.06820	765	17,529	16,764
Probability of re-operation (0.9474)	0.73972	765	16,819	16,054
	0.99867	765	17,785	17,019
Cost of initial ENT consultation (£131.69)	98.77	765	17,554	16,788
	164.61	765	17,620	16,854
Cost of audiological assessment (£57.48)	43.11	765	17,572	16,807
	71.85	765	17,601	16,836
Cost of ENT multiprofessional assessment (£147.36)	110.52	765	17,550	16,785
	184.21	765	17,624	16,858
Cost of day-case surgery for implantation (£4009.14)	3006.86	765	16,214	15,449
	5011.43	765	18,960	18,194
Cost of fixture and abutment (£830.00)	622.50	765	17,411	16,646
	1037.50	765	17,762	16,997
Cost of surgical consumables (£159.50)	119.63	765	17,533	16,767
	199.38	765	17,641	16,875
Cost of follow-up ENT consultations (£90.93)	68.20	765	17,497	16,731
	113.66	765	17,677	16,912
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	765	17,566	16,800
	81.00	765	17,608	16,843
Cost of audiology follow-up in year of surgery (£50.17)	37.62	765	17,554	16,788
	62.71	765	17,620	16,854
Cost of BAHA sound processor (£2191.25)	1820.00	765	17,220	16,454
	2995.00	765	18,381	17,616
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	765	16,419	15,654
	1000.00	765	20,026	19,261
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	765	17,532	16,767
	829.58	765	17,642	16,876

DSA – adult cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	751	14,532	13,781
	0.04310	751	14,538	13,786
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	751	14,529	13,777
	0.13826	751	14,545	13,793
Probability of failure to integrate (0.006)	0.00015	751	14,533	13,782
	0.03291	751	14,531	13,779
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	751	14,828	14,077
	0.06820	751	14,472	13,720
Probability of re-operation (0.9474)	0.73972	751	13,877	13,125
	0.99867	751	14,702	13,951
Cost of initial ENT consultation (£110.78)	83.09	751	14,506	13,754
	138.48	751	14,561	13,810
Cost of audiological assessment (£57.48)	43.11	751	14,519	13,768
	71.85	751	14,548	13,796
Cost of ENT multiprofessional assessment (£147.36)	110.52	751	14,497	13,745
	184.21	751	14,570	13,819
Cost of day-case surgery for implantation (£2004.57)	1503.43	751	13,842	13,090
	2505.71	751	15,225	14,474
Cost of fixture and abutment (£830.00)	622.50	751	14,359	13,607
	1037.50	751	14,708	13,957
Cost of surgical consumables (£159.50)	119.63	751	14,480	13,728
	199.38	751	14,587	13,836
Cost of follow-up ENT consultations (£72.11)	54.09	751	14,462	13,711
	90.14	751	14,604	13,853
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	751	14,512	13,761
	81.00	751	14,555	13,803
Cost of audiology follow-up in year of surgery (£50.17)	37.62	751	14,501	13,749
	62.71	751	14,566	13,815
Cost of BAHA sound processor (£2191.25)	1820.00	751	14,168	13,416
	2995.00	751	15,325	14,574
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	751	13,388	12,636
	1000.00	751	16,927	16,176
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	751	14,480	13,728
	829.58	751	14,587	13,836

DSA – adult cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	751	14,604	13,852
	0.04310	751	14,609	13,858
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	751	14,600	13,849
	0.13826	751	14,616	13,865
Probability of failure to integrate (0.006)	0.00015	751	14,605	13,853
	0.03291	751	14,611	13,860

