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# Cost-benefit Analysis of Cochlear Implants: A Societal Perspective

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**Objectives:** While the costs and outcomes of cochlear implantation (CI) have been widely assessed, most of these analyses were solely performed from the perspective of healthcare costs. This study assesses the costs and benefits of CI in the Netherlands from a broader societal perspective, including health outcomes, healthcare cost, educational cost, and productivity losses and gains.

**Design:** The cost and benefits of CI were analyzed in this cost-benefit analysis, in which a monetary value is put on both the resources needed and the outcomes of CI. The costs and benefits were analyzed by prototypical instances of three groups, representing the majority of cochlear implant patients: prelingually deaf children implanted at the age of 1, adults with progressive profound hearing loss implanted at the age of 40 and seniors implanted at the age of 70 with progressive profound hearing loss. Costs and benefits were estimated over the expected lifetimes of the members of each group, using a Markov state transition model. Model parameters and assumptions were based on published literature. Probabilistic and one-way sensitivity analyses were performed.

**Results:** In all three patient groups, the total benefits of CI exceeded the total cost, leading to a net benefit of CI. Prelingually deaf children with a bilateral CI had a lifetime positive outcome net benefit of €433,000. Adults and seniors with progressive profound hearing loss and a unilateral CI had a total net benefit of €275,000 and €76,000, respectively. These results ensue from health outcomes expressed in monetary terms, reduced educational cost, and increased productivity.

**Conclusions:** Based on estimates from modeling, the increased healthcare costs due to CI were more than compensated by the value of the health benefits and by savings in educational and productivity costs. In particular, for children and working adults, the societal benefit was positive even without taking health benefits into account. Therefore, CI generates an advantage for both patients and society.

**Key words:** Cochlear implants, Cost-benefit analysis, Sensorineural hearing loss

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## INTRODUCTION

Cochlear implantation (CI) can restore, at least partially, the cochlear function of patients with severe and profound hearing loss and have become the standard of care for this patient population (Bond et al. 2009). Increasing numbers of patients are eligible for CI since patients with increasing residual hearing profit from CI (Carlson et al. 2018; Snel-Bongers et al. 2018; Huinck et al. 2019). For example, the recent relaxation of selection criteria in the United Kingdom and Belgium will lead to an approximate 30% increase of patients eligible for CI (Van der Straaten et al. 2020a). Due to the substantial lifelong cost of implantation and maintenance, the total cost of CI will increase in the coming years. Given that the healthcare sector is budget constrained, the risk exists that not all patients eligible for CI will receive CI, or that the rising cost of CI will displace other healthcare treatments.

Despite the rising total cost, unilateral CI is considered cost-effective, as was shown in numerous cost-utility analyses of CI that were performed in various countries and various patient groups (O'Neill et al. 2001; Schulze-Gattermann et al. 2002; Summerfield et al. 2002; UK Cochlear Implant Study Group 2004; Barton et al. 2006b; Lee et al. 2006; Neilson 2006; Chen et al. 2014; Smulders et al. 2016). In addition, two critical reviews evaluated the effectiveness and cost-effectiveness of unilateral and bilateral CI in children and adults (Bond et al. 2009; Ontario 2018). Most of these studies took the healthcare perspective into account, and some also included educational cost of children with and without CI (O'Neill et al. 2001; Schulze-Gattermann et al. 2002; Barton et al. 2006b; Colletti et al. 2011). No study has taken other costs and benefits outside the healthcare sector (i.e., the societal perspective) into account, such as future productivity losses and gains. All these costs combined are called the lifetime societal costs. To put this cost in perspective, the lifetime societal costs of severe to profound hearing loss in the absence of implantation were estimated at $298,000 per person in the United States in 1998 (Mohr et al. 2000).

The healthcare perspective only provides insight into cost in the healthcare sector and does not take all benefits of CI into account. Besides improvements in educational level (De Raeve et al. 2015; van Weerdenburg et al. 2019) and reduction of educational cost (O'Neill et al. 2001; Barton et al. 2006b; Colletti et al. 2011), productivity also improves after CI (Kos et al. 2007; Monteiro et al. 2012). A cost-benefit analysis (CBA) with a societal perspective includes all these benefits and provides a comprehensive understanding of CI consequences. A healthcare perspective yields risks for suboptimal budgetary decision-making at the patient's expense, where a CBA taking a societal perspective can support decision-makers in maximizing social welfare (Krol & Brouwer 2014). This study aimed to include all relevant societal costs and benefits of CI in the Netherlands. Economic modeling generally entails assumptions about future costs and benefits. The wider the perspective that is adopted, the greater the number of such assumptions. For that reason, we

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made extensive tests of the sensitivity of conclusions to variation in parameters whose values were assumed.

## MATERIALS AND METHODS

This CBA from a societal perspective was performed for the situation in the Netherlands in two ways. First, the costs of healthcare, educational provision, and productivity were specific to the Netherlands. Second, Dutch data on healthcare benefits, educational placements, and employment patterns were used when available. Published literature was used for the input of the CBA. Where published literature was inconclusive or absent, the input was based on the opinions of a panel of experts, two of whom were also authors of this paper. The panel consisted of an otorhinolaryngologist specialized in CI (JF), a clinical physicist and audiological scientist (JJB) specialized in CI and a data manager and coordinator of CI. Approval by the Medical Research Ethics Committee was not required since no human subjects were involved.

The costs and benefits were calculated for three different groups, which together represent the majority of CI patients. These three groups were each represented by a prototypical instance of the whole group chosen by the expert panel to estimate the costs and benefits of the whole CI population. The prototypical instances of each group had an age of implantation based on the mean age of implantation of each group in our center since 2008. Group 1 comprises prelingually deaf children who underwent simultaneous bilateral implantation, and the prototypical instance is implanted at the age of one year. Group 2 refers to working-age adults (age 18–67) with progressive profound hearing loss and the prototypical instance had received an implant at the age of 40 years, and group 3 comprises retired seniors (age >67) with progressive profound hearing loss and the prototypical instance received an implant at age 70. Patients in groups 2 and 3 used a contralateral acoustic hearing aid together with their unilateral cochlear implant, which is standard practice in the Netherlands. Three equivalent patient groups without CI were modeled as control groups. These controls received regular care with bilateral hearing aids.

First, a classical cost-utility analysis was performed. In this analysis, the health outcomes and cost were compared between the treatment and control groups. Health outcomes were measured by means of health-related quality of life (HRQL) surveys and expressed on a scale where 1 corresponds to the state of full health and 0 to the state of being dead. The gain in HRQL, that is, the difference in HRQL between the treatment and the control group, is pivotal in the analysis. This gain and the length of life for which the gain is sustained were used to estimated quality-adjusted life years (QALYs) gained by the treatment. The primary outcomes of cost-utility analyses were the additional cost per QALY gained by the treatment. This is called the incremental cost-effectiveness ratio (ICER).

Second, the cost-utility analysis was extended to a CBA, in which all benefits and costs were expressed as monetary values. The health outcomes were converted to euros. Next, the health benefits were combined with benefits outside healthcare, such as increased productivity, resulting in the sum of all benefits. Finally, the sum of all costs was subtracted from the sum of all benefits to assess whether the benefits of the intervention exceeded its costs. This value was called the net benefit.

