The Cost-Effectiveness of Bimodal Stimulation Compared to Unilateral and Bilateral Cochlear Implant Use in Adults With Bilateral Severe to Profound Deafness

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Abstract

Objectives: An increasing number of severe-profoundly deaf adult unilateral cochlear implant (CI) users receive bimodal stimulation; that is, they use a conventional acoustic hearing aid (HA) in their non-implanted ear. The combination of electric and contralateral acoustic hearing provides additional benefits to hearing and also to general health-related quality of life compared to unilateral CI use. Bilateral CI is a treatment alternative to both unilateral CI and bimodal stimulation in some healthcare systems. The objective of this study was to conduct an economic evaluation of bimodal stimulation compared to other management options for adults with bilateral severe to profound deafness.

Design: The economic evaluation took the form of a cost-utility analysis and compared bimodal stimulation (CI+HA) to two treatment alternatives: unilateral and bilateral CI. The analysis used a public healthcare system perspective based on data from the United Kingdom (UK) and the United States (US). Costs and health benefits were identified for both alternatives and estimated across a patient's lifetime using Markov state transition models. Utilities were based on Health Utilities Index (HUI3) estimates and health outcomes were expressed in Quality Adjusted Life Years (QALYs). The results were presented using the Incremental Cost-Effectiveness Ratio (ICER) and the Net Monetary Benefit approach to determine the cost-effectiveness of bimodal stimulation. Probabilistic sensitivity analyses explored the degree of overall uncertainty using Monte Carlo simulation. Deterministic sensitivity analyses and Analysis of Covariance identified parameters to which the model was most sensitive; i.e. whose values had a strong influence on the intervention that was determined to be most cost-effective. A Value Of Information analysis was performed to determine the potential value to be gained from additional research on bimodal stimulation.

Results: The base case model showed that bimodal stimulation was the most cost-effective treatment option with a decision certainty of 72% and 67% in the UK and US, respectively.

Despite producing more QALYs than either unilateral CI or bimodal stimulation, bilateral CI was found not to be cost-effective because it was associated with excessive costs. Compared to unilateral CI, the increased costs of bimodal stimulation were outweighed by the gain in quality of life. Bimodal stimulation was found to cost an extra £174 per person in the UK ($937 in the US) and yielded an additional 0.114 QALYs compared to unilateral CI, resulting in an ICER of £1,521 per QALY gained in the UK ($8,192/QALY in the US). The most influential variable was the utility gained from the simultaneous use of both devices (CI+HA) compared to Unilateral CI. The value of further research was £4,383,922 at £20,000/QALY ($86,955,460 at $50,000/QALY in the US).

Conclusions: This study provides evidence of the most cost-effective treatment alternative for adults with bilateral severe to profound deafness from publicly-funded healthcare perspectives of the UK and US. Bimodal stimulation was found to be more cost-effective than unilateral and bilateral CI across a wide range of willingness-to-pay thresholds. If there is scope for future research, conducting interventional designs to obtain utilities for bimodal stimulation compared to unilateral CI would reduce decision uncertainty considerably.

INTRODUCTION

Cochlear Implants (CI) were formerly considered to be suitable for severe to profoundly deaf patients who could not benefit from acoustic amplification via conventional hearing aids (Tyler et al. 2002; Ching et al. 2004). In recent years, technology development and enhanced patient outcomes have prompted relaxations in the audiometric criteria for cochlear implantation (Neuman & Svirsky 2013; Siburt & Holmes 2015) and there is a growing population of unilateral CI users with severe to profound deafness that have residual hearing in the non-implanted ear (Potts et al. 2009; Dorman & Gifford 2010). Current evidence suggests that these patients can now benefit from acoustic amplification in that ear and should therefore be managed by combining the electric stimulation from their implant with acoustic stimulation from a hearing aid (HA) fitted to the contralateral ear (Ching 2006; Scherf & Arnold 2014). The concurrent use of both hearing devices (CI+HA) is variably referred to as 'bimodal stimulation', 'bimodal aiding', or 'electric and contralateral acoustic stimulation' (Crew et al. 2015) and can offer important functional advantages compared to CI use alone in at least some patients (Siburt & Holmes 2015).

There is a large amount of variability in estimates of the prevalence of bimodal stimulation among adult CI recipients. Most studies report bimodal rates of only 10-32% (Syms et al. 2002; Tyler et al. 2002; Fitzpatrick et al. 2009; Yamaguchi & Goffi-Gomez 2013; Scherf & Arnold, 2014; Devocht et al. 2015). However, there is a growing population of CI recipients with usable residual hearing in the contralateral ear due to changes in candidacy criteria over time (Ching 2005), and more patients may now benefit from using a hearing aid in their non-implanted ear than ever before (Fielden & Kitterick 2016; Neuman et al. 2017). A recent UK study suggested that the proportion of patients aided bimodally since 2009 may be as high as 48% (Fielden et al. 2016a).

Studies have suggested that bimodal stimulation significantly improves outcomes compared to unilateral cochlear implant use alone in the domains of speech recognition and sound localization (Potts et al. 2009; Crew et al. 2015), perception of music (Crew et al. 2015), sound quality (Morera et al. 2012), quality of life (Farinetti et al. 2015), auditory stimulation (Zhang et al. 2010; Farinetti et al. 2015) and functioning in real life environments (Ching et al. 2004). Improvements in patients' hearing ability as a result of bimodal stimulation have been noted in both quiet and noisy conditions (Harris & Hay-McCutcheon 2010; Farinetti et al. 2015).

Despite this evidence for the potential clinical effectiveness of bimodal stimulation, not every patient will receive all of these benefits from the use of a contralateral HA. Evidence suggests that the perceived health benefits of bimodal stimulation may vary due to sub-optimally fitted hearing aids (Harris & MccCutcheon 2010). Yehudai et al. (2013) also reported that HAs were found to be malfunctioning in a high proportion of bimodal recipients (81%) and Ching et al. (2004) observed some level of degradation in speech perception with contralateral HA use. Malfunction of the HA or the interference that it can cause between the two devices is reported to be the main reason why many bimodal recipients discontinue use of acoustic amplification in their non-implanted ear post-implantation (Fitzpatrick & Leblanc 2010; Scherf & Arnold 2014; Fielden & Kitterick 2016).

The decision faced by policy makers is whether managing adults with severe to profound binaural hearing loss with bimodal stimulation is the most cost-effective option compared to unilateral or bilateral CI; that is, do the additional health benefits that bimodal stimulation generates justify the additional costs involved in its provision compared to the other available alternatives. While the cost-effectiveness of unilateral cochlear implantation compared with non-surgical management (i.e. HAs) in adults has been reported in many studies (United Kingdom CI Study Group 2004; Bond et al. 2009; Turchetti et al. 2011), such studies have not assessed bimodal stimulation as a distinct treatment alternative. For example, Bond et al.

(2009) conducted an economic evaluation that compared unilateral cochlear implantation (assuming 70% of individuals used an acoustic hearing aid in their contralateral ear) to conventional best practice (some patients used HAs and some did not). Although they did account for the cost of the contralateral HA (£100 on average) and its replacement over time (every 5 years), they were unable to identify reliable published estimates of the health benefits (‘utility gain’) from bimodal stimulation and therefore only included the incurred HA costs in their model.

Goman (2014) assessed the minimum utility gain required for bimodal stimulation to be cost-effective compared to a unilateral CI in adults. The study considered the additional costs associated with bimodal stimulation including hearing aid appointments (assessment, fitting etc.) and rehabilitation (aftercare, repairs etc.). The study explored four different scenarios varying the assumptions around the frequency of the hearing aid replacement and the percentage of patients receiving rehabilitation. The author estimated a minimum required utility gain of between 0.0022 and 0.0109 (depending on included costs) for bimodal stimulation to be cost-effective at a willingness-to-pay (WTP) threshold of £20,000 per Quality-Adjusted Life Year (QALY). These estimates represent the best-available evidence for whether bimodal stimulation may be a cost-effective alternative to unilateral and bilateral CI.

There is an outstanding need to assess the cost-effectiveness of bimodal stimulation compared to alternative management options. The key methodological issues to overcome are: (a) the collection of information on the size of the health benefits from a more representative sample of bimodally-aided, unilateral and bilateral cochlear implant users; and (b) obtaining such data from a larger population than studied previously. Variability across clinical services in terms of the fitting and management of hearings aids create the necessity to adjust the economic evaluation to account for considerable differences across services in terms of both the costs incurred and the benefits gained (Fielden & Kitterick 2016). The objectives of this study were therefore to: (i) conduct a cost-utility analysis of bimodal stimulation compared to unilateral and bilateral cochlear implantation; (ii) explore how small changes in the health benefits associated with bimodal stimulation could impact the conclusions of the economic assessment; and (iii) assess how sensitive the results are to assumptions around how long severe to profoundly deaf adults will continue to use a contralateral HA after they receive their CI.

MATERIALS AND METHODS

Ethical Approval

This research was granted ethical approval by the Office for Research Ethics Committees Northern Ireland (ORECNI, REC reference 15/NI/0054). The research followed the principles of the Declaration of Helsinki.