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	751	14,850	14,098
	0.06820	751	14,553	13,802
Probability of re-operation (0.9474)	0.73972	751	14,024	13,273
	0.99867	751	14,754	14,003
Cost of initial ENT consultation (£110.78)	83.09	751	14,577	13,826
	138.48	751	14,633	13,881
Cost of audiological assessment (£57.48)	43.11	751	14,590	13,839
	71.85	751	14,619	13,868
Cost of ENT multiprofessional assessment (£147.36)	110.52	751	14,568	13,817
	184.21	751	14,642	13,890
Cost of day-case surgery for implantation (£2004.57)	1503.43	751	13,913	13,161
	2505.71	751	15,297	14,545
Cost of fixture and abutment (£830.00)	622.50	751	14,430	13,679
	1037.50	751	14,780	14,028
Cost of surgical consumables (£159.50)	119.63	751	14,551	13,800
	199.38	751	14,658	13,907
Cost of follow-up ENT consultations (£72.11)	54.09	751	14,534	13,782
	90.14	751	14,676	13,924
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	751	14,584	13,832
	81.00	751	14,626	13,875
Cost of audiology follow-up in year of surgery (£50.17)	37.62	751	14,572	13,821
	62.71	751	14,637	13,886
Cost of BAHA sound processor (£2191.25)	1820.00	751	14,239	13,488
	2995.00	751	15,397	14,645
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	751	13,459	12,707
	1000.00	751	16,999	16,247
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	751	14,551	13,800
	829.58	751	14,658	13,907

BCHA device cost = £183

DSA – paediatric cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	934	17,513	16,579
	0.04310	934	17,518	16,584
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	934	17,509	16,575
	0.13826	934	17,525	16,591
Probability of failure to integrate (0.006)	0.00015	934	17,513	16,580
	0.03291	934	17,501	16,567
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	934	17,840	16,906
	0.06820	934	17,445	16,512
Probability of re-operation (0.9474)	0.73972	934	16,668	15,734
	0.99867	934	17,732	16,798
Cost of initial ENT consultation (£131.69)	98.77	934	17,481	16,547
	164.61	934	17,547	16,613
Cost of audiological assessment (£57.48)	43.11	934	17,499	16,566
	71.85	934	17,528	16,594

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Cost of ENT multiprofessional assessment (£147.36)	110.52	934	17,477	16,543
	184.21	934	17,551	16,617
Cost of day-case surgery for implantation (£4009.14)	3006.86	934	16,141	15,207
	5011.43	934	18,887	17,953
Cost of fixture and abutment (£830.00)	622.50	934	17,338	16,404
	1037.50	934	17,689	16,756
Cost of surgical consumables (£159.50)	119.63	934	17,460	16,526
	199.38	934	17,568	16,634
Cost of follow-up ENT consultations (£90.93)	68.20	934	17,424	16,490
	113.66	934	17,604	16,670
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	934	17,493	16,559
	81.00	934	17,535	16,601
Cost of audiology follow-up in year of surgery (£50.17)	37.62	934	17,481	16,547
	62.71	934	17,547	16,613
Cost of BAHA sound processor (£2191.25)	1820.00	934	17,147	16,213
	2995.00	934	18,308	17,375
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	934	16,346	15,412
	1000.00	934	19,953	19,019
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	934	17,459	16,525
	829.58	934	17,569	16,635

DSA – paediatric cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	934	17,617	16,683
	0.04310	934	17,622	16,688
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	934	17,613	16,679
	0.13826	934	17,629	16,695
Probability of failure to integrate (0.006)	0.00015	934	17,618	16,684
	0.03291	934	17,618	16,684
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	934	17,872	16,938
	0.06820	934	17,565	16,631
Probability of re-operation (0.9474)	0.73972	934	16,884	15,951
	0.99867	934	17,807	16,873
Cost of initial ENT consultation (£131.69)	98.77	934	17,585	16,651
	164.61	934	17,651	16,717
Cost of audiological assessment (£57.48)	43.11	934	17,603	16,670
	71.85	934	17,632	16,698
Cost of ENT multiprofessional assessment (£147.36)	110.52	934	17,581	16,647
	184.21	934	17,655	16,721
Cost of day-case surgery for implantation (£4009.14)	3006.86	934	16,245	15,311
	5011.43	934	18,991	18,057
Cost of fixture and abutment (£830.00)	622.50	934	17,442	16,508
	1037.50	934	17,794	16,860
Cost of surgical consumables (£159.50)	119.63	934	17,564	16,630
	199.38	934	17,672	16,738

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Cost of follow-up ENT consultations (£90.93)	68.20	934	17,528	16,594
	113.66	934	17,708	16,774
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	934	17,597	16,663
	81.00	934	17,639	16,705
Cost of audiology follow-up in year of surgery (£50.17)	37.62	934	17,585	16,651
	62.71	934	17,651	16,717
Cost of BAHA sound processor (£2191.25)	1820.00	934	17,251	16,317
	2995.00	934	18,412	17,479
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	934	16,450	15,516
	1000.00	934	20,057	19,123
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	934	17,563	16,629
	829.58	934	17,673	16,739