### Markov Model

Costs and benefits were estimated over the expected lifetimes of the members of each group using a Markov model. A Markov model is a model that is used in decision analysis for evaluating potential outcomes and costs of a disease. The model can estimate costs and benefits over a longer period using different states of health (so-called Markov states) and cycles. A patient is always in one health state, which has its own health benefits and costs. The transitions from one state to another may occur at the beginning of each cycle. The overall estimated costs and benefits depend on time (number of cycles) spent in each health state (Sonnenberg & Beck 1993). Therefore, a Markov model can assess cost and benefits when the timing of events is important, and when important events can recur, such as is the case in CI. For our two Markov models, in total four different states were used (see Fig. 1): bilateral CI, unilateral CI, inactive cochlear implant (i.e., no CI), and deceased. Transitions from bilateral to unilateral and from unilateral to inactive were caused by internal device failure. When internal device failure occurred, the costs and benefits of unilateral (group 1) or no CI (groups 2 and 3) were used for one cycle. In the next cycle, the original state was restored by a transition from the state of inactive cochlear implant to unilateral cochlear implant or from unilateral cochlear implant to bilateral cochlear implant. A cycle length of one year was used. The probability of dying at a certain age was obtained from Statistics Netherlands, which based this probability on national survival statistics (Statistics Netherlands 2018b) (Supplemental Digital Content 1; http://links.lww.com/EANDH/A762).

Tables 1 and 2 show the underlying model assumptions. Analyses were performed with Microsoft Excel 365 (Microsoft, Redmond, WA). Results above €10,000 were rounded to the nearest €1000, and results below €10,000 were rounded to the nearest €500. Moreover, the validity of the conceptual model, the input data, the computerized model, and techniques of validation were assessed with the AdViSHE (Vemer et al. 2016) checklist (see Supplemental Digital Content 2, http://links.lww.com/EANDH/A766).

### Benefits

QALYs were expressed in monetary terms. Policymakers in many countries have put a value on a QALY gained insofar as they specify the maximum amount that they are prepared to spend to gain a QALY. According to Dutch reference values, a QALY is valued at €20,000, €50,000, or €80,000, depending on a low, medium, or high burden of disease. The burden of disease is expressed on a scale where 0 corresponds to no loss of quality of life and 1 to a complete loss of quality of life. The burden of disease is classified as low 0.1–0.4, medium 0.41–0.7, and high 0.71–1. In the base-case analysis, we used the middle value of €50,000 per QALY, based on the values 0.42 (group 1) and 0.43 (group 2 and 3) (Bobinac et al. 2013; Zwaap et al. 2015). These values were calculated by subtracting the HRQL of no CI (Table 2) from 1.

The expert panel selected the gain in HRQL for the three groups from recent literature, using the following criteria: (1) The paper had to include not only the gain in HRQL but also the absolute HRQL pre and posttreatment; (2) the HRQL had to be reported in tables or text, not only in graphs; (3) preferably the HRQL had to be measured in a Dutch population; (4)

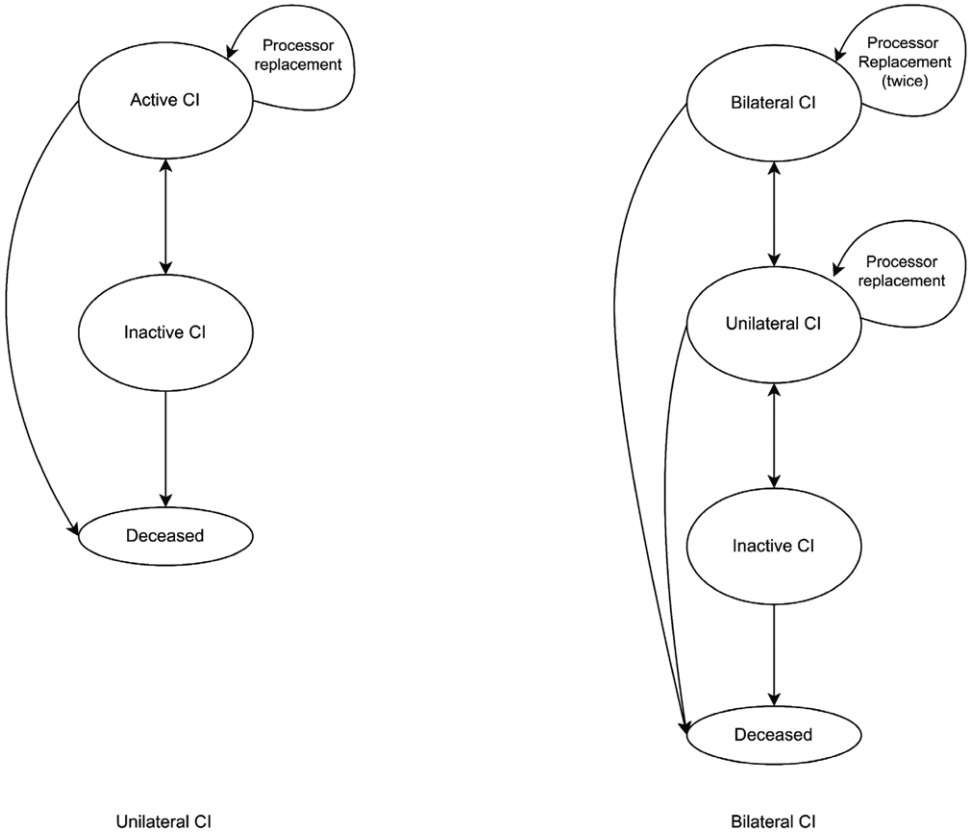


Fig. 1. Markov models for unilateral CI (left) and bilateral CI (right). CI indicates cochlear implantation.

When more than one study was available, the most recent study was chosen since changing implantation strategies influence the HRQL gain. Prelingually deaf children (group 1) are expected to have a higher HRQL gain in more recent studies because the age at implantation has declined over the years. Post lingually deaf adults and seniors (groups 2 and 3) are expected to have a decline in HRQL gain in recent studies since more patients with residual hearing are candidates for CI.

For the group of prelingually deaf children, several studies were identified (Barton et al. 2006b; Lovett et al. 2010; Summerfield et al. 2010; Sparreboom et al. 2012; Pérez-Martín et al. 2017). The study of Sparreboom et al. (2012) was selected with a gain in HRQL of 0.13 for unilateral CI and 0.18 for bilateral CI. These HRQL gains were based on the Health Utility Index 3 (Horsman et al. 2003) in a Dutch population of sequentially bilaterally implanted prelingually deaf children. For the two adult groups, several studies were identified (Summerfield et al. 2002; Summerfield et al. 2006; Bichey & Miyamoto 2008; Klop et al. 2008; Chen et al. 2014; Smulders et al. 2016). The study of Smulders et al. (2016) was selected with a gain in HRQL of 0.17 for unilateral CI and 0.21 for bilateral CI. These utilities were also based on the Health Utility Index 3 in a Dutch population. A detailed overview of the used HRQL values is shown in the Table in Supplemental Digital Content 4, http://links.lww.com/EANDH/A765.

Other benefits of CI, such as the potential reduction of the development of psychopathology in children (Netten et al. 2018) or a raised perception of autonomy in elderly patients (Sonnet et al. 2017), could not be expressed in monetary terms and were therefore not included in the model.

## Costs

Parallel to the benefits, an analysis of costs is performed to determine the incremental costs of treatment. All costs are reported in Euros, adjusted to 2018 price levels. Future costs and health outcomes were discounted at annual rates of 4% and 1.5%, respectively, in line with the guidelines of the Dutch National Health Care Institute (Zorginstituut Nederland) (IJzerman et al. 2016). The use of different or the same discount rates for costs and benefits is part of scientific debate (e.g., Attema et al. 2018); for that reason, we have modeled the effect of using the same discount rate in the sensitivity analysis.

The Markov model contained three different types of costs: healthcare cost, educational cost, and productivity cost. In the Netherlands, the cost of healthcare is bundled in diagnosis treatment combinations (DTC). Since a DTC also includes the cost of rectifying complications, the incidence and cost of complications were not analyzed separately. Only internal device failure, which may be seen as a complication, is analyzed separately because it has its own DTC due to the high cost associated with reimplantation. For CI, there are several DTCs: for the initial implantation, for the replacement of the external processor, which occurs once every five years, and for CI aftercare, which occurs once every three years. The charge for a DTC is negotiated between hospitals and health insurance companies. We used the average charges of all hospitals in 2018 published by the Dutch Health Authority (Dutch Health Authority 2018). The charge for a DTC is used as a proxy for the real healthcare cost. However, we are aware that the real cost of healthcare and charges for DTC could deviate.