Population

The population under consideration were male and female adults with bilateral severe to profound deafness which, in the UK, are defined as having pure-tone average thresholds >90 dB HL at 2 and 4 kHz. The study considered the mean age for implantation to be 50 years, which is consistent with other economic evaluations and assessments of CI provision in the UK (United Kingdom CI Study Group 2004; Bond et al. 2009; Goman 2014). Our analysis extrapolated costs and health benefits over a lifetime horizon to reflect the intended duration of CI use.

Perspective

The economic evaluation was assessed from a Public Healthcare Service (PHS) perspective to determine the treatment alternative that maximises health benefits within a limited budget. The treatment alternatives considered were unilateral cochlear implantation (CI), bimodal stimulation (CI+HA) and simultaneous bilateral cochlear implantation (CI+CI). In our primary analysis we used an NHS and PSS perspective (National Health Service and Personal Social Services), which is United Kingdom (UK) centred. We also conducted a secondary analysis using information from the United States (US) that accounted for differences in treatment and hearing device costs between the two countries.

Model Description

A decision analytic model was constructed and was based on a decision tree (Fig. 1) and Markov models using the cohort simulation approach, which follows a cohort of patients as a whole through each of the possible model states over time. The structure of the model was based on the clinical pathway that patients commonly follow after unilateral cochlear implantation in the UK (Bond et al. 2009; NICE 2013).

All patients were first assessed for cochlear implant candidacy and a proportion were assumed to be ineligible for implantation. Although the modelling of those patients who are not eligible for implantation is identical across all the treatment alternatives considered in the current study, their inclusion in the model is necessary as the proportion of patients in a population who can access a treatment can affect how cost effective it is.

All patients eligible for CI underwent surgery to implant the electrode array ('internal component'). These patients then entered a three-state Markov model (Fig. 2) including one which denotes the use of CI(s) and two absorbent states (i.e. states from which they could not return to being CI users) representing the non-use of CI(s) and death. Adults in the state denoting use of CI(s) subsequently entered a second Markov model comprised of four states that described the success of implantation surgery and function of the device.

The structure of the model was identical for all three treatment options but the model for bilateral CI assumed patients were implanted simultaneously in both ears and thus included the cost of two cochlear implants but one surgery. The model for bimodal stimulation accounted for extra costs and benefits from the additional use of the contralateral HA over unilateral CI but also assumed that not all patients will be willing or able to use a contralateral HA. The proportion of bimodal users was taken from a large-scale cross-sectional UK survey of 359 unilateral cochlear implant recipients that reported a percentage of contralateral HA use of 45.4% (Fielden et al. 2016a, 2017).

The 'working' state assumed that the fitted CI(s) was functioning and there were no adverse effects. It was assumed that all patients experiencing a failure of the sound processor (external failure) needed a replacement processor. In the case of an internal failure or a major complication, patients were assumed to require an operation for re-implantation, while a proportion of those patients would have the implantable component extracted. For bilateral CI, it was assumed that those who required an extraction continued as unilateral CI users with associated benefits gained and costs incurred. The non-use state reflected the results of CI extraction in the case of unilateral CI users and also voluntary permanent non-use.

The patients who were ineligible for CI entered a two-state Markov model with states 'alive' and 'death'. The alive state represented the non-surgical management of severe to profound deafness in which a proportion of adults were assumed to benefit from (and therefore use) HAs. Of those, a proportion were assumed to use two HAs and the remainder a unilateral HA (Bond et al. 2009). The death state was an absorbent state that represented death due to natural causes (ONS, 2017).

A discount rate of 3.5% was applied for both costs and health outcomes, based on the HM Treasury UK (HMS Treasury 2009; NICE 2013). A willingness-to-pay threshold of £20,000/QALY and $50,000/QALY was used for the UK and US analysis, respectively (NICE 2013; Claxton et al. 2015). A cycle length of 6 months was chosen to illustrate the complexity of events the first two years after cochlear implantation. The analysis was conducted in Microsoft Excel 2016.

Parameter Values

Transition Probabilities

The probabilities used in the model are listed in Table 1. The probabilities related to cochlear implantation were obtained from the most up to date economic model of unilateral cochlear implantation in adults in the UK (United Kingdom Cochlear Implant Study Group 2004; Bond et al. 2009). The probabilities of external failure and major complications (from year 2 onwards) were assumed to be constant over time, whereas the probability of internal failure was assumed to be time dependent. To estimate the annual probability of internal failure, survival curves were generated using the latest Cumulative Survival Percentage (CSP) data available from a major manufacturer of CIs (Cochlear Ltd. 2016) (see Supplemental Digital Content 1, which demonstrates the survival curves and best model fits).

Utilities

A postal survey of unilateral CI users in the UK was conducted to determine the 'utility weights' for unilateral CI and bimodal stimulation. Utility weights were estimated by assessing the self-reported Health-Related Quality of Life (HRQoL) of adult CI users by administering the Heath Utilities Index Mark 3 (HUI3) instrument (Feeny et al. 2002), a preference-based measure of health that has been found to be sensitive to interventions that restore hearing (Yang et al. 2013). The questionnaire was open to all adult unilateral CI users managed by two large clinical services in the UK who met the following inclusion criteria: (i) they must be at least 18 years of age; (ii) they must have received their CI in the UK; (iii) they must have been implanted unilaterally. Questionnaires were completed on paper, responses were anonymous, and no identifying personal information was requested.

A total of 91 patients were confirmed to be eligible and completed the HUI3 questionnaire: 31 bimodal users and 60 unilateral CI users. Bimodal users were defined as those who not only reported using a contralateral HA but using it at the same time as their CI. The incremental utility gain associated with bimodal stimulation was evaluated by comparing the utility weights of the bimodal group to the unilateral CI group using non-parametric analyses (Mann Whitney U test) after accounting for differences in the time since implantation. Bias-corrected confidence intervals for the mean utility weights for the unilateral and bimodal groups were computed using bootstrapping (Davison & Hinkley, 1997).

Four studies were relevant for estimating the utility of bilateral CI. Summerfield et al. (2006) reported a utility increment of 0.031 compared to unilateral CI based on the results from a randomized controlled trial, which was the same incremental value found in an earlier study that estimated utility values using a scenario-based approach (Summerfield et al., 2002). Kuthubutheen et al (2016) reported an average utility increment of 0.035 when comparing the values associated with health state descriptions of bilateral with unilateral CI by patients and healthcare professionals. Finally, Smulders et al (2016) found a utility difference of between 0.02 and 0.04 depending on the measurement instrument used after randomizing patients to unilateral or bilateral implantation. The utility increment associated with bilateral CI over unilateral CI from Summerfield et al. (2002, 2006) was used (0.031) both because it was observed using the HUI3, the same instrument used in the current study to estimate the increment from bimodal stimulation, because it was found consistently across two studies using contrasting estimation methods, and because it approximated the average value reported across all four studies. The final utility weights are shown in Table 2.

Resource Use

Direct costs of the hearing aid and the cochlear implant were calculated using the most suitable and up-to-date unit costs (Table 3). The costs related to unilateral CI were obtained from published literature (United Kingdom CI Study Group 2004; Bond et al. 2009) and were inflated using the appropriate inflation ratio from Hospital and Community Health Services

(HCHS) index (Curtis & Burns 2015). Warranty information of the cochlear implant is presented as supplemental material (see Supplemental Digital Content 2).

Compared to unilateral CI, bimodal stimulation was associated with additional costs related to the contralateral hearing aid. Although a proportion of the cohort of severe to profoundly deaf adults were assumed to already use HAs before implantation (Fig. 1), the model assumed that additional appointments were provided to each bimodally-aided patient following implantation in order to ensure the two devices (CI+HA) were optimized to work together. It was assumed that there was only one follow-up visit related to the HA given that these patients were not new HA users. It was assumed that the HAs were replaced every 5 years (Bond et al. 2009; Summerfield et al. 2010; Goman 2014). The costs related to the hearing aids were based on the UK NHS reference costs 2015/2016 (Department of Health and Social Care, 2016).

In the US analysis, the costs for the hearing aids were gathered from Wertz et al. (2017) and for the cochlear implantation from Semenov et al. (2013). Costs were inflated to 2017 levels using the Consumer Price Index (CPI) for the Medical Care system. These parameters are presented in the supplemental material (see Supplemental Digital Content 3, which demonstrates the costs used in the US analysis).

Decision making

The final decision of an economic analysis can be presented in the form of Incremental Cost-Effectiveness Ratios (ICERs) or using the Net Monetary Benefit (NMB) approach. The ICER is defined by the difference in costs between two alternatives divided by the difference in their health effect. The cost-effectiveness decision is whether this ratio between the incremental costs and benefits (also referred to as the ‘cost per QALY’) is below the willingness-to-pay threshold.

Under the NMB method, the health benefits produced by the interventions under consideration are expressed in monetary terms using the threshold, and the monetary value of the additional health benefits is compared to the generated additional costs. An intervention is cost-effective if it generates higher net monetary benefits compared to the net monetary benefits produced by the other alternatives; i.e. the difference between the NMBs of an intervention and the next best alternative (the Incremental NMB, INMB) is greater than zero.