DSA – adult cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	917	14,532	13,616
	0.04310	917	14,538	13,621
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	917	14,529	13,612
	0.13826	917	14,545	13,628
Probability of failure to integrate (0.006)	0.00015	917	14,533	13,617
	0.03291	917	14,531	13,614
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	917	14,828	13,912
	0.06820	917	14,472	13,555
Probability of re-operation (0.9474)	0.73972	917	13,877	12,960
	0.99867	917	14,702	13,786
Cost of initial ENT consultation (£110.78)	83.09	917	14,506	13,589
	138.48	917	14,561	13,645
Cost of audiological assessment (£57.48)	43.11	917	14,519	13,603
	71.85	917	14,548	13,631
Cost of ENT multiprofessional assessment (£147.36)	110.52	917	14,497	13,580
	184.21	917	14,570	13,654
Cost of day-case surgery for implantation (£2004.57)	1503.43	917	13,842	12,925
	2505.71	917	15,225	14,309
Cost of fixture and abutment (£830.00)	622.50	917	14,359	13,442
	1037.50	917	14,708	13,792
Cost of surgical consumables (£159.50)	119.63	917	14,480	13,563
	199.38	917	14,587	13,671
Cost of follow-up ENT consultations (£72.11)	54.09	917	14,462	13,546
	90.14	917	14,604	13,688
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	917	14,512	13,596
	81.00	917	14,555	13,638
Cost of audiology follow-up in year of surgery (£50.17)	37.62	917	14,501	13,584
	62.71	917	14,566	13,650
Cost of BAHA sound processor (£2191.25)	1820.00	917	14,168	13,251
	2995.00	917	15,325	14,409

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	917	13,388	12,471
	1000.00	917	16,927	16,011
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	917	14,480	13,563
	829.58	917	14,587	13,671

DSA – adult cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	917	14,634	13,717
	0.04310	917	14,639	13,723
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	917	14,630	13,714
	0.13826	917	14,646	13,730
Probability of failure to integrate (0.006)	0.00015	917	14,635	13,719
	0.03291	917	14,645	13,728
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	917	14,859	13,942
	0.06820	917	14,588	13,672
Probability of re-operation (0.9474)	0.73972	917	14,088	13,171
	0.99867	917	14,776	13,859
Cost of initial ENT consultation (£110.78)	83.09	917	14,607	13,691
	138.48	917	14,663	13,746
Cost of audiological assessment (£57.48)	43.11	917	14,621	13,704
	71.85	917	14,649	13,733
Cost of ENT multiprofessional assessment (£147.36)	110.52	917	14,598	13,682
	184.21	917	14,672	13,755
Cost of day-case surgery for implantation (£2004.57)	1503.43	917	13,943	13,027
	2505.71	917	15,327	14,411
Cost of fixture and abutment (£830.00)	622.50	917	14,460	13,544
	1037.50	917	14,810	13,893
Cost of surgical consumables (£159.50)	119.63	917	14,582	13,665
	199.38	917	14,689	13,772
Cost of follow-up ENT consultations (£72.11)	54.09	917	14,564	13,648
	90.14	917	14,706	13,790
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	917	14,614	13,697
	81.00	917	14,656	13,740
Cost of audiology follow-up in year of surgery (£50.17)	37.62	917	14,602	13,686
	62.71	917	14,668	13,751
Cost of BAHA sound processor (£2191.25)	1820.00	917	14,269	13,353
	2995.00	917	15,427	14,510
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	917	13,489	12,573
	1000.00	917	17,029	16,113
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	917	14,581	13,665
	829.58	917	14,689	13,772