For internal device failure, cumulative device survival data ("Cochlear Nucleus Implant Reliability Report" 2017) were

**TABLE 1. Assumptions incorporated in the Markov models**

| Model assumptions | Source |
|---|---|
| CI nonuse rate: group 1 = 0%, group 2 and 3 = 1.7% | Clinical practice LUMC |
| Reimplantation after internal failure, duration 1 cycle | Expert panel opinion |
| CI aftercare hospital visit once every 3 yrs | Clinical practice LUMC |
| Processor replacement once every 5 yrs | Clinical practice LUMC |
| Hearing aid replacement once every 5 yrs | Clinical practice LUMC |
| Contralateral hearing aid in case of unilateral CI | Clinical practice LUMC |
| Utilities are stable over time | Group UK CIS, 2004 |
| Life expectancy is similar in CI and control group | Expert panel opinion, conform to other studies |
| Healthcare costs other than CI care are similar in CI and control group | Expert panel opinion, conform to other studies |
| Education is compulsory from 5 to 18 yrs | Legislation Netherlands |
| Employment is taken into account between 18 and 67 yrs | Conform Dutch retirement legislation |
| Bilateral CI are implanted simultaneously | Clinical practice LUMC |
| 2% employment loss per year in the control group, group 2 | Expert panel opinion |
| No difference in education and productivity between patients with bilateral and unilateral CI | Lack of data, Expert panel opinion |
| Cost of bilateral CI = 2× costs of unilateral CI | Expert panel opinion |

obtained from market leader Cochlear (Sydney, Australia) and extrapolated (see Supplemental Digital Content 3, http://links.lww.com/EANDH/A764). Two methods of extrapolation are included in the sensitivity analyses. If internal failure occurred and a new device was implanted, cost of reimplantation within 10 years after initial implantation were not counted as healthcare cost since the manufacturer paid these. If the reimplantation took place more than 10 years after initial implantation, the cost were counted as healthcare cost.

Second, educational cost were taken into account. In the Netherlands, children are obliged to attend education between the age of 5 and 18 years. Therefore, educational cost were only relevant for group 1. In the Netherlands, all prelingually deaf children without CI attend special education. There is not much literature about the percentage of children with CI that attend mainstream education in the Netherlands after an educational reform in 2014. Some studies have provided data on small sample sizes, indicating that 45–55% are attending mainstream education (Langereis & Vermeulen 2015; van Weerdenburg et al. 2019). This seems comparable to studies in Belgium (45–59%) and South Korea (46%), and lower than studies in France (67–88%), UK (76%) and Canada (79%) (Barton et al. 2006a; Uziel et al. 2007; Verhaert et al. 2008; Venail et al. 2010; De Raeve 2016; Bae et al. 2019; Ganek et al. 2020). However, all countries have different educational systems and selection bias is present in most studies. For that reason, the expert panel decided to use a not yet published study from our group, which uses epidemiological data of the education types of all prelingually CI children in the Netherlands from Statistics Netherlands, a governmental organization. Only 35% of the CI children attend mainstream primary education and 46% attend mainstream secondary education, which has several levels from occupational to preacademic (Van der Straaten et al. 2020b). When attending mainstream education, all children received additional support, as were all children attending mainstream education in the studies of Langereis et al. and van Weerderburg et al. The cost of the different education modalities were obtained from reports of the ministry and the inspectorate of education (Dutch Inspectorate of Education 2019).

Third, the cost of productivity of patients and their family members were calculated. The cost of productivity in our model comprised decreased or increased productivity due to CI and productivity loss of patients and informal caregivers due to hospital visits. Travel cost and cost of informal care were not included in the model due to the unavailability of scientific data concerning these topics.

Productivity losses and gains were valued using two different methods: first, the human-capital method, which reflects the employee perspective and multiplies all the working hours lost by the modal hourly wage for adults in employment in the Netherlands, and second, the friction-cost method, which is more conservative and reflects the cost for the employer by limiting productivity cost to the time that is needed to hire and train a replacement worker. The time needed to find and retrain a new worker is the friction period, in which an employer faces productivity losses. Only the productivity losses during the friction period are taken into account in this method (Krol and Brouwer 2014). The length of the friction period is based on national labor market characteristics and unemployment levels. In this study, we adhered to the Dutch guidelines, which advise a friction-cost period of 85 days (IJzerman et al. 2016).

In the primary result, the human-capital method is presented. Wage was used as a proxy for productivity. Average wages per age group were gathered from Statistics Netherlands (Statistics Netherlands 2018a).

In group 1, the prelingually deaf children, the productivity losses and gains are calculated in two stages. First, the proportion of CI patients and controls that are predicted to be employed was determined. Second, the percentage of the national modal income that was received by each employed group was determined. Both assumptions were based on the scarcely available evidence. A Dutch labor study reported, based on data from Statistics Netherlands, a 45% employment rate of people with hearing loss between ages 15 and 64 compared with 65% employment rate in normal-hearing subjects (Josten et al. 2007). In addition, a Canadian and a Spanish study using retrospective questionnaires described an increase in employment rate after implantation from 62 to 75% and 63 to 83% (Clinkard et al. 2015; Huarte et al. 2017). The difference in wage was also described in the Dutch and Canadian studies. The Dutch study reported that based on a literature review, the wage of people with hearing loss was 66% of the national modal income (Josten et al. 2007). In the Canadian study, the postimplantation income increased by $10,000 compared with preimplantation (Clinkard et al. 2015). Although none of those studies was performed in prelingually deaf patients, the expert panel assumed that 45% of the controls and 65% of the CI patients were predicted to