Scenario Analysis

In the primary analysis, it was assumed that all bimodal recipients continue wearing the HA over a lifetime horizon. However, various studies have suggested that a substantial proportion of bimodal users may cease using their contralateral HA at some point after implantation (Cowan and Chin-Lenn 2004; Devoncht et al. 2015; Fielden & Kitterick 2016; Fitzpatrick and LeBlanc, 2010; Neuman et al. 2017). The scenario analysis used the weighted average of this proportion across several published studies (see Supplemental Digital Content 4, which demonstrates the proportion of adult bimodal users that discontinue hearing aid use) and assumed that 39% of users would cease HA use after five years.

There is also published evidence supporting the use of age-adjusted utilities as the utility of a normal-hearing person diminishes over time (Bond et al. 2009). Therefore, a secondary analysis was conducted using age-dependent utilities to prevent the overestimation of quality of life for which a scaling factor that reduced utilities as a function of age was extracted from Bond et al. (2009).

Uncertainty

A univariate sensitivity analysis was conducted by varying the value of each parameter over a plausible range while all other parameters were held constant (Claxton 2008). Each parameter was varied between the 2.5% and 97.5% percentile values derived using its confidence interval or standard error (Tables 1–3). The results were visualised using a tornado plot in which the effect of varying each parameter on the main output of the model (the INMB value) were plotted for each parameter.

The overall level of uncertainty in the model was quantified by conducting a Probabilistic Sensitivity Analysis (PSA) (Claxton et al. 2005). Instead of each parameter being represented by a single value, each parameter was represented by a probability distribution that expressed the likely range of values the parameter could take. Parameters that referred to probabilities were represented by beta distributions (Table 1), and both utilities (Table 2) and costs (Table 3) were represented by gamma distributions[1]. Monte Carlo Simulation was used to run the model 3000 times and generate pairs of incremental costs and QALYs for each alternative by random sampling. The results are presented on a Cost-Effectiveness Acceptability Frontier (CEAF), a form of graph that illustrates the uncertainty associated with the optimal treatment (i.e. the treatment with the highest expected NMB) for different values of the willingness-to-pay threshold (Fenwick et al. 2001; Barton et al. 2008). The PSA was conducted with Visual Basic for Applications (VBA) in Microsoft Excel 2016.

In addition to capturing overall uncertainty, the results of the PSA can be analysed to assess the relative effect of each parameter on the total amount of uncertainty. In other words, it is possible to identify the parameters that explain the most uncertainty (i.e. variance) in the model outputs (Campbell et al. 2015). Parameters of interest were identified by analysing the output

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[1] A challenge with utilities is they are constrained in the interval $(-\infty, 1]$ and no distribution fits well. This challenge was overcome by transforming the utilities into disutilities (1-Utility), which are bounded between $[0, +\infty)$ and can be represented by a gamma distribution.

of the 3000 simulation runs of the PSA using Analysis of Covariance (ANCOVA) (Briggs et al. 2006). The ANCOVA analysis was conducted in SPSS Statistics 24.

Value Of Information (VOI)

When policy makers use the results of an economic analysis to inform their decision making, they can choose either to approve/reject a new intervention given the current level of uncertainty around the decision, or alternatively they can choose to wait until further (more precise) evidence is obtained from additional research (Briggs et al. 2006). Value Of Information (VOI) analysis quantifies the value that can be gained from resolving uncertainty; that is, it estimates the value of conducting additional research. The Expected Value of Perfect Information (EVPI) is calculated and represents the upper bound of the expected per-patient benefits of further research. The EVPI can also be estimated at a population level (pop EVPI) by incorporating information about the size of the relevant patient population.

The size of the patient population was estimated using the prevalence of adults with profound deafness who are considered likely to access CI services in the UK and US. Figures from the British Cochlear Implant Group (BCIG) indicate that on average 800 adults have been implanted unilaterally each year between 2011 and 2017. The VOI analysis assumed that the total population of adults who are likely to avail of cochlear implantation in the UK is equal to 800 per year over the next 10 years, which after discounting over that period equated to a population of 6,886 adults. For the US, the number of adults implanted has increased from 41,000 in 2010 (NIH, 2010) to 58,000 in 2012 (NIH, 2016), a growth of 5,667 per year, resulting in a 10-year discounted population estimate of 48,780. The discounting period of 10 years was chosen as a time horizon over which the benefits of an optimal decision could be expected to accrue, but not so long as to mean that some of the treatment options or assumptions in the model may no longer be applicable.

RESULTS

Base case results are summarized in Table 4. Bimodal stimulation generates an ICER of £1,521 per QALY compared to unilateral cochlear implantation. This ICER is below the £20,000 threshold adopted by NICE in the UK, suggesting that bimodal stimulation is more cost-effective than unilateral CI. Bilateral CI is deemed not to be cost-effective as it generates an ICER of £219,900/QALY compared to bimodal stimulation, and is even less cost-effective when compared to unilateral CI. The economic evaluation looking at costs from a US perspective arrived at similar results. Bimodal stimulation generates an ICER of $8,192/QALY over unilateral CI, while bilateral CI is again not cost-effective compared to the other two alternatives.

Under the Net Monetary Benefit framework, the INMB from bimodal stimulation compared to unilateral CI was positive for both the UK (£2114) and US ($4784) perspectives, and is therefore the preferred alternative among the three treatment options considered. For the UK, the individual QALY gain (+0.11 years) from offering bimodal stimulation compared to unilateral CI was reached at an additional cost of £174 per person. The accrual of incremental NMB at a threshold of £20,000 per QALY over a lifetime horizon is shown in Figure 3 (‘base case’). Bimodal stimulation offers increasingly more benefits throughout the years compared to unilateral CI use and starts to generate additional monetary benefits over unilateral CI by the end of the 1st year after cochlear implantation.

The first scenario analysis re-considered the cost-effectiveness of bimodal stimulation assuming that approximately 39% of bimodal users would discontinue the use of the HA voluntarily (Fig. 3, ‘Stop being bimodal’). The incremental NMB was smaller than the base case but still positive (£1511), and still identified bimodal stimulation as the most cost-effective alternative. Having less bimodal recipients reduced the expected costs by 0.11% and the amount of QALYs by an even greater extent (drop of 0.53%) compared to the base case. In other words, the reduced health benefits gained from having fewer patients aided bimodally still outweighed the cost savings arising from having fewer contralateral hearing aid users to support. The same conclusion can be derived from the US analysis.

The second scenario analysis used utilities that diminished over the patient's lifetime to reflect aging-related changes in health. Diminishing utilities generated less NMBs making bimodal stimulation not cost-effective compared to unilateral CI with a negative INMB of -£1392. A similar pattern is illustrated in the US analysis. While bimodal stimulation does generate positive incremental net monetary benefits for the first three decades following implantation, the utility gained from bimodal use is outweighed by the additional costs once the average age of the cohort reaches 79 (Fig. 3, 'Diminishing Utilities'). The cost-effectiveness of bimodal stimulation under diminishing utilities will therefore be dependent on the assumed average age at implantation (50 years in the current study), the number of years lived following CI surgery (23 on average in the current study based on the observed average life expectancy in the cohort of 83 years), and the time horizon adopted for the analysis (a lifetime horizon was adopted following the approach taken by NICE when formulating guidance on cochlear implants).

The results of the univariate sensitivity analyses are presented in the left panels of Figure 4. The key parameters shown in the tornado plot were identified using ANCOVA, which indicated that the utility parameters had the highest impact on the variance of the model outputs, contributing to 84% (98% for US) of the overall uncertainty in the INMB. The uncertainty in the bimodal utility explained 54% (64% for US) of the variance in INMB. A drop in the bimodal utility of more than 0.029 (0.027 for US) would lead to a negative incremental NMB, making unilateral cochlear implantation more cost-effective than bimodal stimulation. On the other hand, a small increase in the bimodal utility resulted in high incremental NMBs; e.g. an increase of only 6% in the bimodal utility doubled the incremental NMB. Bilateral CI is not cost-effective compared to either bimodal stimulation and unilateral CI even when using the highest reported utility value (an increment of 0.04 compared to unilateral CI) (Smulders et al, 2016).

The model outputs were insensitive to the cost of the acoustic hearing aid in both countries. Similarly, the appointment costs related to hearing aids (assessment, fitting & follow-up) did not influence the economic outputs to the extent needed to affect the conclusions of the base case analysis (Fig. 4, left panels). The univariate sensitivity analysis suggested that the cost parameters in the US analysis were slightly more influential than the UK analysis, although the cost-effectiveness of bimodal stimulation remained robust; i.e. no plausible value examined for the costs related to hearing aids resulted in negative INMB values. The INMB remained positive for HA device prices up to $6,000. Another influential parameter was the proportion of patients receiving bimodal stimulation, which explained 13% and 27% of the output variance in the UK and US analysis, respectively. The relationship between bimodal use and NMB was positive such that increasing the proportion of bimodally-aided patients resulted in a higher INMB in both analyses (UK & US). A rise in the number of bimodal users led to greater increases in health benefits than in costs.

