A decision-analytic model to project the expected lifetime costs and quality of life (QoL) for individuals receiving bilateral cochlear implants during first life year

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List of abbreviations

CI	Cochlear implantation
DRG	Diagnosis-related groups
EQ-5D	EuroQoL-5D
HELFO	The Norwegian Health Economics Administration
HTA	Health Technology Assessment
ICER	Incremental Cost-Effectiveness Ratio
KINDL	Questionnaire for Measuring Health-Related Quality of Life in Children and Adolescents
NMB	Net Monetary Benefit
NHB	Net Health Benefit
NoMA	Norwegian Medicines Agency
OUH	Oslo University Hospital
PedSQL	Paediatric Quality of Life Inventory
PPT	Education and psychological counselling service
QALYs	Quality Adjusted Life Years
STA	Single Technology Assessment
Statped	Public special education service
WTP	Willingness-to-pay

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Abstract

Introduction: Deafness and hearing impairment are common conditions in infants with an estimated prevalence of hearing impairment of 1 - 2 per 1000 new-borns (Butcher et al., 2019; CDC, 2019; Uhlén et al., 2020). A core treatment for deafness or severe hearing loss is cochlear implantation (CI). Cochlear implants do not restore normal hearing, but CI can make it possible for children born deaf or with severe hearing loss to interpret sounds and communicate effectively (Bond et al., 2009). Several factors have shown to affect the language outcomes by CI such as implantation early in life and bilateral versus unilateral implantation. Auditory Verbal Therapy (AVT) is another intervention believed to improve language outcome after CI.

To date few economic analyses have described the health and economic consequences of hearing interventions over a lifetime. To our knowledge, there are no studies in Norway that have assessed the lifelong resource use and health related quality of life (HRQoL) after CI. Therefore, our objective was to conceptualize a disease simulation model that captured the lifelong costs and effects on HRQoL associated with standard of care for individuals identified with severe-to-profound hearing loss receiving bilateral CI <12 months with no other disabilities and an IQ-level above 75. Secondly, we performed an exploratory analysis to estimate the health effects, costs, and cost-effectiveness of AVT compared to standard of care.

Methods: We developed a Markov-model with three different health states related to language skills, and a state of death for a population of children who had received bilateral CI. Resource use from the health care sector, the education sector and production loss were included, and three different perspectives were applied, the health care, the extended health care, and the societal perspective. We relied on primary data from two Norwegian studies (one cross-sectional and one longitudinal study) to inform our health states, as well as the transition probabilities and HRQoL. For resource use, we relied on a combination of expert opinion, national guidelines, and national tariffs. Outcomes were discounted lifetime costs (2021 Norwegian Krone (NOK)) and quality adjusted life years (QALYs). Results from the exploratory analysis with AVT were given as ICERs and compared with willingness-to-pay thresholds of NOK 300 000 and NOK 500 000 to evaluate cost-effectiveness.

Results: The total discounted expected lifetime costs per patient, including all the sectors and production loss was NOK 2 375 698 (NOK 5 347 950 undiscounted). When we restricted the costs to the extended health care sector, the expected discounted costs decreased by more than half, i.e., to NOK 1 060 858 (NOK 2 160 826 undiscounted). Total discounted lifetime QALYS were 22.4 (60.5 undiscounted). In the exploratory analysis of a 1-year AVT program, from the broadest societal perspective, the AVT was considered cost-effective with an ICER of NOK 76 569, while from an extended health care perspective, the ICER was generally higher than the willingness to pay threshold, i.e., NOK 731 036 per QALY. When only medical costs in health care sector were included the ICER was NOK 163 823 per QALY indicating cost-effectiveness.

Conclusions: With the help of clinical experts in the field, for the first time in Norway, we developed a model to capture the long-term health and economic consequences for children receiving bilateral CI. Costs from different sectors were included, and we have obtained insights into the types of costs associated with cochlear implantation. The conceptualized model will be able to formally evaluate AVT programs once data become available. Core parameters of the CI model were uncertain, but the results indicate considerable lifetime costs of CI and lower HRQoL than in the general population. AVT may be a cost-effective means of improving language outcomes.

1 Introduction

Deafness and hearing impairment are common conditions in infants with an estimated prevalence of hearing impairment of 1 - 2 per 1000 new-borns (Butcher et al., 2019; CDC, 2019; Uhlén et al., 2020). The prevalence increases to 3- 4 per 1000 children by school age (Fortnum et al., 2001; Mehra et al., 2009).

Today, hearing loss often is detected early in life due to the implementation of screening of new-borns in many countries. Universal neonatal screening for hearing loss was implemented in Norway in 2008. Screening makes it possible to start interventions at an early age.

A core treatment for deafness or severe hearing loss is cochlear implantation (CI). A cochlear implant is an electronic device operated into the cochlea and works by directly stimulating the auditory nerve through electrodes. Even if cochlear implants do not restore normal hearing, CI can make it possible for children born deaf or with severe hearing loss to interpret sounds and communicate effectively (Bond et al., 2009).