**TABLE 2. Model input parameters**

| Parameter | Value | SE | Alpha | Beta | Distribution | Source |
|---|---|---|---|---|---|---|
| **HRQL** | | | | | | |
| CI children | 0.71° | | 7.6 | 3.1 | beta | Sparreboom et al. 2012 |
| Bilateral CI child | 0.76° | | 16.7 | 5.3 | beta | Sparreboom et al. 2012 |
| No CI child | 0.58° | | 26.4 | 19.9 | beta | Sparreboom et al. 2012 |
| CI adult | 0.74° | | 15.2 | 5.4 | beta | Smulders et al. 2016 |
| Bilateral CI adult | 0.78° | | 12.7 | 3.6 | beta | Smulders et al. 2016 |
| No CI adult | 0.57° | | 26.2 | 18.9 | beta | Smulders et al. 2016 |
| **Costs** | | | | | | |
| DTC CI implantation | €43,345 | 5529* | 61.5 | 705.2 | gamma | Dutch Health Authority |
| DTC CI aftercare | €820 | 105* | 61.5 | 13.3 | gamma | Dutch Health Authority |
| DTC processor replacement | €9915 | 1265* | 61.5 | 161.3 | gamma | Dutch Health Authority |
| Hearing aid (1 side) | €2000 | 255* | 61.5 | 32.5 | gamma | Expert panel opinion |
| **Education** | | | | | | |
| % CI mainstream primary edu | 35% | 0.04* | 32.7 | 38.4 | beta | Van der Straaten et al. (2020b) |
| % CI mainstream secondary edu | 46% | 0.06* | 39.6 | 73.5 | beta | Van der Straaten et al. (2020b) |
| Special education | €40,341 | 5146* | | | normal | Dutch Inspectorate of education |
| Additional support | €2225 | 284* | | | normal | Dutch Inspectorate of education |
| Primary education | €5300 | 676* | | | normal | Dutch Inspectorate of education |
| Secondary education | €6400 | 816* | | | normal | Dutch Inspectorate of education |
| **Productivity** | | | | | | |
| Modal income 18–24 yr | €14,685 | | | | | Statistics Netherlands |
| 25–29 yr | €27,723 | | | | | Statistics Netherlands |
| 30–34 yr | €35,331 | | | | | Statistics Netherlands |
| 35–39 yr | €40,192 | | | | | Statistics Netherlands |
| 40–44 yr | €43,436 | | | | | Statistics Netherlands |
| 45–49 yr | €44,921 | | | | | Statistics Netherlands |
| 50–54 yr | €44,951 | | | | | Statistics Netherlands |
| 55–59 yr | €44,382 | | | | | Statistics Netherlands |
| 60–64 yr | €42,297 | | | | | Statistics Netherlands |
| 65–67 yr | €28,801 | | | | | Statistics Netherlands |
| Daily income | €128 | 16* | | | normal | Statistics Netherlands |
| **Group 1 employment** | | | | | | |
| % of wage controls | 66% | 0.08* | 20.2 | 10.4 | beta | Expert opinion based on Josten et al. 2007 |
| % of wage CI | 85% | 0.11* | 8.4 | 1.5 | beta | Expert opinion based on Clinkard et al. 2015 |
| % of employment controls | 45% | 0.06* | 33.4 | 40.8 | beta | Josten et al. 2007 |
| % of employment CI | 55% | 0.07* | 27.1 | 22.2 | beta | Expert opinion based on Josten et al. 2007 |
| **Group 2 employment** | | | | | | |
| Baseline % employed | 50% | | | | | Kos et al. 2007 |
| Proportion promotion | 6% | | | | | Kos et al. 2007 |
| Proportion no promotion | 30% | | | | | Kos et al. 2007 |
| Unemployment rise (%/yr) | 2% | | | | | Expert opinion based on Kos et al. 2007 |
| **Economic** | | | | | | |
| Discount rate costs | 4% | | | | | Dutch National Health Care Institute |
| Discount rate effect | 1.5% | | | | | Dutch National Health Care Institute |
| QALY value | €50,000 | | | | | Pomp et al. 2014, Bobinac et al. 2013 |
| Intern device failure | Shown in SDC 3 | | | | | Cochlear CSR |
| Life expectancy | Shown in SDC 1 | | | | | Statistics Netherlands |
| Time horizon | Lifetime | | | | | |

*For variables with a beta or gamma distributions, alpha and beta parameters were calculated.*Estimated standard error, based on 95% confidence interval ±25% of value; °, see SDC 4 for more detailed information about the used HRQL values.HRQL indicates health-related quality of life; SDC, supplemental digital content; SE, standard error.*

become employed at a working age. Of these employed people, the wages were assumed to be 66% (controls) and 85% (CI patients) of the national modal income.

In group 2, the adult patients developed their hearing loss during their working life. Therefore, assumptions for this group were based on a Swiss study (Kos et al. 2007) on professional occupation after CI. In this retrospective study, approximately half of the patients had been unemployed before implantation and remained unemployed afterward. The professionally active patients largely maintained their activities after implantation and only a minority had stopped working (Kos et al. 2007). It was assumed that 50% of the CI patients had lost their employment before implantation and remained unemployed after implantation, that 14% would become unemployed in the 7 years (2 percentage point/year) after implantation, that 30% would retain their current employment and 6% would retain their employment as well as gain promotion. In the control group, it was assumed that 50% were unemployed at the starting point of the modeling and that the other 50% would gradually become unemployed in the 25 years after the starting point (2 percentage point/year).

In group 3, which assumes implantation at 70 years of age (i.e., after retirement), productivity and education are not relevant. Therefore, in this group, the societal perspective was identical to the healthcare perspective.

All input variables of the Markov model are shown in Table 2.

### Sensitivity Analyses

The impact of parameter uncertainty was tested with a probabilistic sensitivity analysis. In this analysis, all input parameters were randomly varied to simulate the events that occurred to each member of a cohort of 1000 patients. To vary the parameters, distributions were set around the parameters based on the median or mean and standard error when normally distributed and based on the alpha and beta parameters in case of a beta or gamma distribution. When the standard error was not accessible, we assumed that the mean or median ±25% would be representative of the 95% confidence interval around the mean or median. Alpha and beta parameters of the beta or gamma distributions were calculated using the median/mean and the estimated standard errors (Briggs et al. 2006). The results of the probabilistic sensitivity analysis are presented in an ICER scatterplot, in which every dot is a simulation of the model with randomly chosen, slightly different input variables. This plot shows the distribution of outcomes over four quadrants. The upper right quadrant represents better health outcomes at higher costs, whereas the lower right quadrant represents better health outcomes at lower costs. The left quadrants represent worse health outcomes at higher (upper) or lower (lower) costs. In addition to the ICER scatterplot, a cost-effectiveness acceptability curve (CEAC) is presented. This curve shows the probability that the intervention is cost-effective compared with the alternative given a certain monetary value for a QALY gained. This is called the willingness-to-pay threshold. The curve, therefore, represents the percentage of the 1000 samples (Y axis), which are considered as cost-effective at increasing willingness-to-pay thresholds (X axis). Confidence intervals of incremental costs, QALYs, and the net benefit were calculated using the 2.5th and 97.5th percentile of the 1000 samples (Wood 2004).

In addition, the influence of single parameters on the model outcomes was checked with one-way sensitivity analyses, in which one parameter is varied at a time to assess the impact of the change of that specific parameter on the model results. The results of the one-way sensitivity analyses are plotted in a so-called Tornado diagram, which shows the impact of varying each parameter with the parameters ranked from the most to the least influential.

## RESULTS

The results for each group are presented as the net benefit, which is the sum of all costs subtracted from the sum of all benefits. All costs and benefits are expressed in euro per person and are aggregated over the expected lifetimes of patients following implantation. Table 3 shows the results of the classic cost-utility analyses of each group calculated with the fixed parameters that are shown in Table 2. Table 4 shows the outcomes of the CBA for all groups based on the fixed parameters.

### Group 1: Prelingually Deaf Children

The results for the prelingually deaf children are shown in Tables 3 and 4 and Fig. 2A. The direct healthcare cost of CI were more (€173,000) than the healthcare cost in the control group. In contrast, the educational cost (€118,000) and the productivity cost (€61,000) were lower in the CI group. The ICER of bilateral CI versus no CI was negative. This indicates that bilateral CI is both more effective and less costly than no CI. When the QALY cost, which were in favor of CI, are included (€425,000), the net benefit of bilateral CI compared with no CI was €433,000 (95% confidence interval −€166,000; €936,000) at €50,000 per QALY, as is shown in Table 4.

Compared with unilateral CI, bilateral CI was estimated to have a considerably lower net benefit at €38,000 (95% confidence interval −€683,000; €801,000). Healthcare cost and QALY gains for unilateral CI are about half those for bilateral CI, but in the model, we have used the same educational and productivity cost. The ICER of bilateral versus unilateral CI was estimated at €34,000/QALY. This indicates that societal costs for bilateral CI, although higher than those for unilateral CI, are estimated to be below the willingness-to-pay threshold of €50,000/QALY.

### Group 2: Adults With Progressive Profound Hearing Loss

The results of the second group are shown in Tables 3 and 4 and Fig. 2B. The total costs to society of providing CI were similar to the costs of not providing CI, as healthcare cost of CI were equivalent to the productivity gains resulting from CI. The ICER of CI versus no CI was €200/QALY, indicating a considerable QALY gain at negligible societal costs. Combined with the health benefits, the calculated net benefit of CI was €275,000 (95% confidence interval −€110,000; €604,000).