The right panels of Figure 4 also demonstrates the differences in QALYs and costs from the PSA on a cost-effectiveness plane (scatter plot). Overall, there was little decision uncertainty surrounding the optimal strategy; the majority of simulation runs were gathered around a tight cluster below the threshold (i.e. produced a positive INMB) offering robustness to the base case results. In the UK analysis, the simulations spread more horizontally (across the incremental QALYs axis), while there was little variance in the incremental costs. This pattern is compatible with the results of the univariate sensitivity analysis that identified the utility weights as the key parameters causing uncertainty in the model. In the US analysis, cost parameters have a bigger role in the overall uncertainty and thus the simulations show a greater degree of spread vertically along the incremental cost axis, reflecting the plausible ranges of costs obtained from the published literature.

Figure 5 plots the Cost-Effectiveness Acceptability Frontier (CEAF) that illustrates both the optimal decision (the treatment alternative that generates the highest net monetary benefits), the probability of the decision being correct (the amount of uncertainty around the decision), and the population-level EVPI (the value of conducting further research) as a function of willingness-to-pay threshold. The CEAF shows that bimodal stimulation has a high probability of being cost-effective ($p \approx 0.7$) across most thresholds compared to the other two alternatives. For willingness-to-pay thresholds of £20,000/QALY and £30,000/QALY the probability of bimodal stimulation being the most cost-effective alternative was 72% and 73%, respectively. In the US analysis, bimodal stimulation was cost-effective with a 67% and 59% certainty at a $50,000/QALY and $100,000/QALY threshold, respectively (Table 4).

Figure 5 also shows that the EVPI in the UK analysis reached an initial peak where uncertainty between unilateral CI and bimodal was the highest; i.e. around the threshold value of £1,900/QALY. The continuous increase in the population EVPI at thresholds above that point indicates that there is more decision uncertainty around higher thresholds, mostly likely because the probability of bilateral CI becoming cost-effective increases. At a £20,000/QALY threshold ($50,000/QALY in the US analysis), the estimated value of reducing uncertainty through further research is £637 ($1783 in the US) per patient, leading to a population EVPI estimate of £4,383,922 ($86,955,460 in the US).

DISCUSSION

This economic evaluation found that bimodal stimulation is more cost-effective than unilateral or bilateral cochlear implantation for adults with bilateral severe to profound sensorineural deafness in both the UK and the US from a public health service perspective. With an ICER of £1,521/QALY over unilateral CI, bimodal stimulation would be considered highly cost-effective as it is well below the lowest limit of the cost-effectiveness threshold range adopted by NICE (£20,000/QALY). Similar results are derived from the US analysis, where bimodal use generated an ICER of $8,192 compared to unilateral CI, well below the $50,000/QALY threshold. Bimodal stimulation generated greater health benefits than unilateral cochlear implantation, and on average those benefits outweighed the extra cost burden related to the maintaining of the contralateral acoustic hearing aids. Although bilateral CI produces more health benefits (QALYs) than the other treatment alternatives, the excessive costs associated with providing two cochlear implants means that it is highly unlikely to be the most cost-effective alternative.

The decision on the cost-effectiveness of bimodal stimulation is not sensitive to the choice of WTP threshold as it is only at thresholds lower than £1,900/QALY that costs of the additional hearing aids outweigh the benefits they provide to adult CI users. The robustness of the UK model output is demonstrated by the high level of decision certainty after accounting for the joint uncertainty in all parameters; i.e. bimodal stimulation was the most cost-effective alternative in 72% of the model runs at a threshold of £20,000/QALY. An examination of the proportion of simulations (28%) for which bimodal stimulation was not the most cost-effective option (in which unilateral CI was more cost-effective) indicated that those results were mainly driven by bimodal stimulation generating less health than unilateral CI. This situation is not implausible as there have been reports of poorer outcomes due to interference between electrical and acoustic inputs and the perceptual differences between the two inputs (Scherf &

Arnold 2014; Yehudai et al. 2013). This possibility was incorporated in the model by adjusting the standard errors of the two utility parameters in such a way as to allow for the possibility that bimodal stimulation could generate less health benefits than unilateral CI use. If improving the fitting of both hearing devices in bimodal stimulation decreased the possibility of a reduction in health benefits compared to unilateral CI, decision uncertainty would be reduced even further, although even without such assumptions decision uncertainty with current information is low.

The only previous study that has considered the cost-effectiveness of bimodal stimulation estimated that a utility gain of at least 0.0109 was required to make bimodal stimulation cost-effective. The current study obtained HUI3 data from 91 adults managed at two large CI services in the UK and estimated a utility gain of 0.032 by comparing a bimodally-aided group with a unilateral CI group while controlling for years of experience with CI use (Table 2, unilateral 0.478 vs bimodal 0.510). The size of this utility gain was sufficient to outweigh the extra costs of maintaining the contralateral HAs if utilities did not diminish over time, and even if it was assumed a proportion of patients stopped using their contralateral HAs. When utilities diminished over time under the assumption of an age-related decline in health, bimodal stimulation only ceased being cost-effective after the patients reached ~80 years of age; i.e. after approximately 30 years of device use. Thus, even under the assumption that utilities diminish with age, bimodally-aided adults will get benefit well into their working life. The basic implication from the age-adjusted utilities analysis is that as patients get to an advanced age their health looks increasingly similar regardless of the hearing devices they use; i.e. unilateral CI or CI+HA.

The utility gain for bimodal stimulation used in the present analysis was obtained from UK patients who had received no rehabilitation specific to bimodal stimulation as it is not routinely provided in the UK (Fielden & Kitterick 2016). This fact underpinned the assumption that the only additional costs for bimodal stimulation associated with that utility gain were the costs required to ensure the hearing aid was up to date and maintained over time, which is also in line with the assumptions made by Bond et al. (2009). Even if we used higher cost estimates as Goman (2014) reported, who assumed a greater level of extra rehabilitation and aftercare in the bimodal group, the final decision on the cost-effectiveness of bimodal stimulation remains the same. The overall costs for bimodal stimulation were also higher in the US vs the UK model in the present evaluation, resulting in an additional cost of $937 per person for bimodal users, but bimodal stimulation still remained cost-effective. Thus, the current results suggest that bimodal stimulation would be affordable under a public healthcare system in the US. The provision of bimodal stimulation in the US not only has the potential to increase patient benefit, but also reduce inequalities within the healthcare system as contralateral acoustic HAs are not provided through the public health system in the US, but instead are purchased privately. Bainbridge and Ramachandran (2014) reported that the prevalence of hearing aid use among older adults was 28% to 66% higher in those with higher incomes compared to the adults on the lower end of the income-to-poverty distribution. It is possible that similar differences could arise in contralateral HA use among adult CI users in the US.

The EVPI depicts the upper price that a healthcare system should be willing to spend for obtaining additional evidence. Performing more trials and conducting further research provides more accuracy around the input parameters and resolves part of the uncertainty around the final outcome of the model. It would appear feasible for randomised controlled trials or other types of formal clinical evaluations to be conducted within these funding limits (£4,383,922 at £20,000/QALY; $86,955,460 at $50,000/QALY). If there is scope for further investment, it should be where the uncertainty is highest. Results from the sensitivity analyses suggest utility weights as the key driver of parameter uncertainty. Future research could address this issue by conducting interventional designs (e.g. randomized controlled trials) to obtain utility weights for bimodal stimulation compared to unilateral CI, rather than rely on data from observational studies on which the current utility weights are based. The lack of health utility data in the field of cochlear implantation in general is an important evidence gap to address given the important role that economic evaluations play in determining whether such a low-volume high-cost intervention is good value for money from the payer's perspective.

Economic evaluations typically use generic health instruments to obtain data on health-related quality of life; that is, instruments that are by design relevant to a wide range of health conditions. Although the EQ-5D (Brooks, 1996; The Euroqol Group, 1990) and Health Utilities Index Mark 3 (HUI3) (Boyle et al. 1995; Feeny et al. 2002) are both standardized instruments used in a wide range of health conditions, both are limited in detecting differences between degrees of hearing loss and changes due to hearing-related interventions. The EQ-5D has been found to perform poorly in hearing-related conditions in terms of its sensitivity to change (Barton et al. 2005; Grutters et al. 2007; Longworth et al. 2014). The HUI3 has been found to be more sensitive although largely to the comparison between 'no hearing' to 'some hearing' rather than to different degrees of hearing (or to bilateral versus unilateral hearing) (Lovett et al. 2009; Goman 2014). Although the EQ-5D is the preferred instrument of NICE (NICE, 2013), it has accepted evidence of the effectiveness of cochlear implantation based on HUI3 data when forming its guidance on who should be able to access cochlear implantation in the UK (NICE, 2009). This fact and the availability of HUI3 data on the utility gain from bilateral implantation led to the use of the HUI3 in the current study. However, it is possible that this choice of instrument was suboptimal given that it's dimensions do not explicitly cover aspects of hearing that are contingent on binaural hearing (or at least bilateral access to sound). The lack of well-validated preference-based measures of health-related quality of life that are sensitive to hearing and hearing-related interventions but also whose use is suitable for informing economic evaluations poses an ongoing challenge for the application of health economics to hearing healthcare.