It is well established that early identification and intervention of hearing loss in children may give improved speech and language outcomes (Fulcher et al., 2012; Karltorp et al., 2020). Studies have also shown that children with bilateral cochlear implantation achieve better language outcomes than those with unilateral implants (Boons et al., 2012; Jacobs et al., 2016; Lammers et al., 2014; Leigh et al., 2013; Sarant et al., 2014; Tait et al., 2010). Other studies have shown that simultaneous versus sequential implantation have better outcomes (Gordon et al., 2013; Kral et al., 2016). Early bilateral simultaneous implantation has been the standard in Norway since 2004 (Wie et al., 2020).

Educational training, e.g., Auditory Verbal Therapy (AVT), given after cochlear implantation is another factor that is believed to affect the language outcomes (Binos et al., 2021; Dettman et al., 2013; Dornan et al., 2010; Percy-Smith et al., 2018; Thomas & Zwolan, 2019). AVT programs are already established in several countries, for example in Denmark, UK and Australia as part of early interventions programs in hearing loss (Auditory Verbal UK, 2023; Firstvoice, 2023; Hallstrøm, 2022). In Denmark the training is given for 3 years, but the duration may differ between countries.

The importance of knowing more about the lifelong consequences of hearing loss, including patients with CI, has been emphasised in a recent systematic review by an US group of investigators (Borre et al., 2021). This review summarizes cost-effectiveness analyses of cochlear implantation that has been performed (Borre et al., 2021). These analyses had demonstrated cost-effectiveness of bilateral over unilateral implantation, CI versus hearing aids, simultaneous over sequential implantation, and for early implantation over later implantation. The authors, however, conclude that few economic analyses have investigating hearing interventions over a lifetime.

To our knowledge, no studies in Norway have assessed the lifelong resource use and HRQoL after CI. Together with a team closely involved in the field of hearing loss and CI, we decided to develop a Markov-model following children with CI over their lifetime. We included health states related to language skills and the associated health related quality of life to be able to project interventions at a later stage. Another discussion with the team was how language skills and AVT given early in life could influence HRQoL and other achievements later in life. The degree of speech and hearing abilities may affect many different aspects of life, as being able to attend a mainstream school, getting an education, having a job, or even getting a partner. All these events in life may influence the quality of life (QoL). Whether a person has a job or not have societal economic consequences. We therefore wanted to project AVT to the model.

First, we conceptualized a model that captured the lifelong costs and effects associate with standard of care for individuals identified with severe or profound hearing loss receiving bilateral CI <12 months with no other disabilities and an IQ-level above 75.

Secondly, we performed an exploratory analysis to evaluate the outcomes, costs, and cost-effectiveness of AVT given early in life compared to standard of care.

2 Background

2.1 Hearing loss and interventions

Prenatal hearing loss or deafness are mostly caused by genetic factors or maternal infections during pregnancy. Common causes in early childhood are ear inflammations (otitis media) and infections like meningitis (WHO, 2021).

The severity of hearing loss varies and can be categorized as mild, moderate, moderately-severe, severe, profound or complete. It can involve one or both ears. Which category a person belongs to depends on that person's hearing threshold measured in decibels (dB) (WHO, 2021).

There are two main types of hearing loss, sensorineural and conductive hearing loss. These two types refer to the cause and the location of the problem (WHO, 2021). It is also possible to have a mixed type with a combination of these two types of hearing loss. Sensorineural hearing loss is the most common and is caused by damage to the inner ear (cochlea = sense organ) or to the nerve pathways from the inner ear to the brain (PenTAG, 2007). Sensory hair cells located in the cochlea may be damaged or be lacking thus causing sensorineural hearing loss. Conductive hearing loss is caused by problems in the ear canal or middle ear reducing the transfer of physical energy from the ear drum to the inner ear (WHO, 2021).

A well-known treatment for hearing loss is acoustic hearing aids (HA). These can be used in both sensorineural and conductive hearing loss, but do not provide enough benefit for people with severe-to-profound sensorineural hearing loss. For these people cochlear implant (CI) may be a better option. Audiologists define severe-to-profound hearing loss as having hearing thresholds of 80-90 decibel and profound hearing loss as > 90 decibel, but the criterion for reimbursement of CI is broadening and some countries may have more flexible criteria (T. Busch, personal communication, 27-Aug-2021).

A cochlear implant is an electronic device and works by directly stimulating the auditory nerve through electrodes operated into the cochlea. Figure 2.1 (WHO, 2021) shows the different components of the implant. An external microphone senses sound and a speech processor (1) transforms this sound into electrical stimuli which are sent to an external transmitter (2). The external transmitter sends the electrical signals through the skin to an internal receiver (3). The stimuli are then processed before they go through a cable to reach the electrodes in the cochlea (4). The device makes it possible to bypass any damages in the middle- and inner ear structures.

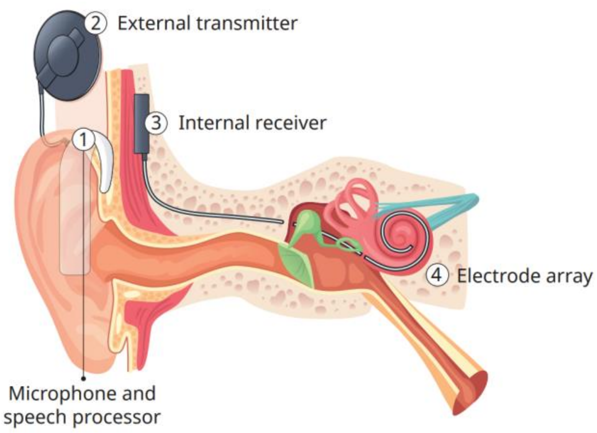

Figure 2.1 Illustration of the external and internal components of the cochlea implant
Reproduced from the World Report on Hearing (WHO, 2021).