In an additional analysis, the age of implantation was varied, that is, the indication for CI at different ages was used as a starting point for modeling the costs. The results are shown in Table 5. The indication of a CI at any age during a person's working life had a positive net benefit from the societal perspective. In addition, early treatment after eligibility for CI led to

**TABLE 3. Results of the cost-utility analysis of the base-case calculated with fixed parameters (shown in Table 2)**

| | Incremental costs (95% CI) | Incremental QALY (95%CI) | ICER |
|---|---|---|---|
| **Group 1** | | | |
| BCI vs. no CI | –€7000 (–98,000;77,000) | 8.5 (–2.5;18.9) | –€800 |
| BCI vs. CI | €80,000 (65,000;97,000) | 2.4 (–11.4;17.5) | €34,000 |
| **Group 2** | | | |
| CI vs. no CI | €1000 (–14,000;16,000) | 5.5 (–1.8;12.0) | €200 |
| BCI vs. CI | €74,000 (60,000;89,000) | 1.2 (–7.8;9,5) | €64,000 |
| **Group 3** | | | |
| CI vs. no CI | €64,000 (53,000;78,000) | 2.8 (–0.6;5.9) | €23,000 |
| BCI vs. CI | €58,000 (46,000;70,000) | 0.3 (–4.1;4.1) | €220,000 |

*Incremental costs show the point estimate and 95% confidence interval of the sum of the monetary values assigned to the costs and cost-savings incurred in healthcare, education, and work.BCI indicates bilateral CI; 95% CI, 95% confidence interval based on 2.5th and 97.5th percentile; ICER, incremental cost-effectiveness ratio; QALY, quality-adjusted life year.*

**TABLE 4. Results of the cost-benefit analysis of the base case calculated with fixed parameters (shown in Table 2)**

| | Healthcare cost | Educational cost | Productivity cost | QALY | Net benefit (95%CI) |
|---|---|---|---|---|---|
| **Group 1** | | | | | |
| BCI vs. no CI | €173,000 | −€118,000 | −€61,000 | −€425,000 | **€433,000 (−€166,000;€936,000)** |
| BCI vs. CI | €80,000 | – | – | −€118,000 | **€38,000 (−€683,000;€801,000)** |
| **Group 2** | | | | | |
| CI vs. no CI | €83,000 | – | −€81,000 | −€276,000 | **€275,000 (−€110,000;€604,000)** |
| BCI vs. CI | €74,000 | – | – | −€58,000 | **−€16,000 (−€454,000;€399,000)** |
| **Group 3** | | | | | |
| CI vs. no CI | €64,000 | – | – | −€141,000 | **€76,000 (−€101,000; €228,000)** |
| BCI vs. CI | €58,000 | – | – | −€13,000 | **−€45,000 (−€263.000;€142,000)** |

*The total incremental costs are divided over four cost types. Benefits are presented as negative costs. QALYs are expressed in monetary terms €50,000/QALY. Net benefit shows the sum of all cost categories.BCI indicates bilateral CI; 95% CI, 95% confidence interval based on 2.5th and 97.5th percentile.*

higher societal benefits. The lower net benefit for CI at an age of 20 years is largely caused by the lower modal income at this age.

Compared with unilateral CI, bilateral CI was not cost-effective, provided that we have used the same productivity cost for unilateral and bilateral CI. The health gain was 1.2 QALY. This resulted in an ICER of bilateral CI versus unilateral CI €64,000/QALY. The net benefit was −€16,000 (95% confidence interval −€454,000; €399,000).

### Group 3: Seniors With Progressive Profound Hearing Loss

The results of group 3 are depicted in Tables 3 and 4 and Fig. 2C. In this group, which assumes implantation at 70 years of age (i.e., after retirement), productivity and education are not relevant. Healthcare cost were somewhat similar to those of the other groups, but the QALY gain was lower due to these seniors' shorter life span after implantation. The health gain was estimated at 2.8 QALYs for unilateral CI and an additional 0.3 QALYs for bilateral CI. As a result, the cost-effectiveness of CI in this population was less than for the other groups. Still, the ICER of CI versus no CI was at an acceptable €23,000/QALY (95% confidence interval −€101,000; €228,000). However, the ICER of bilateral CI versus unilateral CI was calculated at an unacceptably high €220,000/QALY, with a negative net benefit of −€45,000 (95% confidence interval −€263.000; €142,000).

### Sensitivity Analyses

Table 6 shows the comparison of the cost calculated with the human-capital method, which were presented in the results above and the cost calculated with the friction-cost method. As expected, the net benefit was reduced when the more conservative friction-cost method was used. The productivity cost dropped because the human-capital methods included the productivity loss during the patients' future working life, whereas the friction-cost method only took into account the productivity losses during a period of 85 days. However, the net benefit for CI remained positive.

In addition, a probabilistic sensitivity analysis was performed for each group. The results are shown in Figure 3 in ICER scatterplots and a CEAC.

The majority of the outcomes for group 1 (Fig. 3A1) are situated in the lower right quadrant (75%). This figure shows a wide spread of both costs and QALYs gained, but overall the comparison of BCI versus no CI is cost-effective at a willingness-to-pay thresholds of €20,000 and €50,000 per QALY gained. The CEAC (Fig. 3A3) shows a 94% probability of cost-effectiveness at a willingness-to-pay level of €20,000. The outcomes for group 2 (Fig. 3B1) and 3 (Fig. 3C1), which are mainly situated in the upper right quadrant, have less uncertainty than those of group 1 due to the shorter life span after implantation in these groups because the CI was performed at an older age. In group 2, the CEAC (Fig. 3B3) was similar to the CEAC of group 1, insofar group 2 compared CI versus no CI and group 2 BCI versus no CI. Both are far below the threshold for reimbursement (€50,000) in the Netherlands (Zwaap et al. 2015). However, the CEAC of group 3 (Fig. 3C3) presents an 84% probability of cost-effectiveness for CI at a willingness-to-pay level of €50,000. This probability rises to 90% at a willingness-to-pay level of €80,000.

The probabilities of cost-effectiveness for bilateral CI compared with unilateral CI are depicted in columns 2 and 3 of Figure 3. At a willingness-to-pay level of €20,000, the probability of cost-effectiveness is 42%, 30%, and 9% for groups 1, 2, and 3, respectively. This probability increases to 53%, 50%, and 32% at a willingness-to-pay level of €50,000, and to 55%, 55%, and 41% at a willingness-to-pay level of €80,000.

The results of the one-way sensitivity analyses are shown in Figure 4. For each group, a tornado diagram was plotted. In all groups, changing the gain in HRQL and the value per QALY had the most substantial impact on the net benefit. The discount rate also had a noticeable impact on the net benefit in groups 1 and 2, with a long lifetime. When both benefits and costs are discounted at the same rate of 4%, the net benefit remained positive but declined with €200,000, €130,000, and €35,000 in groups 1, 2, and 3, respectively. Notably, in group 3, the net benefit of CI turned slightly negative when the HRQL gain was reduced. Furthermore, the net benefit turned slightly negative when QALYs were valued at €20,000.