The scenario analyses indicated that the incremental net monetary benefits from bimodal stimulation compared to unilateral CI were reduced substantially if it was assumed that a proportion of bimodally-aided patients ceased use of their HA after a period of time. Scherf and Arnold (2014) reported that the main reason for rejecting the hearing aid was the absence of any perceived benefits. Poor provision and management of bimodal stimulation could potentially lead to bad synchronisation and malfunctioning of the two devices, reduce benefit, and ultimately lead to non-use. Differences have been observed across clinical practices in the procedures used to fit and tune the HA after implantation (Scherf & Arnold 2014) and a majority of practices in the UK do not have an agreed protocol on the best approach of fitting both devices simultaneously (Fielden & Kitterick 2016). Surveys of cochlear implant audiologists have suggested that clinicians need guidelines around issues of bimodal candidacy and management (Scherf & Arnold 2014; Siburt & Holmes 2015; Fielden & Kitterick, 2016). Such guidelines could help reduce non-use of the HA by optimizing bimodal fitting procedures to maximise patient benefit or by ensuring that only patients likely to benefit are aided bimodally.

The current economic model suggests that a higher number of bimodal users as a proportion of all CI recipients would lead to even greater net benefits. An increase in bimodal usage in the UK of approximately 34% that occurred around 2009 has been attributed to the change in guidance on candidacy criteria in the UK that permitted candidates to have greater access to residual hearing (Fielden et al. 2016a). Bimodal usage could increase further if the candidacy criteria for cochlear implantation are expanded to those with even greater levels of residual hearing. Such an eventuality would seem possible as the candidacy criteria adopted in UK are some of the most restrictive in the world (Vickers et al. 2016). In the US, providing HAs through the public healthcare system might increase the rate of contralateral HA use among CI users without bimodal stimulation necessarily being offered as a distinct treatment option. Although any increase in bimodal usage would render bimodal stimulation even more cost-effective than it currently appears to be, it would necessitate audiologists having to manage an even greater number of bimodally-aided patients in the future. This growing trend places additional emphasis on the need to develop guidance around maintaining both devices simultaneously, to develop enhanced fitting procedures, and to identify which patients have the capacity to derive benefit from a contralateral HA.

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FIGURE CAPTIONS

Figure 1. The decision tree and Markov models used for evaluating costs and health-related outcomes of combining electric and contralateral acoustic stimulation (CI+HA, ‘Bimodal stimulation’) compared to electric stimulation alone (unilateral and bilateral CI).

Figure 2. Schematic representation of the Markov Model for use of one or two CIs. Ellipses indicate distinct health states and arrows show the permitted directions in which the simulated cohort could move from one state to another. The states reflect the possible post-operative outcomes following unilateral or bilateral cochlear implantation and two absorbent states of non-use and death due to natural causes.

Figure 3. Base-case and scenario analysis results of the comparison between bimodal stimulation and unilateral CI at a willingness-to-pay threshold of £20,000/QALY. Results from the US analysis are not displayed because they are similar to the UK analysis.

Figure 4. Tornado plots (left) of the one-way sensitivity analyses of bimodal stimulation versus unilateral CI for the UK (top) and US (bottom) summarizing the uncertainty attached to each individual parameter. Key parameters were identified using analysis of covariance. Each parameter was varied between the 2.5% and 97.5% percentile. Scatter plots (right) of incremental costs and incremental quality-adjusted life years of bimodal stimulation versus unilateral CI. The majority of simulation runs (UK 72%; US 67%) were observed to lie below the willingness-to-pay threshold.

Figure 5. Cost-Effectiveness Acceptability Frontier (CEAF) representing the results from the probabilistic sensitivity analysis. The graph illustrates the management option with the highest expected net monetary benefits (NMB), the probability that this intervention is cost-effective, and the population Expected Value of Perfect Information (pop EVPI) across a range of willingness-to-pay thresholds. All three treatment alternatives were considered (unilateral CI,

bimodal stimulation and bilateral CI) although no part of the graph relates to bilateral CI as it was not the optimal choice at any of the WTP threshold values considered. Results from the US analysis are not displayed because they are similar to the UK analyses.

Figure 1

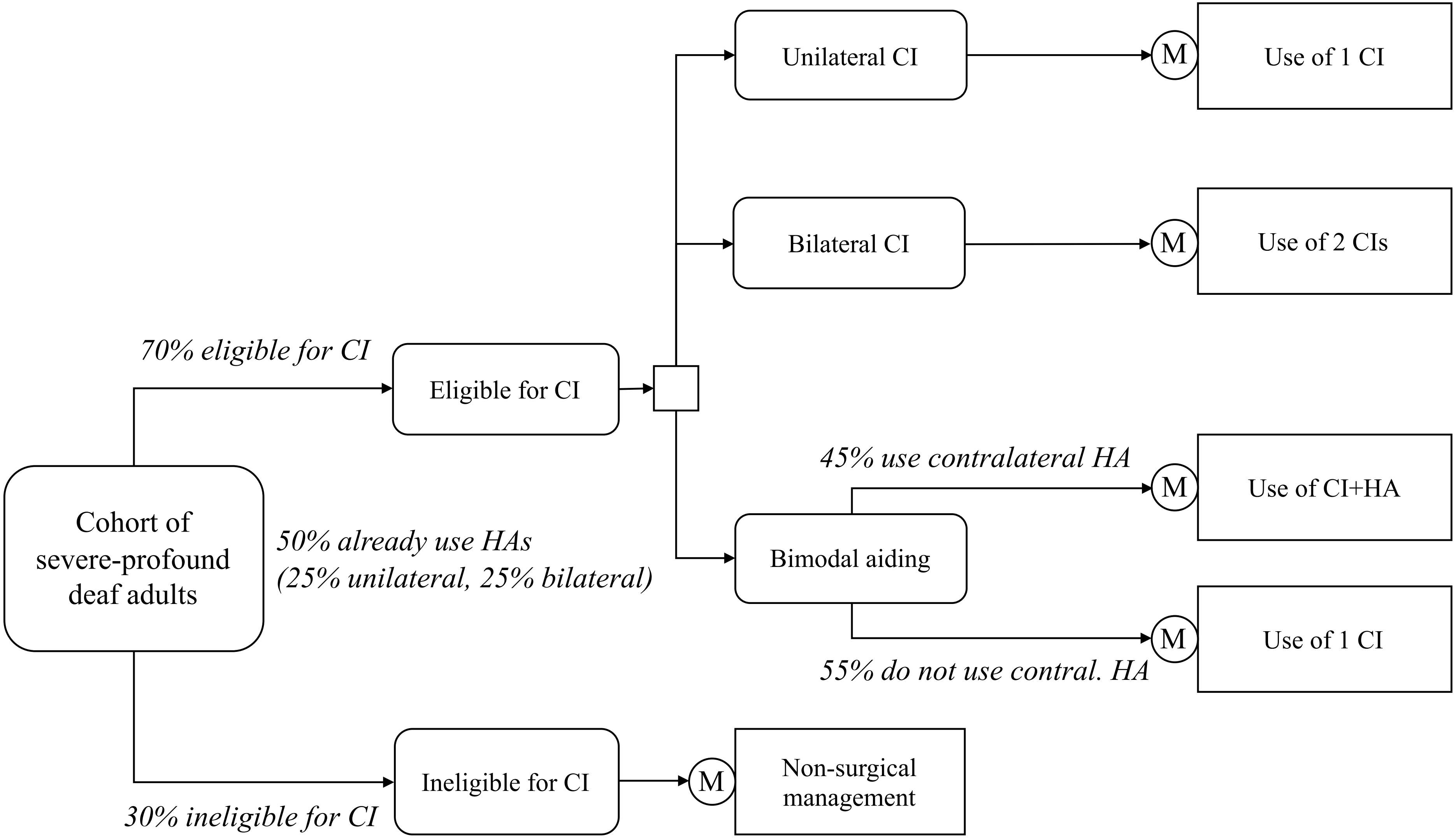
Unilateral CI
M
Use of 1 CI
Bilateral CI
M
Use of 2 CIs
70% eligible for CI
Eligible for CI
45% use contralateral HA
M
Use of CI+HA
Cohort of severe-profound deaf adults
50% already use HAs
(25% unilateral, 25% bilateral)
Bimodal aiding
M
Use of 1 CI
55% do not use contral. HA
Ineligible for CI
M
Non-surgical management
30% ineligible for CI