Patients with cochlear implants require life-long follow-up due to refitting of the device, changing environments and to compensate for the brain's acclimatization to the CI. The limitations of the CI, especially in noisy conditions may also require accommodations in school and workplace either with technical equipment or support staff.

2.2 Factors influencing language outcomes

Receiving implantation early in life has shown to provide better language outcomes (Karltorp et al., 2020; Niparko et al., 2010; Purcell et al., 2021). A meta-analysis including 21 studies found the benefit of CI to be greatest when received before 12 months of age (Ruben, 2018). Further, bilateral, and simultaneous implantation have shown better results for language skills than unilateral and sequential implantation (Gordon et al., 2013; Kral et al., 2016). Another factor that is believed to affect the language outcomes is the educational training given after CI.

One educational intervention that has been found to be more effective than many other approaches is AVT (Auditory Verbal Therapy) (Binos et al., 2021; Dettman et al., 2013; Dornan et al., 2010; Percy-Smith et al., 2018; Thomas & Zwolan, 2019). AVT is (1) characterized by a focus on auditory learning, that is, "learning through listening", (2) it includes the family as an essential part of the intervention, and (3) constantly assess the child's language abilities and adapts the intervention accordingly. Although AVT is a flexible program that is adapted to each individual child's needs, it is a uniform program in the sense that it follows specific principles.

Several other factors influence language outcomes after CI, including additional disabilities, non-verbal IQ and family characteristics (Geers, 2002). The advances in technology since the first children received CI until today also contributes to better outcomes in general.

2.3 Cochlear implantation in Norway

The first operations with CI in Norway using intra cochlear implants took place in late 1986 (L.R. Opheim, personal communication, 18-Aug-2021). Unilateral implantation was common practice during the first years, but from 2004 bilateral implantation has been offered to all children if medical appropriate (Wie, 2010). All operations in children are centralized and performed at Oslo University Hospital, Rikshospitalet. CI may be performed as early as in 5-6 months old.

In Norway, approximately 65 children are born with impaired hearing or deafness every year. These cases include all severities and on average 33 of these children receive CI.

The children are followed up and trained in their local communities. The educational training, however, is not standardized and the amount, and the content of the training depends on where the children live. Type of training offered can be dependent upon economic resources and priorities in the municipality, caregivers' preferences, teachers, paramedical professional's expertise, or even ideological standpoints. Because the number of children with CI is relatively low, there is also a challenge with getting enough local expertise in a setting with a decentralized follow-up.

2.3.1 Auditory Verbal Therapy

An ongoing project, initiated at Rikshospitalet Oslo University Hospital (OUH) in 2022, offer all implanted children Auditory Verbal Therapy for one year after implantation. The training can be done either digitally or by physical meetings. However, both the short- and long-term implications of AVT are currently unknown in Norway but are essential to inform priority setting decisions.

2.4 Prioritization between health investments in Norway

In Norway, the three main criteria for prioritization between health interventions are resource use (costs), health benefit, and severity of the condition. Costs are expressed in Norwegian Krone (NOK) and health benefits in terms of quality adjusted life years (QALYs) and presented as the ratio of costs to QALYs. There exist no official thresholds for willingness to pay for QALYs, but it is accepted to use more resources by higher severity (Meld. St. 34 (2015–2016)). Based on UK studies, the estimate of the opportunity costs of one QALY is 275 000 NOK/QALY (Meld. St. 34 (2015–2016)).

2.5 Cost-effectiveness of cochlear implantation

The main type of analysis for priority setting in health care is cost-effectiveness analyses (CEAs) where costs and health effects of standard (status quo) treatments and new interventions are estimated and expressed as a cost-effectiveness ratio (cost per QALY). There is a growing number of cost-effectiveness analyses in the field of hearing loss. Borre at al. (2021) conducted a systematic review of economic analyses using models with all types of hearing interventions and identified 34 studies of cochlear implantation (Borre et al., 2021). Most of these analyses assessed bilateral versus unilateral implantation. Other studies assessed CI versus hearing aids, simultaneous versus sequential implantation, deaf education, or bimodal hearing technology (CI + HA). It also included a cost-utility study with a societal perspective (Semenov et al., 2013) showing that early implantation was more cost-effective when compared with later implantation.

The review mentioned above, concluded that there are few economic analyses investigating hearing interventions over a lifetime, and this represents an important gap in knowledge.

3 Aim and objectives

The primary aim of this study was to conceptualize and develop a decision-analytical model to enable the quantification of the lifetime health and economic consequences of current standard of care for infants receiving a bilateral CIs aged <12 months with severe-to-profound hearing loss with no other disabilities and an IQ-level above 75.

The secondary objective was to conduct an exploratory analysis to project outcomes of educational training interventions, such as Auditory Verbal Therapy (AVT), compared with standard of care training.