## DISCUSSION

This CI CBA from a societal perspective showed a positive net benefit for all prototypical instances of three patient groups that together are representative for the majority of CI patients: prelingually deaf children who underwent simultaneous bilateral implantation, adults with progressive profound hearing loss, and seniors with progressive profound hearing loss. The net benefit even remained positive when productivity cost were calculated with the conservative friction-cost method. All results should be interpreted with caution since they were based on a model that drew on data from several sources and made assumptions about future costs and benefits. The uncertain

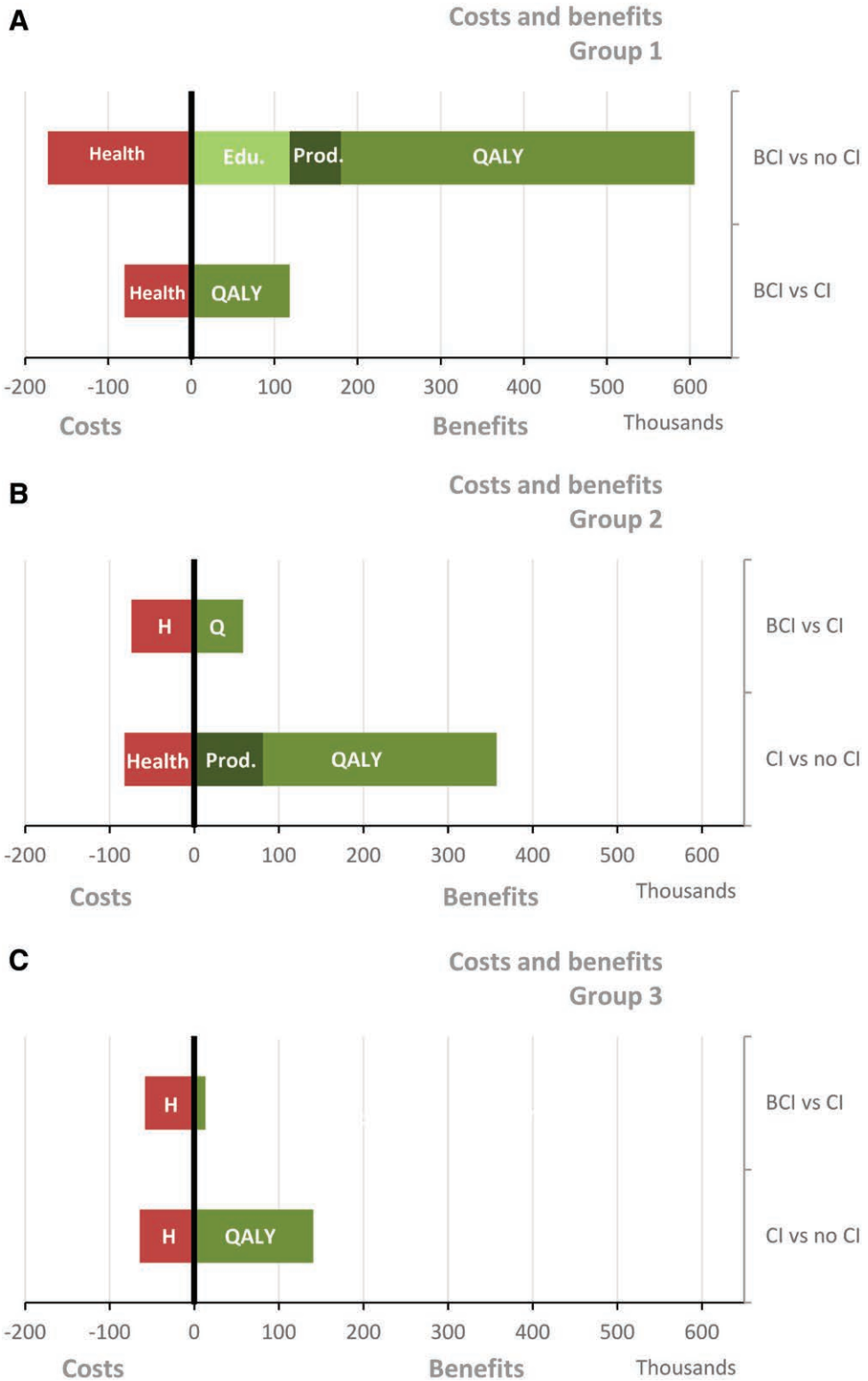


Fig. 2. Costs and benefits per group. BCI indicates bilateral CI; edu., education cost; H(ealth), healthcare cost; P(rod.), productivity cost; Q(ALY), QALYs gained in monetary terms.

nature of long-term modeling studies is reflected in the wide confidence intervals surrounding the net benefit.

To our knowledge, there are no comparable cost-benefit analyses of CI from a societal perspective in literature. However, several cost-utility analyses have been performed from the healthcare perspective. To enable a fair comparison between the results of our study and the outcomes of previous cost-utility analyses, our results need to be translated to a healthcare perspective by omitting the costs in other sectors. In group 1, the ICER from the healthcare perspective would have

**TABLE 5. Results Markov model group 2 (CI vs. no CI) with different age of indication for CI**

| | Incr. costs | Incr. QALY | ICER | **Net benefit** |
|---|---|---|---|---|
| 20 yr | €11,500 | 6.9 | €1700 | **€335,000** |
| 30 yr | –€23,000 | 6.3 | –€3600 | **€337,000** |
| 40 yr | €1000 | 5.5 | €200 | **€275,000** |
| 50 yr | €51,000 | 4.7 | €11,000 | **€182,000** |
| 60 yr | €74,000 | 3.8 | €19,500 | **€114,000** |

*ICER-indicates incremental cost-effectiveness ratio; Incr., incremental; QALY, quality-adjusted life year.*

**TABLE 6. Comparison of estimating productivity losses and gains with the Friction-cost method and the human-capital method**

| | Incr. costs | Incr. QALY | ICER | **Net benefit** |
|---|---|---|---|---|
| **Group 1** | | | | |
| Human capital | –€7000 | 8.5 | –€800 | **€433,000** |
| Friction costs | €54,000 | | €6500 | **€372,000** |
| **Group 2** | | | | |
| Human capital | –€1000 | 5.5 | €200 | **€275,000** |
| Friction costs | €79,000 | | €14,000 | **€197,000** |

*ICER indicates incremental cost-effectiveness ratio; Incr., incremental; QALY, quality-adjusted life year.*

been €20,000/QALY for BCI versus no CI. This is similar to the ICER in the Ontario HTA (Ontario 2018), which reported an ICER of €24,000/QALY* (*converted to euros and price level of 2018). In group 2, the present study's ICER from the healthcare perspective would be €15,000/QALY, which is comparable to the results of Chen et al. (2014) (€9400/QALY*) and somewhat lower than the HTA of Bond et al. (2009) (€26,000/QALY*). Since the results are sensitive to the estimated value of the HRQL gain, differences with the results of Chen et al. are likely to reflect the fact that they used a larger estimate of the HRQL gain (0.27) than Smulders et al. (0.21). For the population in group 3, the UK CI group (UK Cochlear Implant Study Group 2004) reported an ICER of €60,000/QALY*, which is remarkably higher than the ICER €23,000/QALY in this study. This difference is caused by higher healthcare cost (20%) and a smaller estimate of the HRQL gain (0.15) in the UK study (UK Cochlear Implant Study Group 2004).

From the societal perspective, which includes benefits of CI in other sectors than healthcare, the present study found that the ICERs improved. In group 1, CI was more effective and less expensive from the societal perspective compared with €20,000/QALY from the healthcare perspective. In group 2, the ICER improved from €15,000/QALY to €200/QALY from the broader societal perspective.

In this study, CI in group 1 was found to lead to a reduction in educational cost of approximately €118,000. By contrast, a study by Barton et al. (2006a), who meticulously calculated and compared the educational cost of CI patients and controls without CI in the United Kingdom, reported a difference in cost of €44,000*. However, this difference appears to be due to differences between the educational systems in the United Kingdom and the Netherlands. In the study of Barton et al., 76% of the CI children attended mainstream education, whereas in the Netherlands, this percentage is lower (35 and 46%) and the children receive additional support (van Weerdenburg et al. 2019). More importantly, in the British study, only 41% of the control group attended special education, whereas, in the Netherlands, all prelingually deaf children without CI receive special education.