Figure 2

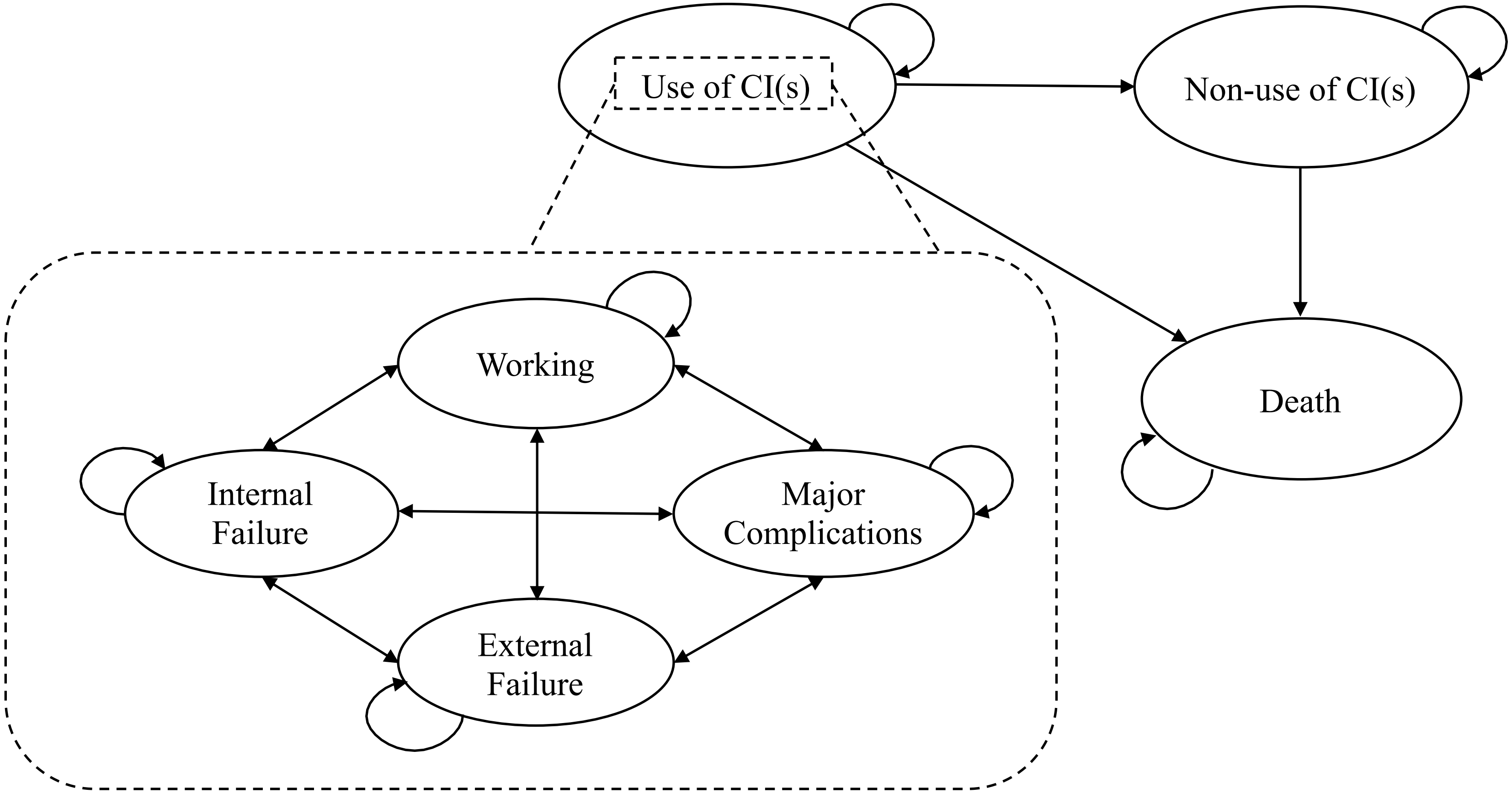
Use of CI(s)
Non-use of CI(s)
Working
Death
Internal Failure
Major Complications
External Failure

Figure 3

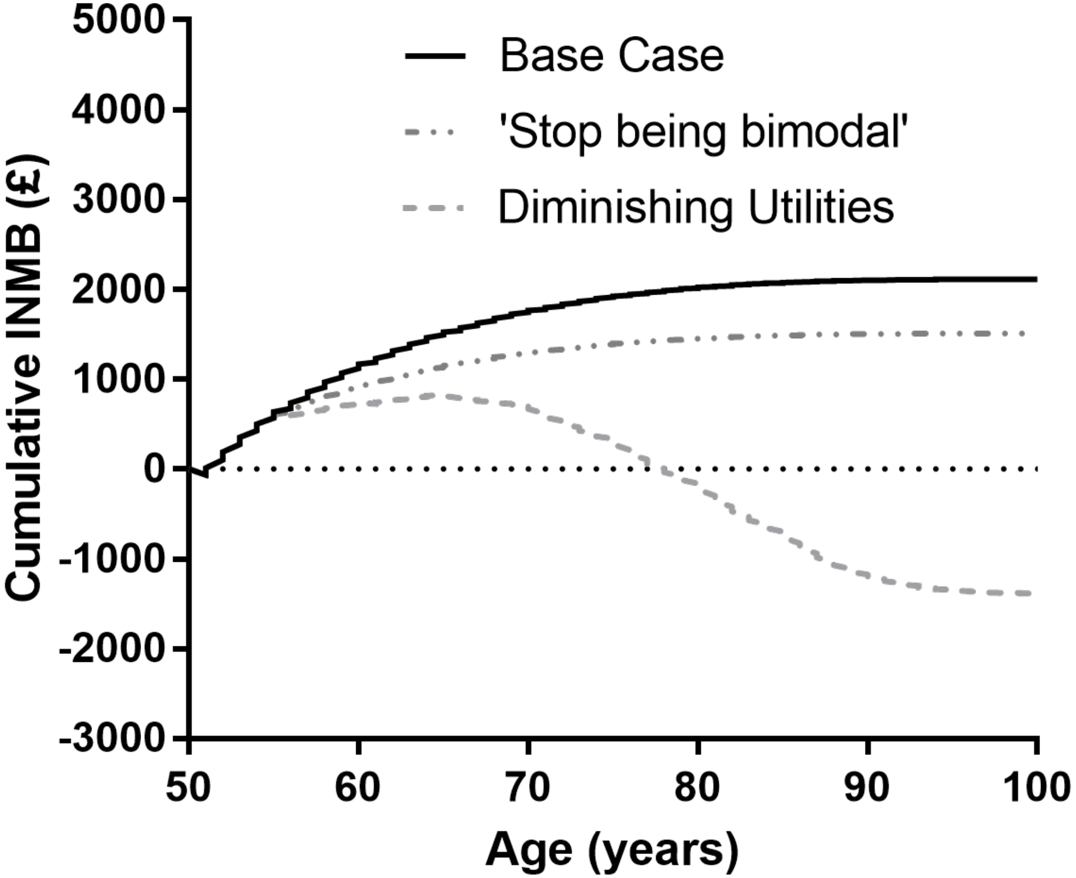
Base Case
'Stop being bimodal'
Diminishing Utilities
Cumulative INMB (£)
5000
4000
3000
2000
1000
0
-1000
-2000
-3000
50
60
70
80
90
100
Age (years)