4 Theory

4.1 Health economic evaluation

Economic evaluation is performed to inform decision makers and stakeholders on how to use scarce resources in the best possible way. It can be defined as:

> *"The comparative analysis of alternative causes of action in terms of both costs and consequences" (Drummond et al., 2015, p. 4).*

The different types of economic evaluations are named after how, or in which units, the outcomes are measured. Examples are cost-effectiveness analyses (CEA), cost-utility analyses (CUA), and cost-benefit analyses (CBA).

In CEAs outcomes are measured in clinical ("natural") endpoints such as a drug's *effect* on blood pressure or a screening program's ability to detect cancer cases (Drummond et al., 2015, p. 7). CUAs have a generic outcome measure of *utility,* mostly quality-adjusted life years (QALYs). QALYs are based on both quality and length of life. With a generic measure, it is possible to compare costs and outcomes between different economic evaluations independently of disease or health condition. A CUA is a variant of a CEA, and the term CEA is more often used. In contrast, CBAs the outcomes are translated into monetary values. Prices related to the health benefits are found through willingness-to-pay studies.

QALY is the main outcome measure used in health technology assessments (HTAs). A QALY is a generic measure and is used across different diseases and conditions. It includes effects on both the length and the HRQoL. An intervention may affect both HRQoL and longevity or only one of them. To calculate the QALY, the quality of life is multiplied by the length of life. For example, if the quality of life under a disease is 0,8 in 1 year, the QALY is 0,8 x 1 year = 0,8 QALYs. Figure 4.1 (Drummond et al., 2015) below illustrates the two dimensions of the QALY. The X-axis represents the length of life, and the y-axis the HRQoL, which may take on a number between 0 and 1. The figure demonstrates the QoL and length of life with and without a treatment, A or B.

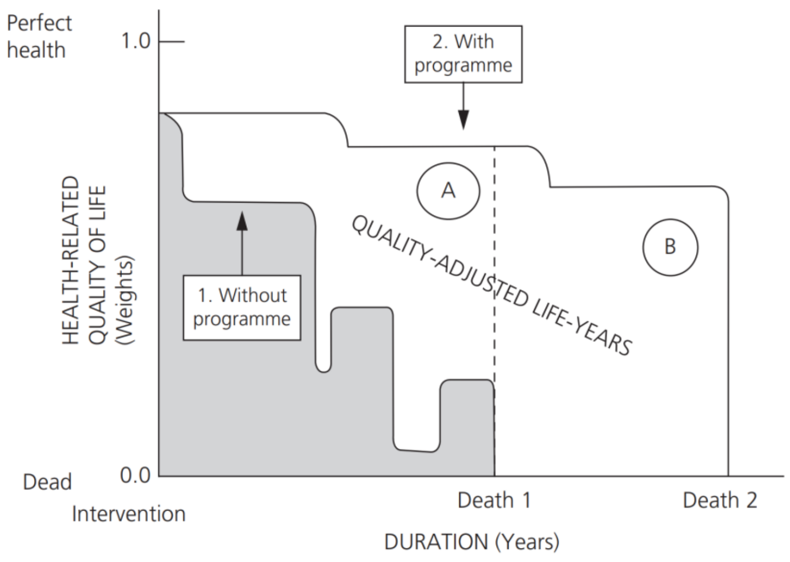

Figure 4.1 The two dimensions of the QALY-measure; length and HRQoL.
Reproduced from Drummond et al, 2015.

4.2 Measuring Health-Related Quality of Life in hearing loss

HRQoL may be measured by either disease-specific or generic questionnaires. Generic questionnaires need to be general and simple enough to apply across diseases and may not be as sensitive as disease-specific measures to capture changes in HRQoL. However, an advantage with generic measures is that HRQoL across different types of diseases more easily can be compared (Hunink et al., 2014).

The most used generic instrument for HRQoL, the EQ-5D (Rabin & de Charro, 2001), has been reported to have poor validity and responsiveness in hearing disorders (Finch et al., 2018). The standard version of the EQ-5D is not developed for children, but alternatives are available including a Norwegian version for children 8-11 years (EQ-5D-Y) (Oslo University Hospital (OUH), 2016).

The assessment of HRQoL in children can be proxy- or self-reported. For young children under the age of 3, there are only proxy reported HRQoL, i.e., conducted by caregiver. The recommended age for self-reporting varies between different instruments. In a systematic literature review by (Mpundu-Kaambwa et al., 2022), the minimum recommended age for self-reporting varied from 7 to 12 years between 7 different instruments.

Several different instruments are being used in the assessment of HRQoL in children with hearing loss, but no single tool has become the standard (Roland et al., 2016). Two common instruments are the KINDL and the PedSQL (T. Busch, personal communication, 23-Sep-2021) and data from both are available from Norwegian studies. These instruments are described below.

4.2.1 The KINDL

The KINDL is an generic instrument in the form of a questionnaires for measuring HRQoL in children aged 4-17 years (Ravens-Sieberer & Bullinger, 1998). The questionnaire is available in a self-reported form from age 4 and a proxy-reported version. KINDL has six dimensions: Physical well-being, Emotional well-being, Self-esteem, Family, Friends, and Everyday functioning (school or nursery school/kindergarten). Each dimensions have four items each, making it a total of 24 items. The total score is based on those 24 items. Additionally, several modules with 6 items have been developed to assess HRQoL in chronic diseases: one module for chronically ill hospitalized children and 7 other disease-specific modules. No module has been developed for hearing, but the 6-items questionnaire was adapted to be used in the 500-project. The maximum score is 100.