In groups 2 and 3, the effects and costs of bilateral CI were also modeled and compared with the effects and costs of unilateral CI. In group 3, bilateral CI was not cost-effective (Fig. 3C2, C3). In group 2, the net benefit of bilateral CI was negative (Table 4 and Fig. 3B2, B3) with ICER €64,000, which is above the threshold of €50,000 used for evaluation in the Netherlands (Zwaap et al. 2015). Therefore, from the societal perspective, bilateral CI is just above the limits of cost-effectiveness. Other studies that considered the cost-effectiveness of bilateral CI, albeit from the healthcare perspective only, showed varying results. Smulders et al. (2016) reported bilateral CI to be cost-effective in the Netherlands; however, they did not discount these patients' future costs and health outcomes. Summerfield et al. (2010) and Bond et al. (2009) showed that the applicable thresholds for cost-effectiveness in the United Kingdom were not met for adults with profound hearing loss. By contrast, the Ontario HTA (Ontario 2018) concluded that bilateral CI in adults was cost-effective at commonly used willingness-to-pay thresholds. In this study, the difference in costs between unilateral and bilateral CI was caused by differences in healthcare cost and differences in utilities, while we were unable to find evidence for differences in productivity. In future research, these effects of bilateral CI should be investigated further.

In addition to the educational cost, productivity cost and gains were also included in the present CBA. In general, people with hearing loss are more likely to be unemployed, have higher levels of sick leave and need more time to recover from a working day than people with normal hearing (Kramer et al. 2006; Nachtegaal et al. 2009; Stam et al. 2013). Improving the hearing ability might positively affect employment, which can be important since employment can have major advantages from a patient's perspective. Being employed is associated with increased self-esteem and self-worth. Furthermore, work provides relationships and social connections and a higher level of social status (Jahoda 1982). Being unemployed is associated with poorer health status and negative psychological symptoms (Montgomery et al. 1999). Therefore, empowering people with hearing loss to become or remain employed might improve their lives, even without considering the financial and economic advantages. Most likely, however, these additional effects of employment will be expressed in the utilities used in the model and were therefore accounted for in the present analyses.

While studies on the effect of productivity of CI are scarce, they all concluded that CI can empower patients to improve or retain productivity (Kos et al. 2007; Clinkard et al. 2015; Huarte et al. 2017). The present study showed that the economic impact of this effect may be considerable, although more research into the effects of CI on productivity could improve the accuracy of the productivity predictions.

## Limitations

This CBA was based on the situation in the Netherlands. First, the cost of healthcare and the patterns of the educational provision were specific to the Netherlands. Second, there was a preference, where possible, to use data on healthcare benefits and employment patterns that had been gathered in the Netherlands. However, such data were not always available and it was necessary to source data from other countries: Canada,

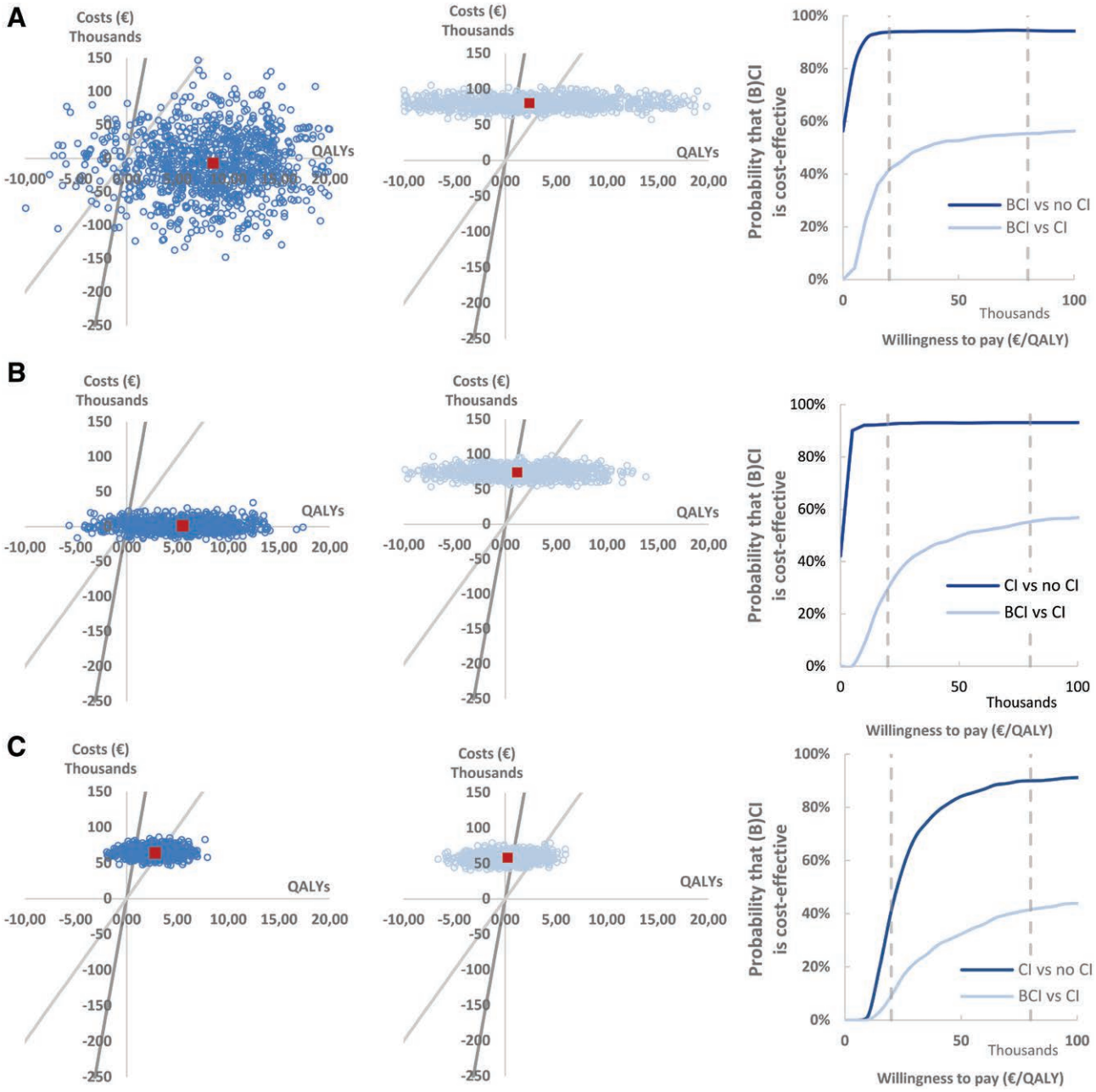


Fig. 3. Probabilistic sensitivity analysis. A, group 1; BCI vs. no CI (A1), BCI vs. CI (A2), and both CEACs (A3); B, group 2; CI vs. no CI (B1), BCI vs. CI (B2), and both CEACs (B3); C, group 3; CI vs. no CI (C1), BCI vs. CI (C2), and both CEACs (C3). In the scatterplots, the gray lines correspond the €80,000/QALY (dark) and €20,000/QALY (light). The red dot is the outcome of the base-case Markov model. BCI indicates bilateral CI; CEAC, cost-effectiveness acceptability curve; ICER, incremental cost-effectiveness ratio.

Spain, and Switzerland, for example. Given considerable variation in data between studies, the particular choices of these external sources may be critical. Since healthcare, education, and productivity cost vary across countries, the results may not be representative of other countries. However, as is shown in our sensitivity analysis, gain in HRQL was the most important factor that influenced the net benefit and it is likely that the gain in HRQL associated with CI is similar to values measured in the Netherlands. The second most important factor is the value of a QALY, which is country-specific. However, the value used in the sensitivity analysis (Table 4) of €20,000 is comparable with other countries such as the United Kingdom National Institute of Clinical Excellence guidelines of £20,000–30,000 per QALY (€22,700–34,100 in 2018).