Figure 4

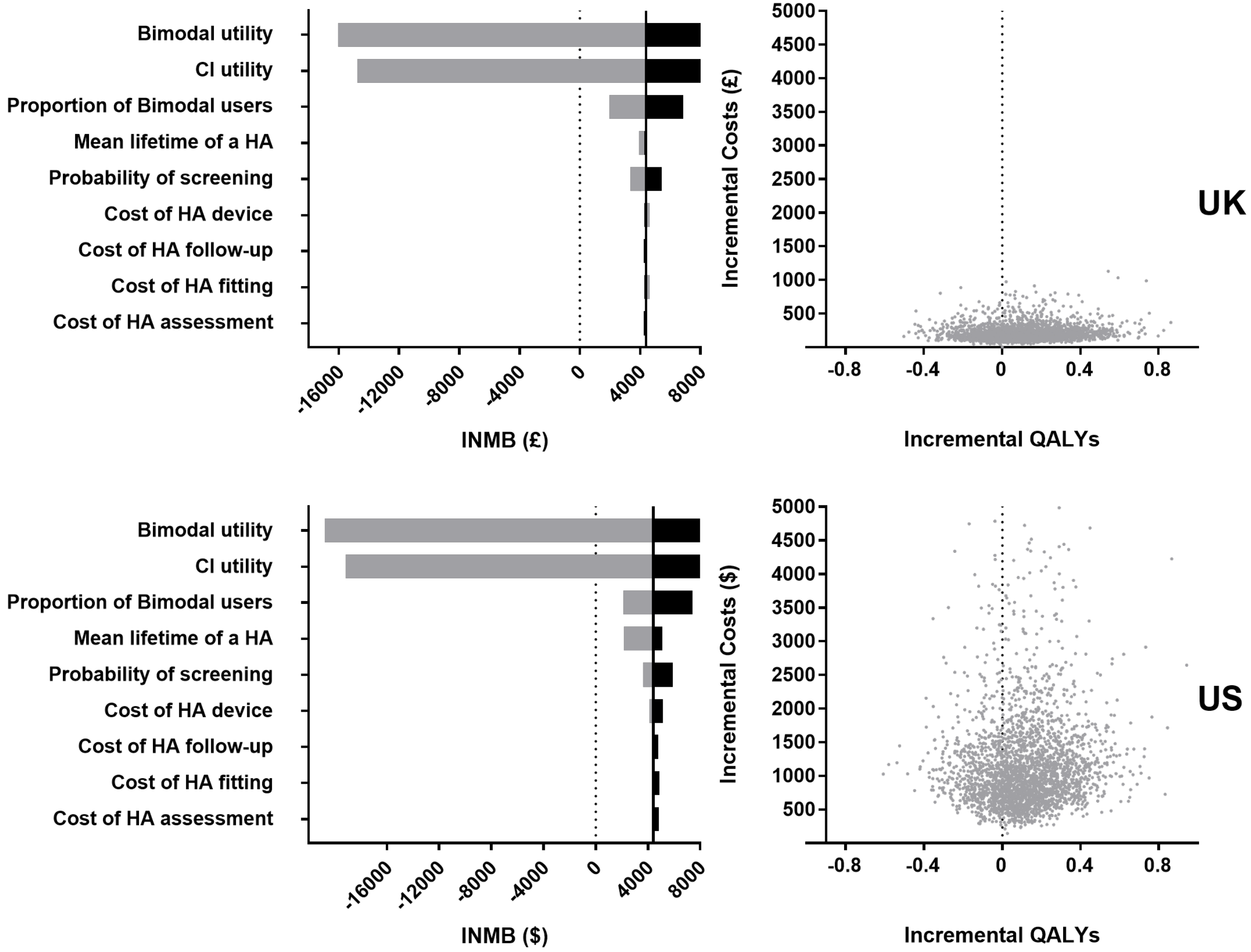
Bimodal utility
CI utility
Proportion of Bimodal users
Mean lifetime of a HA
Probability of screening
Cost of HA device
Cost of HA follow-up
Cost of HA fitting
Cost of HA assessment
-16000
-12000
-8000
-4000
0
4000
8000
INMB (£)
Incremental Costs (£)
5000
4500
4000
3500
3000
2500
2000
1500
1000
500
-0.8
-0.4
0
0.4
0.8
Incremental QALYs
UK
Bimodal utility
CI utility
Proportion of Bimodal users
Mean lifetime of a HA
Probability of screening
Cost of HA device
Cost of HA follow-up
Cost of HA fitting
Cost of HA assessment
-16000
-12000
-8000
-4000
0
4000
8000
INMB ($)
Incremental Costs ($)
5000
4500
4000
3500
3000
2500
2000
1500
1000
500
-0.8
-0.4
0
0.4
0.8
Incremental QALYs
US

Figure 5

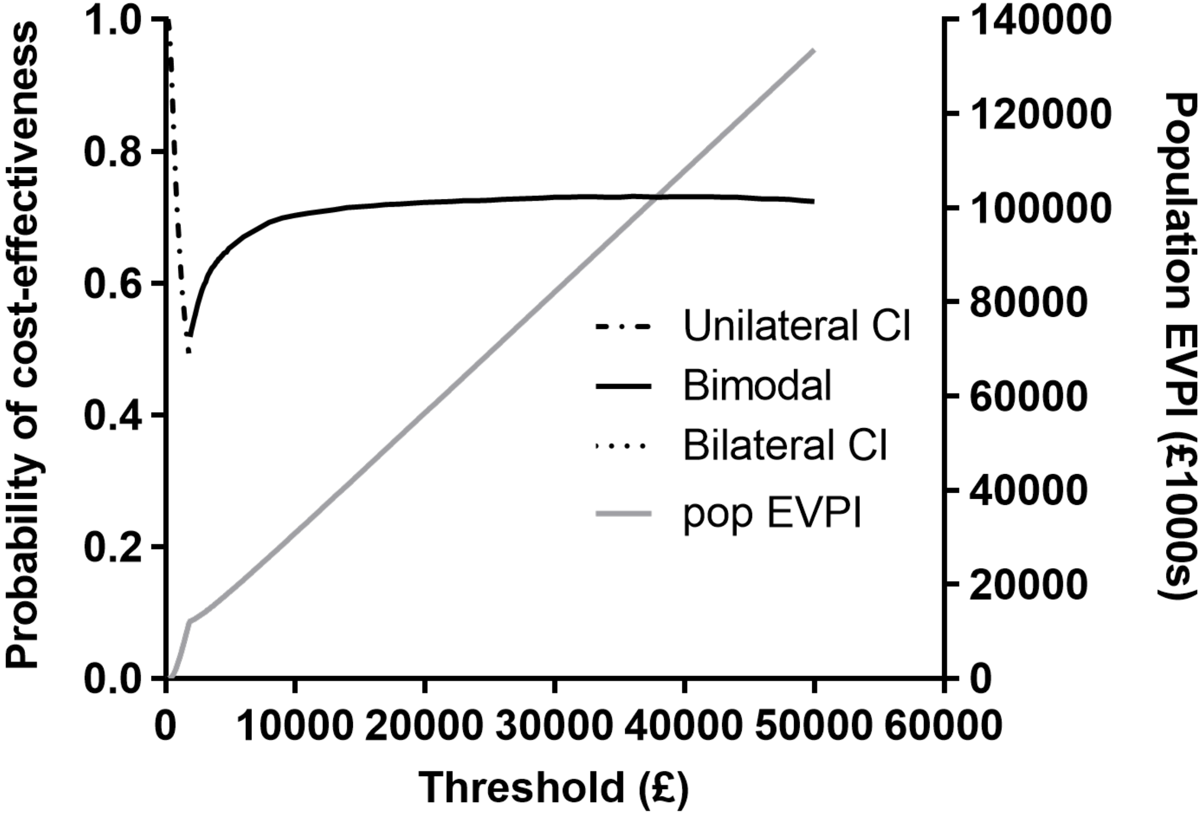
1.0
0.8
0.6
0.4
0.2
0.0
Probability of cost-effectiveness
140000
120000
100000
80000
60000
40000
20000
0
Population EVPI (£1000s)
0
10000
20000
30000
40000
50000
60000
Threshold (£)
Unilateral CI
Bimodal
Bilateral CI
pop EVPI

Table 1. Probability parameters of the Markov state transition models. The means and standard errors (SEs) were used to define beta distributions for the probabilistic sensitivity analyses.

Parameter Name	Mean	SE	Source	Description
Screening	0.3	0.075	Bond 2009	Proportion of initial referrals not undergoing an operation to fit a CI (i.e. who receive non-surgical management)
Bimodal users	0.454	0.1135	Fielden 2016, 2017	Proportion of CI recipients who use a contralateral HA
Surgical Death	0	N/A	Bond 2009	Probability of death from cochlear implant surgery
External failure*	0.062	0.0155	Bond 2009	6-month probability of external component (sound processor) failure
Internal failure*	Time-dependent	N/A	Cochlear Europe submission, Conboy and Gibbin 2004	6-month probability of internal component (receiver-stimulator & electrode array) failure
Major complications yr 1*	0.02	0.005	Bond 2009	6-month probability of major complication† in year 1
Major complications yr 2+*	0.002	0.0005	Bond 2009	6-month probability of major complication† in year 2+
CI extraction	0.115	0.0049	Bond 2009	Probability of non-reimplantation of a cochlear implant due to internal failure or major complications
Elective non-use of CI	0.0236	0.0059	Bond 2009	Probability of voluntary non-use of implants (applied once at the end of 2nd year of implant use)
Use of 1 HA	0.25	0.125	Bond 2009	Proportion of candidates for CI who use 1 HA
Use of 2 HAs	0.25	0.125	Bond 2009	Proportion of candidates for CI who use 2 HAs

*Probability is doubled for bilateral cochlear implantation. †Event requiring any form of operation not related to a device failure.

Table 2. Utility parameters of the Markov state transition models. All utilities were based on data obtained using the HUI3 instrument. Lower and Upper 95% Confidence Intervals (LCI, UCI) were calculated by bootstrapping, which was also used as the sampling method for these parameters in the probabilistic sensitivity analyses.

Parameter Name	Mean	LCI	UCI	Source	Description
Absolute utilities					
Non-surgical management	0.433	0.411	0.455	Bond 2009	Utility of profoundly deaf adults with access to HAs
Unilateral CI	0.478	0.420	0.536	Current study	Utility of unilateral cochlear implant use
Bimodal	0.510	0.428	0.592	Current study	Utility of bimodal use
Incremental utilities					
Bilateral CI	0.031	0.018	0.042	Summerfield 2010	Incremental utility gain from using bilateral cochlear implants over a unilateral implant

Table 3. Cost parameters of the Markov state transition models measured in Pound Sterling (£). The means and Lower and Upper 95% Confidence Intervals (LCI, UCI) or standard errors (SEs) were used to define gamma distributions for the probabilistic sensitivity analyses.