4.2.2 The Pediatric Quality of Life Inventory

The Pediatric Quality of Life Inventory (PedsQL) is, as the KINDL, a generic instrument. (PedsQL, 2022). The targeted age group is 2 – 18 years. The questionnaire has four scales functioning dimensions: Physical (8 items), Emotional (5 items), Social (5 items) and School (5 items). PedSQL has both a self-reported and proxy-reported version. The child may self-report from age 5 years. Disease specific modules for have been developed for six diseases, and more are in development. The maximum score is 100.

4.3 Measuring language outcomes in hearing loss

HRQOL is a core outcome for any health intervention, but for CI the ability to hear enough to communicate by speech is crucial. Language skills may be measured through standardized questionnaires adapted to age, of which there are several tests. For example, there are receptive tests, expressive tests, vocabulary tests and comprehensive language assessments. The questionnaires can either be answered by proxies, e.g., a parent, or the child. For very young children (<3 years) only proxy questionnaires exist. There is no typical gold standard of test. These tests are designed to capture different domains of language, e.g., receptive, and functional language.

Receptive language is the understanding of speech. An example of a test is the British Picture Vocabulary Scale (BPVS). The BPVS is performed by a test leader speaking a word and the child pointing on a picture representing the meaning of that word (Dunn et al., 1997). Expressive language is the "output" of language. One test measuring expressive language is the Mullen Scale of Early Learning (MSEL). Functional spoken language could be described as communication using functional hearing and communication skills in relation to day to day situations as in teaching, in working life or in other social situations and includes both auditive receptive and expressive language (Hjelmervik et al., 2009; Wie, 2011).

4.4 Cost analysis

A cost analysis in economic evaluations often follows the three steps of identifying, quantifying and valuing resources as described in section 4.4.2 below. The term cost is often used when referring to expenditures, meaning the amount of money that are spent on a product or a service (Drummond et al., 2015). Costs, however, may also be opportunity costs, which may be defined as the value of the next best alternative that is foregone when another alternative is chosen. An example is a caregiver's use of time when caring for an ill person. The next best use of the time could be paid work, and the opportunity cost is the lost benefit of receiving wages.

Which costs to collect in an economic analysis will depend on the perspective taken and various other factors. First, an analyst should consider whether the analysis will estimate the total costs related to each intervention compared or whether the analysis will measure only the incremental costs differences, i.e., excluding routine costs related to both the comparator and intervention.

Second, the analyst should consider how detailed the cost analysis should be. In the hospital, the level of precision can vary from an average cost per patient to micro-costing where each component of resource use is estimated. Sometimes the options will be limited by the availability of data (Drummond et al., 2015). A more detailed description of precision levels is given in Table 4.1 below.

Table 4.1 Levels of precision in costing for hospital costs

Levels of precision in costing for hospital costs		
Micro-costing	Each component of resource use (e.g. laboratory tests, days of stay by ward, drugs) is estimated and a unit cost derived for each	Most precise ↑
Case-mix group	Gives the cost for each category of case or hospital patient. Takes account of length of stay. Precision depends on the level of detail in specifying the types of cases	
Disease-specific per diem	Gives the average daily cost for treatments in each disease category. These may still be quite broad (e.g., orthopaedic surgery)	
Average per diem (or daily cost)	Averages the per diem over all categories of patient. Available in most health care systems	Least precise

Adapted from Drummond 2015.

Third, the analyst needs to consider whether costs at all can impact the result. Small costs may not be worth to include if the effort to collect them is high and they are assumed not to have any impact on the outcomes.

4.4.1 Top-down and bottom-up cost estimation

Costing can be categorized as bottom-up and top-down processes (Chapko et al., 2009). In a top-down process, total costs are allocated downwards (by use of formulae) to a patient group or disease, for example from an hospital department to a patient's treatment. The bottom-up approach is more detailed and based on resource use at disease or patient level. All costs for procedures, laboratory services, medications, housekeeping etc are summed to give the aggregated total costs. The two methods may give different costs estimates for a single project (Cunnama et al., 2016). Previous studies comparing both methods have found the bottom-up method to be most accurate (Hendriks et al., 2014), but this is also debated (Cunnama et al., 2016). A draw-back with the bottom-up process is that it is time-consuming. And as a process become very detailed, costs may also be overlooked or are not even available. A bottom-up process for economic evaluations in healthcare is described below.

4.4.2 The costing-process

Drummond et al. (2015) present costing as a 3-step process of: Identification, Quantification and Valuation.

4.4.2.1 Identification and Quantification

The aim of the identification step is to identify the type of resources used in the management of a specific health problem. When the types of resources are identified, quantification can be started by finding information about the amount of each resource type. Some knowledge about the condition or disease and how it is treated is essential to both these steps. Different approaches can be taken to identify and quantify costs. One approach is by the help of experts in the field.

4.4.2.1.1 Expert opinion

General information about procedures and resource use may not always be present in the literature or easily available elsewhere. Experts in the field of interest could then be consulted. These experts could be health care personnel involved in the treatment of patients or other personnel following up patients outside the health care sector. Experts can also possess more detailed information about the actual resources used at a specific site or hospital than the information found in other sources. Expert-advice is used together with other sources of information.