BCHA device cost = £350

DSA – paediatric cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1360	17,513	16,153
	0.04310	1360	17,518	16,158
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1360	17,509	16,149
	0.13826	1360	17,525	16,166
Probability of failure to integrate (0.006)	0.00015	1360	17,513	16,154
	0.03291	1360	17,501	16,142
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1360	17,840	16,480
	0.06820	1360	17,445	16,086
Probability of re-operation (0.9474)	0.73972	1360	16,668	15,308
	0.99867	1360	17,732	16,372
Cost of initial ENT consultation (£131.69)	98.77	1360	17,481	16,121
	164.61	1360	17,547	16,187
Cost of audiological assessment (£57.48)	43.11	1360	17,499	16,140
	71.85	1360	17,528	16,169
Cost of ENT multiprofessional assessment (£147.36)	110.52	1360	17,477	16,117
	184.21	1360	17,551	16,191
Cost of day-case surgery for implantation (£4009.14)	3006.86	1360	16,141	14,781
	5011.43	1360	18,887	17,527
Cost of fixture and abutment (£830.00)	622.50	1360	17,338	15,979
	1037.50	1360	17,689	16,330
Cost of surgical consumables (£159.50)	119.63	1360	17,460	16,100
	199.38	1360	17,568	16,208
Cost of follow-up ENT consultations (£90.93)	68.20	1360	17,424	16,064
	113.66	1360	17,604	16,244
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1360	17,493	16,133
	81.00	1360	17,535	16,176
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1360	17,481	16,121
	62.71	1360	17,547	16,187
Cost of BAHA sound processor (£2191.25)	1820.00	1360	17,147	15,787
	2995.00	1360	18,308	16,949
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1360	16,346	14,987
	1000.00	1360	19,953	18,594
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1360	17,459	16,099
	829.58	1360	17,569	16,209

DSA – paediatric cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1360	17,695	16,336
	0.04310	1360	17,701	16,341
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1360	17,691	16,332
	0.13826	1360	17,708	16,348

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of failure to integrate (0.006)	0.00015	1360	17,696	16,337
	0.03291	1360	17,706	16,346
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1360	17,896	16,536
	0.06820	1360	17,654	16,295
Probability of re-operation (0.9474)	0.73972	1360	17,049	15,689
	0.99867	1360	17,863	16,504
Cost of initial ENT consultation (£131.69)	98.77	1360	17,663	16,304
	164.61	1360	17,729	16,370
Cost of audiological assessment (£57.48)	43.11	1360	17,682	16,322
	71.85	1360	17,711	16,351
Cost of ENT multiprofessional assessment (£147.36)	110.52	1360	17,659	16,300
	184.21	1360	17,733	16,374
Cost of day-case surgery for implantation (£4009.14)	3006.86	1360	16,324	14,964
	5011.43	1360	19,069	17,710
Cost of fixture and abutment (£830)	622.50	1360	17,521	16,161
	1037.50	1360	17,872	16,512
Cost of surgical consumables (£159.50)	119.63	1360	17,642	16,283
	199.38	1360	17,750	16,391
Cost of follow-up ENT consultations (£90.93)	68.20	1360	17,606	16,247
	113.66	1360	17,787	16,427
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1360	17,675	16,316
	81.00	1360	17,718	16,358
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1360	17,663	16,304
	62.71	1360	17,729	16,370
Cost of BAHA sound processor (£2191.25)	1820.00	1360	17,329	15,970
	2995.00	1360	18,491	17,131
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1360	16,529	15,169
	1000.00	1360	20,136	18,776
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1360	17,641	16,282
	829.58	1360	17,751	16,392

DSA – adult cases, with treatment failures not switching to alternative hearing aid

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1334	14,532	13,198
	0.04310	1334	14,538	13,203
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1334	14,529	13,194
	0.13826	1334	14,545	13,210
Probability of failure to integrate (0.006)	0.00015	1334	14,533	13,199
	0.03291	1334	14,531	13,196
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1334	14,828	13,494
	0.06820	1334	14,472	13,137
Probability of re-operation (0.9474)	0.73972	1334	13,877	12,542
	0.99867	1334	14,702	13,368
Cost of initial ENT consultation (£110.78)	83.09	1334	14,506	13,171
	138.48	1334	14,561	13,227