Parameter Name	Mean	LCI-UCI (SE)	Source	Description
Costs of HA				
Hearing Aid	86	65-108	NHS Reference Costs 2015-2016	Cost of a hearing aid
Assessment	53	40-66	NHS Reference Costs 2015-2016	Cost of a hearing aid assessment
Fitting	71	53-89	NHS Reference Costs 2015-2016	Cost of fitting a hearing aid
Follow-up	53	40-67	NHS Reference Costs 2015-2016	Cost of follow-up after hearing aid fitting
Costs of a CI				
Candidacy	4945	3907-5587	Bond 2009*	Presurgical candidacy costs
CI surgery (Unilateral)	3469	1144-7528	Bond 2009*	Unilateral implantation costs (excluding system costs)
CI surgery (Bilateral)	5204	(1041†)	Bond 2009*	Bilateral implantation costs (excluding system costs)
CI device (Unilateral)	14900	(3603†)	NICE 2009	Mean cost of unilateral cochlear implant system
CI device (Bilateral)	23840	(4768†)	NICE 2009	Mean cost of bilateral cochlear implant system, assuming a 40% discount for the second implant (NICE, 2009)
Maintenance yr1 (Unilateral)	6164	5425-6534	Bond 2009*	Tuning and maintenance costs in year 1
Maintenance yr2	984	757-1436	Bond 2009*	Maintenance costs in year 2
Maintenance yr3	932	392-1435	Bond 2009*	Maintenance costs in year 3
Maintenance yr4+	735	391-1079	Bond 2009*	Maintenance costs in years 4+
Upgrade (Unilateral)	5072	(101†)	Bond 2009*	Processor upgrade
Major Complications (Unilateral)	9588	9004-10519	Bond 2009*	Medical event requiring surgery not related to device failure

*Inflated to 2015/16 prices. †Variance of costs set to 1/5 of the mean value following Summerfield et al. (2010).

Table 4. Results of the cost-effectiveness analyses for the base case.

Treatment Alternative	Expected QALYS	Expected Costs	ICER*	NMB	INMB*	Probability of being cost-effective†	
						£20,000/QALY	**£30,000/QALY**
United Kingdom (WTP threshold £20,000/QALY)							
Unilateral CI	5.87	£33,227	-	£84,050	-	28%	27%
Bimodal	5.98	£33,401	£1,521/QALY	£86,165	£2,114	72%	73%
Bilateral CI	6.11	£62,688	£219,900/QALY	£59,542	-£26,623	0%	0%
United States (WTP threshold $50,000/QALY)						**$50,000/QALY**	**$100,000/QALY**
Unilateral CI	5.87	$46,229	-	$246,964	-	30%	24%
Bimodal	5.98	$47,166	$8,192/QALY	$251,748	$4,784	67%	59%
Bilateral CI	6.11	$79,120	$239,926/QALY	$226,453	-$25,295	3%	17%

***Incremental values calculated by comparing adjacent rows; i.e. Bimodal aiding to Unilateral CI and Bilateral CI to Bimodal aiding. †Probabilities represent the proportion of simulations for which each treatment alternative generated the greatest net monetary benefits.**

APPENDICES

Survival Curves to inform the probability of failure of the internal component of a single cochlear implant.

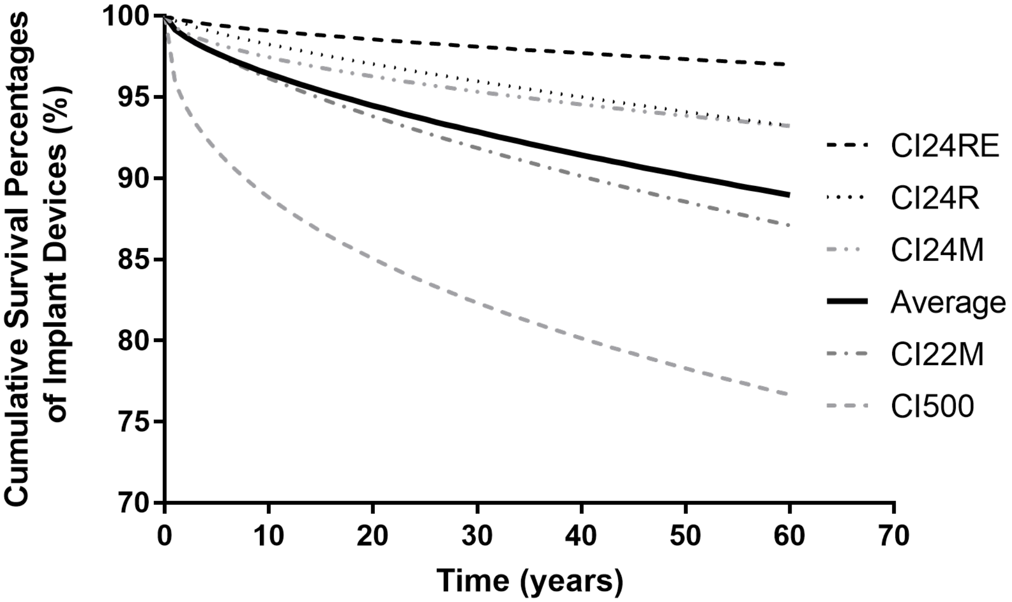

Table A. Warranty and lifetime parameters for the hearing devices considered in the Markov models.

Parameter	Mean	SE	Source	Description
Proportions in warranty				
Proportion of internal failures	0.007		Bond 2009	Proportion of internal component failures occurring during warranty period
Proportion of external failures	0.318		Bond 2009	Proportion of external component failures occurring during warranty period
Warranty costs (£)				
Internal Failure (during warranty)	3469	694†	Bond 2009*	Cost of internal component failure (during warranty period)
Internal Failure (after warranty)	21483	4297†	Bond 2009*	Cost of internal component failure (in years after warranty period)
External Failure (during warranty)	0	19†	Bond 2009*	Cost of external component failure (during warranty period)
External Failure (after warranty)	5072	1014†	Bond 2009*	Cost of external component failure (in years after warranty period)
Lifetime in years				
Lifetime of an HA	5		Bond 2009 Summerfield 2010 Goman 2014	The number of years after which an acoustic hearing aid is likely to be upgraded in routine clinical practice.
Warranty of CI (internal)	10		Bond 2009	The warranty period of the internal part of a CI
Warranty of CI (external)	3		Bond 2009	The warranty period of the external part of a CI

*Inflated to 2015/16 prices. †Variance of costs set to 1/5 of the mean value following Summerfield et al. (2010).

Table B. Cost parameters of the Markov state transition models. Measures in US Dollars ($)*.

Parameter Name	Mean	SE†	Source	Description
Costs of HA				
Hearing Aid	700	140	Wertz et al. 2017	Cost of a hearing aid
Assessment	234	47	Wertz et al. 2017	Cost of assessment of a hearing aid
Fitting	197	39	Wertz et al. 2017	Cost of fitting of a hearing aid
Follow-up	197	39	Wertz et al. 2017	Cost of follow-up of a hearing aid
Costs of CI				
Candidacy	1650	330	Semenov et al. 2013	Presurgical candidacy costs
CI surgery (Unilateral)	6010	1202	Semenov et al. 2013	Unilateral implantation costs (excluding system costs)
CI surgery (Bilateral)	9015	1803	Bond et al. 2009	Bilateral implantation costs (excluding system costs)
CI (Unilateral)	36162	7232	Semenov et al. 2013	Mean cost of unilateral cochlear implant system
CI (Bilateral)	57859	11572	Bond et al. 2009	Mean cost of bilateral cochlear implant system, assuming a 40% discount for the second implant
Maintenance yr1-3	1997	399	Semenov et al. 2013	Annual tuning and maintenance costs in year 1-3 including sound processor insurance
Maintenance yr4+	1579	316	Semenov et al. 2013	Maintenance costs in years 4+ including sound processor insurance
Upgrade	2976	595	Semenov et al. 2013	Processor update every 10 years
Major Complications	6259	1252	Semenov et al. 2013	Cost of major complications (unilateral)
Warranty costs				
Replacement of external component	-	-	Semenov et al. 2013	Cost of external component replacement is covered by insurance
Replacement of internal component (during warranty)	6010	1202	Semenov et al. 2013	Cost of internal component replacement

*Inflated to 2017 prices, based on the Consumer Price Index (Bureau of Labor Statistics)

†Variance of costs set to 1/5 of its mean value following Summerfield et al. (2010).

Table C. Estimates of the proportion of adult bimodal users that discontinue hearing aid use.

Study	Proportion of sample stopping HA use (%)	Sample size	Follow up	Source
Fielden et al. (2016)	59	38	5 years	Estimates from CI audiologists, UK
Neuman et al. (2017)	15	94	3 months	Unilateral CI adults, New York
Devocht et al. (2015)	36	77	1 year	Unilateral CI adults, Netherlands
Yamaguchi et al. (2013)	85	82	Not specified	Unilateral CI adults, Brazil
Fitzpatrick and Leblanc (2010)	51	96	6 months	Unilateral CI adults, Canada
Fitzpatrick et al. (2009)	21	24	6 months	Unilateral CI adults, Canada
Cowan and Chin-Lenn (2004)	21	71	Not specified	Unilateral CI adults, Australia