4.4.2.2 Valuation

In the valuation step, prices or values are assigned to the resource use. In principle, valuation should be based on the opportunity cost method, preferably through market prices. In practice, resource use is valued from a variety of sources including previous CEAs, national registries, DRG price lists, NoMA's national unit database and in literature. The valuation of informal care and production loss may need some further description as follows.

One method of valuing informal care is by the opportunity costs of time (Hoefman et al., 2013; Koopmanschap et al., 2008). The opportunity cost of time represents the alternative use of the caregiver's or patient's time and is often valued by the average wage that could have been earned per hour. As wage varies widely between people, a common rate is often used. In Norway, this is the average hourly wage for all sector and occupations and the data are published by Statistics Norway. Several other methods for valuing informal care, such as the proxy good method, wellbeing method or the contingent valuation method are described by Hoefman et al. (2013) and Koopmanschap et al. (2008).

The two main methods of calculating production loss are the human-capital and the friction-cost methods. The human capital approach assumes no unemployment and that all hours not worked are considered lost production. The valuation of the production loss is done by multiplying the average wage rate, inclusive payroll tax and social costs, with the unemployment rate. The friction method only counts those hours lost until another employee overtakes the work. The friction period is the time it takes to restore the initial production level. Results from these two methods will almost always differ. The human capital method has been criticized for overestimating costs, while the friction-cost method for underestimating costs (van den Hout, 2010).

4.5 Analytical perspectives of economic evaluations

Economic evaluation can take on different perspectives depending on which costs and effects are included in the analysis. The most common are the health care perspective and the societal perspective. The health care perspective may be divided into direct and extended health care perspectives. The direct health care includes only costs directly related to the health care sector, such as costs of equipment and personnel in an hospital and drug costs. The extended health care perspective additionally includes non-medical costs, such as travel, and patient's or caregivers use of time related to appointments in the health care sector. The societal perspective includes all effects and costs that can be affected by a health intervention regardless of who pays the costs or who the health improvement affects. Examples are production loss and social services (Sanders et al., 2016). The three perspectives described above, and examples of costs associated with each perspective are given in Table 4.2.

Table 4.2 Analytical perspectives and examples of associated costs

	Perspectives		
	Direct health care	**Extended health care**[1]	**Societal**
Associated costs	Drugs, equipment, laboratory tests, health care personnel, facilities	Direct health care costs plus transportation costs, costs for hotel stays, costs of informal care (time) to attend health-related appointments	Productivity and production loss, other non-health sectors, e.g., impact on educational achievements, social services

Which analytic perspective is relevant, depends on the decision in question, but the issue has been controversial in health economics for decades. Arguments for both including and excluding societal costs are brought up in current discussions (Melberg, 2023). One argument against the societal perspective with inclusion of production gains and losses is that interventions targeting the elderly will be less cost-effective than those targeting employed people. This may lead to discrimination or favouring of certain groups. On the other hand, excluding factors considered only in the societal perspective may not give the full picture of the costs and gains of an intervention.

The guidelines from the Washington panel task force II recommends having two reference cases of the societal and health care perspectives (Sanders et al., 2016). However, the requirements by submissions of HTAs differ between countries. In Norway, the extended health care perspective, but not the societal perspective, is required by submission of single technology assessment (STAs) of medical devices and diagnostic interventions (NiPH, 2021).

[1] Caregiver's and patient's time costs were categorized as non-medical costs together with transportation and hotel costs in the extended health care perspective. The categorization is in line with the impact inventory from Washington panel II (Sanders et al. 2016), and with the NoMA's guidelines for HTA submissions. In the non-health sector, i.e., education sector, time costs were included as societal costs.

Even if the societal perspective is recommended in economic evaluations, many studies do not include this perspective. Further, studies claiming taking the societal perspective often miss relevant costs and outcomes. It is not always clear which costs and outcomes to include, and different objectives may give different judgements on what to add (Walker et al., 2019). Walker et al. (2019) suggests an extension to the "impact inventory" from Second Panel on Cost-Effectiveness in Health and Medicine (Sanders et al., 2016) to make a tool better adapted for inclusion of the societal perspective. The extended impact inventory is set up with outcomes or *dimensions of interest* to better inform decision makers across sectors and from different parts of a health care system. Objectives and consequences of an intervention to an individual may vary depending on value judgements and institutional settings. The dimensions should include outcomes and consequences of importance to decision makers in a specific setting or from their *dimension of interest*. By using this inventory, the analyst is forced to think through what outcomes to include, and on which sectors the outcomes and costs fall. Each *dimension* includes both direct costs and opportunity costs.

4.6 Estimating outcomes of economic evaluation

4.6.1 The expected value

An expected value is a weighted mean value calculated by the sum of different means multiplied with the probability of its occurrence and can be written as:

$$E[X] = \Sigma\, xi * p$$

E[X]: Expected value, xi: unit value, p: probability.

This value is used when presenting average results on group or individual level in models.