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Cost of audiological assessment (£57.48)	43.11	1334	14,519	13,185
	71.85	1334	14,548	13,213
Cost of ENT multiprofessional assessment (£147.36)	110.52	1334	14,497	13,162
	184.21	1334	14,570	13,236
Cost of day-case surgery for implantation (£2004.57)	1503.43	1334	13,842	12,507
	2505.71	1334	15,225	13,891
Cost of fixture and abutment (£830.00)	622.50	1334	14,359	13,024
	1037.50	1334	14,708	13,374
Cost of surgical consumables (£159.50)	119.63	1334	14,480	13,145
	199.38	1334	14,587	13,253
Cost of follow-up ENT consultations (£72.11)	54.09	1334	14,462	13,128
	90.14	1334	14,604	13,270
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1334	14,512	13,178
	81.00	1334	14,555	13,220
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1334	14,501	13,166
	62.71	1334	14,566	13,232
Cost of BAHA sound processor (£2191.25)	1820.00	1334	14,168	12,833
	2995.00	1334	15,325	13,991
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1334	13,388	12,053
	1000.00	1334	16,927	15,593
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1334	14,480	13,145
	829.58	1334	14,587	13,253

DSA – adult cases, with treatment failures switching to alternative hearing aid (BCHA)

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	1334	14,711	13,376
	0.04310	1334	14,716	13,381
Probability of surgical reduction of skin growth/soft tissue thickening around abutment (0.0848)	0.04716	1334	14,707	13,372
	0.13826	1334	14,723	13,388
Probability of failure to integrate (0.006)	0.00015	1334	14,712	13,377
	0.03291	1334	14,731	13,396
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	1334	14,883	13,548
	0.06820	1334	14,676	13,341
Probability of re-operation (0.9474)	0.73972	1334	14,248	12,914
	0.99867	1334	14,831	13,497
Cost of initial ENT consultation (£110.78)	83.09	1334	14,684	13,350
	138.48	1334	14,739	13,405
Cost of audiological assessment (£57.48)	43.11	1334	14,697	13,363
	71.85	1334	14,726	13,392
Cost of ENT multiprofessional assessment (£147.36)	110.52	1334	14,675	13,340
	184.21	1334	14,749	13,414
Cost of day-case surgery for implantation (£2004.57)	1503.43	1334	14,020	12,685
	2505.71	1334	15,404	14,069
Cost of fixture and abutment (£830.00)	622.50	1334	14,537	13,203
	1037.50	1334	14,886	13,552

Parameter (base-case value)	Input value	BCHA (£)	BAHA (£)	Incremental cost per case (£)
Cost of surgical consumables (£159.50)	119.63	1334	14,658	13,324
	199.38	1334	14,765	13,431
Cost of follow-up ENT consultations (£72.11)	54.09	1334	14,641	13,306
	90.14	1334	14,783	13,448
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	1334	14,691	13,356
	81.00	1334	14,733	13,398
Cost of audiology follow-up in year of surgery (£50.17)	37.62	1334	14,679	13,345
	62.71	1334	14,744	13,410
Cost of BAHA sound processor (£2191.25)	1820.00	1334	14,346	13,012
	2995.00	1334	15,504	14,169
Cost of BAHA sound processor maintenance plan (£736.25)	610.00	1334	13,566	12,231
	1000.00	1334	17,106	15,771
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	1334	14,658	13,324
	829.58	1334	14,765	13,431

Cost-effectiveness analysis

BCHA device cost = £117

DSA – impact on ICER

		ICER (£ per QALY gained)			
		Paediatric (P)		Adult (A)	
Input parameter (base-case value)	Input value	QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	121,353	56,568	102,022	47,557
	0.04310	121,393	56,587	102,062	47,576
Probability of surgical reduction of skin growth/thickening around abutment (0.0848)	0.04716	121,327	56,556	101,995	47,544
	0.13826	121,444	56,610	102,113	47,600
Probability of failure to integrate (0.006)	0.00015	121,457	56,617	102,112	47,599
	0.03291	126,383	58,913	106,330	49,565
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	119,273	55,599	100,408	46,805
	0.06820	121,817	56,784	102,384	47,726
Probability of re-operation (0.9474)	0.73972	121,480	56,628	102,491	47,776
	0.99867	121,346	56,565	101,928	47,513
Cost of initial ENT consultation [£131.69 (P) and £110.78 (A)]	98.77 (P)	121,125	56,462	101,827	47,466
	83.09 (A)				
	164.61 (P)	121,600	56,683	102,235	47,656
	138.48 (A)				
Cost of audiological assessment (£57.48)	43.11	121,259	56,524	101,925	47,512
	71.85	121,466	56,621	102,137	47,611
Cost of ENT multiprofessional assessment (£147.36)	110.52	121,097	56,449	101,760	47,435
	184.21	121,628	56,697	102,302	47,688
Cost of day-case surgery for implantation (£2004.57)	1503.43	111,458	51,956	96,935	45,186
	2505.71	131,267	61,190	107,127	49,937
Cost of fixture and abutment (£830.00)	622.50	120,095	55,982	100,744	46,961
	1037.50	122,630	57,163	103,318	48,161
Cost of surgical consumables (£159.50)	119.63	120,974	56,391	101,636	47,377
	199.38	121,751	56,754	102,426	47,745