The uncertainty of the net benefit is caused by both the uncertainty of input parameters and the uncertainty of the model structure (Walker & Fox-Rushby 2001). In this study, we used several sensitivity analyses to assess the impact of the first and, to some extent, the second type of uncertainty. Besides, the uncertainty which is inherent in long-term economic evaluations due to their lifetime horizon and discounting is presented in confidence intervals around costs, QALYs, and net benefit (Tables 3 and 4) and illustrated with ICER scatterplots and CEAC (Fig. 3).

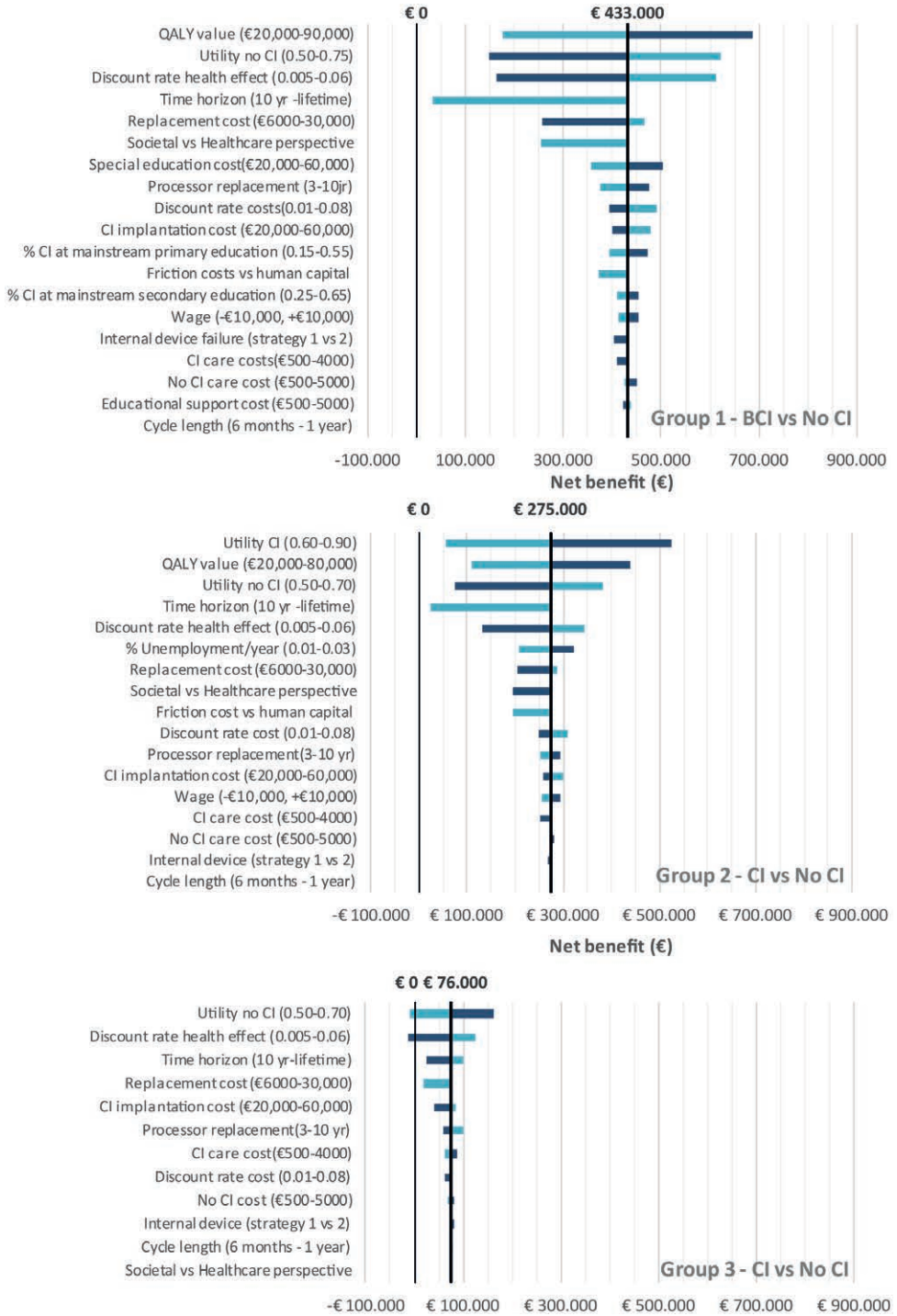


Fig. 4. Results of one-way sensitivity analyses for each group. The dark and light blue bars show the effect on the net benefit of adopting the high and low values of the input variables.

In addition, while a CBA expresses all costs and benefits in monetary terms, it can be difficult or even impossible to determine the monetary value of certain costs and benefits, such as the cost of informal care, the benefits of increased autonomy, and the value of health outcomes in general. For the latter, the present study used Dutch guidelines, which advice to value a QALY at €50,000 (IJzerman et al. 2016). However, the valuation of health outcomes can differ between countries. Furthermore, it matters whether QALYs are valued from the societal perspective or an individual perspective. In countries with collectively funded healthcare systems, such as the Netherlands, cost-benefit analyses should not use an individual's valuation of his or her health gain but the valuation of societal health gain in general (Bobinac et al. 2013).

Since every model is a simplification of reality, this study could not include all variables of influence in the model. For example, in group 3, the cost of informal care and the benefits of postponing residential care were not included since no scientific data are

available on these topics. In addition, assumptions have been made. If available, assumptions were based on scientific literature, and if literature was not available, they were based on expert opinion. Since expert-based assumptions limit the reliability of the model, all assumptions used in the present study were challenged in the sensitivity analyses. Future studies that will systematically analyze HRQL gain, educational placement, and employment patterns using a meta-analysis would be of great value.

To include the lifespan of the internal cochlear implant in the model, we used survival statistics provided by market leader Cochlear. These data cover a period of 30 years, but for the Markov model, we needed to extrapolate these data to 90 years. It might be that such an extrapolation is not realistic. For that reason, an additional analysis was performed in which the internal survival at 50 years after implantation was assumed to be 0% (see Supplemental Digital Content 3, http://links.lww.com/EANDH/A764). This had only a limited impact on the results (i.e., an increase in lifetime costs of approximately €7000 to €29,000), due to the discounting of long-term costs and benefits.

**Implications for Practice** • In 2018 in the Netherlands, 473 new patients underwent a CI, 66 of whom were children who underwent bilateral CI (CION 2018). The members of the expert panel maintained that the three broad categories of bilateral severely profoundly deaf candidates implanted as young children, adults of working age, and retired adults—intending to improve hearing—include 85% of patients currently implanted in The Netherlands. Consequently, for 2018, the group of prelingually deaf children represents 12% of all implantations, which accounts for 60 patients and a net benefit of 26 million euros. The group of adults with progressive hearing loss was estimated to represent 33% of all indications, which amounts to 156 patients and a net benefit of 43 million euros. The group of seniors with hearing loss was estimated to represent 40% of indications, which amounts to 189 patients and a net benefit of 14 million euros. Together, these patients with CI in 2018 represent an estimated lifetime net benefit of 83 million euros in the Netherlands.

## CONCLUSION

This CBA provided a comprehensive overview of the costs and benefits of CI. The majority of CI indications were included, and healthcare, educational, and productivity costs, and benefits were taken into account to model predictions of future costs and benefits. As expected, the Dutch, Belgium, and UK criteria for unilateral and bilateral CI were in line with the positive net benefit of all three groups. From a societal perspective, the costs of CI in prelingually deaf children were predicted to be lower than the costs of not implanting a CI, even without taking the health benefits into account. For adults with progressive hearing loss, the predicted costs of CI were equal to the costs of not implanting a CI, also without taking the health benefits into account. When the health benefits were taken into account, the advantages of CI outweighed those of care without CI, and CI provided clear benefits for both the patients and society.

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