4.6.2 Incremental Cost-Effectiveness Ratio (ICER)

The relationship between costs and consequences in a CEA is usually presented as the incremental costs between the alternative(s) studied divided by the incremental effect. This is referred to as the ICER (incremental cost-effectiveness ratio) and gives information of the extra cost per extra unit of health effect. If ICER < Willingness-to-pay (WTP), this indicates cost-effectiveness of an intervention.

$$ICER = (Cost\ A - Cost\ B)/(Effect\ A - Effect\ B) = (\Delta\, Cost)/(\Delta\, Effect)$$

4.6.3 Net Monetary Benefit or Net Health Benefit

Another common way to present the result is by the Net Monetary Benefit (NMB) or Net Health Benefit (NHB). The WTP is included in the formula, denoted as lambda (λ):

$$\text{Net monetary benefit (NMB)} = \lambda * \Delta E - \Delta C$$

$$\text{Net health benefit (NHB)} = \Delta E - \Delta C/\lambda$$

λ: WTP threshold, ΔE: incremental effectiveness, ΔC: incremental cost

If NMB or NHB is positive, the intervention is cost-effective. If several strategies are present, the strategy with the highest NMB or NHB is considered the most cost effective.

4.6.4 Discounting

Discounting is a process of converting future costs and benefits into present values (PV). Decision models often simulates costs and outcomes over several years and costs and effects in different time periods are commonly discounted. Discounting relates to the opportunity costs of investing money now rather than in the future. In health economic evaluations the health gains (QALYs) are usually discounted with the same rate as the costs (Solberg et al., 2020).

The real interest rate (interest rate adjusted for inflation) could be used as an estimate of the discount rate (Turner et al., 2019). Because the uncertainty of the interest rate is higher in far future, the discount rate is lowered after certain years. In Norway, the discount rates for economic analyses of public interventions is, according to guidelines from the Ministry of Finance, set to 4% the first 40 years following an intervention, thereafter 3% until 75 years and 2% after 75 years (Finansdepartementet, 2021).

A formula for the present value for a single year is:

$$Present\ value(PV) = \ Future\ value\ (FV)/(1+i)t$$

where *i* refers to the interest rate and *t* to the cycle.

4.7 Decision-analytic modelling

The need to calculate the health and economic outcomes necessitates modelling, because clinical trials cannot capture all the relevant consequences. Decision-analytic modelling makes it possible to combine and analyse data from different kind of sources. Models can project future costs and effects, extrapolate results beyond the duration of a clinical trial and assess cost-effectiveness in heterogenic subgroups. Modelling can be used to replace or complement evidence from clinical trials. Models are a simplification of reality, but the aim is to reflect real numbers in the best possible way. One commonly used type of model in CEAs is the Markov-model.

4.7.1 The Markov-model

The Markov-model is suitable when assessing costs and effects over a long time-horizon and/or when events are re-occurring over time (Kuntz et al., 2016). It consists of mutually exclusive health states with the possibility of patients transitioning between states at discrete time intervals (cycles) (Briggs et al., 2006). The transfer to another state is conditioned on the previous state and transition probabilities are included in the model to calculate the proportions (or individuals) going from one state to another. Costs and effects are attributed to each health state and outcomes are calculated based on the time spent in the health states multiplied with the costs and effects associated with those health states.

Markov models are memoryless in the sense that transition probabilities, costs and outcomes are not influenced by what happens earlier in the model. Transition probabilities may be constant across cycles (so-called Markov chain model) or vary by time in the model (Briggs et al., 2006). Constant transition probabilities are unrealistic in a medical context but make the modelling easier to perform. Technically, transition probabilities may vary by time, but valid data may be scarce.

4.7.1.1 Discontinuity correction

In Markov models, costs and effects are set to occur either in the start or in the end of a cycle. In real life these costs and effects can occur anytime during the cycle. Half-cycle correction is a common method used to average the timing of events or costs incurring within the cycle.

If for example, the costs are modelled to occur in the start of the cycle, the costs in the rest of that cycle are not counted until the beginning of the next. Half-cycle correction adjusts for this and accounts for the fact that, in reality, the transitions on average happens in the middle of a cycle (Naimark et al., 2008). It is calculated by adding costs or effects for two subsequent cycles and dividing by 2. Initial costs are mostly not half-cycle corrected as they are incurred at start of the model.

$$Average\ costs\ in\ current\ cycle\ = (Costs\ in\ previous\ cycle + Costs\ in\ current\ cycle)/2$$

4.8 Uncertainty

All outcomes of economic evaluations are accompanied with uncertainty. Two of the main types of uncertainty to be considered in modelling are structural uncertainty and parameter uncertainty.

4.8.1 Structural uncertainty

Structural uncertainty relates to the choice of model type and how the model is set up, i.e., the conceptualizing of the model. A model needs to reflect the nature of a condition or disease. In general, several types of models can be used for the same research question. Models have different characteristics, and a specific model type may be more suitable than another for the problem we want to study. When choosing type of model we need to consider - among other things - if we want to study individuals or groups, what time horizon to have and if events or transitions happen more than once (Roberts et al., 2012). The assumptions we make, when setting the cycle length in a Markov-model and choosing which parameters to include in the model etc., should reflect reality. Which tests or instruments we base the input parameters on also matters. If the model is not appropriate for the question studied, the outcomes can be misleading.

4.8.2 Parameter uncertainty

Parameter uncertainty is present because most inputs used in a model are only sample estimates of the true values. Data from the whole population in question are hardly ever available. Input data for costs and effects are taken from individual samples or sample populations representing the individuals or the group studied. The degree of uncertainty will be affected by the amount and quality of the data used in the model.