Input parameter (base-case value)	Input value	ICER (£ per QALY gained)			
		Paediatric (P)		Adult (A)	
		QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Cost of follow-up ENT consultations [£90.93 (P) and £72.11 (A)]	68.20 (P)	120,712	56,269	101,508	47,318
	54.09 (A)				
	113.66 (P)	122,013	56,876	102,554	47,805
	90.14 (A)				
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	121,209	56,501	101,875	47,489
	81.00	121,516	56,644	102,186	47,634
Cost of audiology follow-up in year of surgery (£50.17)	37.62	121,125	56,462	101,790	47,449
	62.71	121,600	56,683	102,272	47,673
Cost of BAHA sound processor (£2191.25)	1820.00	118,715	55,338	99,337	46,306
	2995.00	127,094	59,245	107,863	50,280
Cost of BAHA sound processor maintenance plan (£735.25)	610.00	112,938	52,646	93,591	43,627
	1000.00	138,962	64,776	119,663	55,780
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	120,967	56,388	101,636	47,377
	829.58	121,758	56,757	102,426	47,745
Proportion of cohort using BCHA for >8 hours per day (0.90)	0.683	37,644	17,548	31,648	14,752
	0.988	1,236,900	576,576	1,039,880	484,736
Proportion using BCHA at lower limit of 95% CI, 0.683, and proportion using BAHA at lower limit of 95% CI, 0.832 (BCHA = 0.90; BAHA = 1.00)	0.683	83,663	38,999	70,328	32,783
	0.832				

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Chapter 4, Quality of life*, for full details.

BCHA device cost = £183
DSA – impact on ICER

Input parameter (base-case value)	Input value	ICER (£ per QALY gained)			
		Paediatric (P)		Adult (A)	
		QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	120,363	56,107	101,028	47,094
	0.04310	120,402	56,125	101,068	47,113
Probability of surgical reduction of skin growth/thickening around abutment (0.0848)	0.04716	120,337	56,094	101,001	47,081
	0.13826	120,453	56,149	101,120	47,137
Probability of failure to integrate (0.006)	0.00015	120,467	56,155	101,118	47,136
	0.03291	125,379	58,445	105,324	49,096
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	118,166	55,083	99,299	46,288
	0.06820	120,852	56,335	101,416	47,274
Probability of re-operation (0.9474)	0.73972	120,699	56,264	101,706	47,410
	0.99867	120,305	56,080	100,884	47,027
Cost of initial ENT consultation [£131.69 (P) and £110.78 (A)]	98.77 (P)	120,135	56,000	100,834	47,003
	83.09 (A)				
	164.61 (P)	120,610	56,222	101,242	47,193
	138.48 (A)				
Cost of audiological assessment (£57.48)	43.11	120,268	56,063	100,932	47,049
	71.85	120,476	56,159	101,143	47,148

Input parameter (base-case value)	Input value	ICER (£ per QALY gained)			
		Paediatric (P)		Adult (A)	
		QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Cost of ENT multiprofessional assessment (£147.36)	110.52	120,106	55,987	100,766	46,972
	184.21	120,638	56,235	101,309	47,225
Cost of day-case surgery for implantation (£2004.57)	1503.43	110,468	51,494	95,941	44,723
	2505.71	130,277	60,728	106,134	49,474
Cost of fixture and abutment (£830.00)	622.50	119,105	55,520	99,751	46,498
	1037.50	121,640	56,702	102,324	47,698
Cost of surgical consumables (£159.50)	119.63	119,983	55,930	100,643	46,914
	199.38	120,761	56,292	101,432	47,282
Cost of follow-up ENT consultations [£90.93 (P) and £72.11 (A)]	68.20 (P) 54.09 (A)	119,721	55,808	100,515	46,855
	113.66 (P) 90.14 (A)	121,023	56,414	101,560	47,342
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	120,219	56,039	100,882	47,026
	81.00	120,526	56,183	101,193	47,171
Cost of audiology follow-up in year of surgery (£50.17)	37.62	120,135	56,000	100,797	46,986
	62.71	120,610	56,222	101,278	47,210
Cost of BAHA sound processor (£2191.25)	1820.00	117,725	54,877	98,344	45,843
	2995.00	126,104	58,783	106,869	49,817
Cost of BAHA sound processor maintenance plan (£735.25)	610.00	111,948	52,184	92,598	43,164
	1000.00	137,971	64,315	118,670	55,317
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	119,977	55,927	100,643	46,914
	829.58	120,768	56,295	101,433	47,282
Proportion of cohort using BCHA for >8 hours per day (0.90)	0.683	37,337	17,404	31,340	14,609
	0.988	1,226,807	571,871	1,029,756	480,016
Proportion using BCHA at lower limit of 95% CI, 0.683, and proportion using BAHA at lower limit of 95% CI, 0.832 (BCHA = 0.90; BAHA = 1.00)	0.683 0.832	82,980	38,681	69,643	32,464

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Chapter 4, Quality of life* for full details.

BCHA device cost = £350

DSA – impact on ICER

Input parameter (base-case value)	Input value	ICER (£ per QALY gained)			
		Paediatric (P)		Adult (A)	
		QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Probability of bleeding within 24 hours of surgery (0.0121)	0.00147	117,857	54,939	98,515	45,922
	0.04310	117,897	54,957	98,555	45,941
Probability of surgical reduction of skin growth/thickening around abutment (0.0848)	0.04716	117,831	54,926	98,488	45,910
	0.13826	117,948	54,981	98,606	45,965
Probability of failure to integrate (0.006)	0.00015	117,960	54,987	98,604	45,964
	0.03291	122,840	57,262	102,776	47,909
Probability of intolerable pain requiring removal of abutment and flange fixture (0.0272)	0.00746	115,365	53,777	96,491	44,979
	0.06820	118,410	55,197	98,966	46,133

Input parameter (base-case value)	Input value	ICER (£ per QALY gained)			
		Paediatric (P)		Adult (A)	
		QALY1[a]	QALY2[b]	QALY1[a]	QALY2[b]
Probability of re-operation (0.9474)	0.73972	118,724	55,343	99,719	46,484
	0.99867	117,671	54,852	98,244	45,796
Cost of initial ENT consultation [£131.69 (P) and £110.78 (A)]	98.77 (P)	117,629	54,832	98,320	45,832
	83.09 (A)				
	164.61 (P)	118,104	55,054	98,728	46,022
	138.48 (A)				
Cost of audiological assessment (£57.48)	43.11	117,763	54,895	98,418	45,877
	71.85	117,970	54,991	98,630	45,976
Cost of ENT multiprofessional assessment (£147.36)	110.52	117,601	54,819	98,253	45,800
	184.21	118,132	55,067	98,795	46,053
Cost of day-case surgery for implantation (£2004.57)	1503.43	107,962	50,326	93,428	43,551
	2505.71	127,771	59,560	103,620	48,302
Cost of fixture and abutment (£830.00)	622.50	116,599	54,352	97,237	45,327
	1037.50	119,134	55,534	99,811	46,527
Cost of surgical consumables (£159.50)	119.63	117,478	54,762	98,129	45,743
	199.38	118,255	55,124	98,919	46,111
Cost of follow-up ENT consultations [£90.93 (P) and 72.11 (A)]	68.20 (P)	117,216	54,640	98,001	45,683
	54.09 (A)				
	113.66 (P)	118,517	55,246	99,047	46,170
	90.14 (A)				
Cost of audiological consultation to fit/commission sound processor (£64.80)	48.60	117,713	54,871	98,369	45,854
	81.00	118,020	55,014	98,680	45,999
Cost of audiology follow-up in year of surgery (£50.17)	37.62	117,629	54,832	98,283	45,814
	62.71	118,104	55,054	98,765	46,039
Cost of BAHA sound processor (£2191.25)	1820.00	115,219	53,709	95,830	44,671
	2995.00	123,598	57,615	104,356	48,645
Cost of BAHA sound processor maintenance plan (£735.25)	610.00	109,442	51,016	90,084	41,992
	1000.00	135,466	63,147	116,156	54,146
Cost of surgical revision for grade 3 skin reaction (£663.66)	497.75	117,471	54,759	98,129	45,742
	829.58	118,262	55,127	98,919	46,111
Proportion of cohort using BCHA for >8 hours per day (0.90)	0.683	36,560	17,042	30,560	14,245
	0.988	1,201,269	559,966	1,004,140	468,075
Proportion using BCHA at lower limit of 95% CI, 0.683, and proportion using BAHA at lower limit of 95% CI, 0.832 (BCHA = 0.90; BAHA = 1.00)	0.683	81,253	37,876	67,911	31,656
	0.832				

a Moving from level 6 of the HUI hearing attribute to level 5.
b Moving from level 6 of the HUI hearing attribute to level 3.
See *Chapter 4, Quality of life* for full details.

Appendix 13

Variables included in the probabilistic sensitivity analyses

Results for each parameter are provided on two lines – the first gives the results at the lower limit of the output parameter and the second gives the results at the upper limit.

Input variable	Mean	Variability	Distribution	Sampling method/parameters of distribution
BCHA cost (£)	N/A	N/A	N/A	Sample from table of possible values (117, 183, 250, 350) with equal probability
BAHA sound processor cost (£)	N/A	N/A	N/A	Sample from table of possible values (1820, 1980, 1970, 2995) with equal probability
BAHA sound processor maintenance cost (£)	N/A	N/A	N/A	Sample from table of possible values (610, 665, 670, 1000) with equal probability
BAHA implant day-case surgery (£)	1918	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 15.2136$
Loss of bone integration	0.741 (at 7 years)	95% CI of Kaplan–Meier estimate (0.4868 to 0.8767)	Beta	$\alpha = 14.3726$; $\beta = 5.0210$
Difference in use of hearing aid (BAHA–BCHA)	0.1	N/A	Beta	$r = 2$; $n = 20$
Probability of initial failure of bone integration	0.0060	N/A	Beta	$r = 1$; $n = 167$
Probability of failure owing to intolerable pain	0.0272	N/A	Beta	$r = 4$; $n = 147$
Probability of re-operation following failure of bone integration or removal of implant due to grade 4 skin reaction	0.9474	N/A	Beta	$r = 18$; $n = 19$
Probability of grade 1/2 skin reaction	0.1188	95% CI (0.0665 to 0.1960)	Beta	$\alpha = 11.4037$; $\beta = 84.5776$
Probability of grade 3 skin reaction	0.0396	95% CI (0.0129 to 0.0924)	Beta	$\alpha = 3.6565$; $\beta = 88.6690$
Probability of grade 4 skin reaction	0.0079	95% CI (0.0002 to 0.0441)	Beta	$\alpha = 0.4956$; $\beta = 62.0683$
Cost of surgery for grade 3 skin reaction (£)	635	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 5.0368$
Cost of outpatient attendance for grade 2 skin reaction (£)	113	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.8963$
Cost of initial ENT consultation (paediatric) (£)	126	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.9994$
Cost of follow-up ENT consultation (paediatric) (£)	87	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.6901$
Cost of initial ENT consultation (adult) (£)	106	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.8408$
Cost of follow-up ENT consultation (adult) (£)	69	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.5473$
Cost of initial audiological assessment (£)	55	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.4363$

Input variable	Mean	Variability	Distribution	Sampling method/parameters of distribution
Cost of attendance to fit BAHA (£)	62	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.4918$
Cost of follow-up audiological assessments (£)	48	± 25% of mean	Gamma	$\alpha = 126.0710$; $\beta = 0.3807$

N/A, not applicable.

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Feedback

The HTA programme and the authors would like to know your views about this report.

The Correspondence Page on the HTA website (www.hta.ac.uk) is a convenient way to publish your comments. If you prefer, you can send your comments to the address below, telling us whether you would like us to transfer them to the website.

We look forward to hearing from you.

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