The potential impact of uncertainty should be explored in sensitivity analyses. By changing the inputs in the model, we observe how uncertainty may impact the outcomes and if it affects the decision.

4.9 Sensitivity analyses

Three common sensitivity analyses to account for uncertainty are structural, deterministic, and probabilistic sensitivity analyses and are described below.

4.9.1 Structural sensitivity analysis

There are different ways to classify types of uncertainty, but sources of uncertainty including simplifications and scientific judgments made when constructing a model could be classified as structural uncertainty (Bojke et al., 2009). A part of a structural sensitivity analysis can for example be performed by changing the test the transition probabilities are based on and run the model with each of the tests. By comparing the outcomes, it can be explored how sensitive the results are to the change in input data.

4.9.2 Deterministic sensitivity analysis

In deterministic analyses, outcomes are estimated based on the point estimates of the parameters in the model. The point estimates are the means or the expected values, and the outcomes are calculated directly from these. To account for uncertainty, we can vary the value of one or more of the input parameters at a time, to see how the results are affected. Typically, each parameter value is varied 25% up and down from expected value (one way sensitivity analysis). More than one parameter value may be changed at a time (multiway sensitivity analyses) (Drummond et al., 2015).

4.9.3 Probabilistic analysis

In probabilistic analysis, probability distributions are assigned to the probabilities, costs, and utilities (Aas, 2020). By doing this, the point estimates are replaced by a distribution and the parameters can take on a range of possible values. The standard errors for the distributions are either collected from the literature or estimated. The model is set to make random draws from the distributions by simulations. The draws from each distribution are repeated many times and each draw give a different number for the parameter. The outcome resulting from one draw may therefore differ from another. One of the alternatives may be estimated to be the most cost effective based one draw, but the other alternative in another draw. Probabilistic analyses can account for uncertainty in all relevant parameters at the same time, i.e., joint uncertainty. The mean probabilistic outcome values are given as the average values of all the repeated draws.

Probabilistic analyses make it possible to calculate the probability of an intervention being cost effective at given WTP thresholds. Calculating the proportion of the results under a certain threshold give the probability of being cost-effective at that threshold. Similarly, the results can be used to calculate the probability of cost-effectiveness at several different thresholds using the net monetary benefit (NMB).

While probabilistic sensitivity analyses capture uncertainty in many parameters at the time, it also accounts for nonlinearity of a Markov model. The outcomes are based on addition of several inputs multiplicated with their probabilities and these mathematical equations are inherently nonlinear. Because of this nonlinearity, the calculations of the outcome values directly from the expected values of the input parameters may have bias: the expectation of the means is not the mean of the expectations. By making the model probabilistic, it will yield the expected values and the joint uncertainty distributions for the outcomes (Briggs et al., 2006). The probabilistic outcome values may not differ much from the deterministic values.

4.10 Transparency and validation

The International Society for Pharmacoeconomics and Outcomes Research (ISPOR) and the Society for Medical Decision Making (SMDM) task force have published recommendations for how to present a model to achieve transparency and for how to perform model validation (Eddy et al., 2012).

Model transparency refers to the fact that a model should be understandable by readers with different types of expertise. Both a non-technical description of the model and more detailed technical information should be given. The technical information should be detailed enough for someone with expertise in modelling to check and it should be sufficient for possibly replicating the model.

Validation are methods used to test the model’s accuracy and if the problem analysed reflects real life or clinical practice. There are different types of validation including face validity and internal validity, which both are relevant for our analysis.

Face validity means to check whether the model correspond to current science and evidence, and to reality. Face validity should be done by people with expertise in the field of question. Important aspects are the model structure, data sources, problem formulation and results. Internal validity includes checking calculations and equations in the model for mistakes and accuracy.

5 Methods

5.1 Analytic overview

We used a model-based approach to follow a cohort of Norwegian children receiving bilateral CI before 12 months of age. The analysis was performed assuming a homogeneous population with no other disabilities and an IQ score above 75 measured by Raven scale (Raven, 2004). For estimation of the total lifetime costs and health outcomes associated with current cochlear implantation practice, we included resource use common for all individuals independent of language skills as well as resource use and HRQoL stratified by three achieved health states of language skills.

We selected three different analytic perspectives depending on who incurred the costs and who obtained the effects: 1) the health care, 2) the extended health care and 3) the societal perspective. Resource use from the health care sector, the education sector and for production loss was included. The analyses involved a primary descriptive analysis to map the health and economic consequences of the standard of care over the lifetime of bilateral CI recipients. For estimating the potential impact of AVT as an intervention after CI, we conducted an exploratory cost-effectiveness analysis of AVT compared to standard of care. Probabilistic analyses were performed for parameter uncertainty. Sensitivity analyses were performed by changing the type of HRQoL-questionnaire the input parameters were based on.

The outcomes were discounted lifetime costs (2021 Norwegian Krone (NOK)) and QALYs[2]. Results from the exploratory analysis with AVT are given as ICERs and compared with two chosen willingness-to-pay thresholds of NOK 300 000 and NOK 500 000 to evaluate cost-effectiveness. Costs and effects were discounted according to the Norwegian guidelines for submissions of single technology assessments (NiPH, 2021) with a discount rate of 4% the first 39 years, 3% from 50-75 years and 2% from 76 years after intervention. Excel was used as technical tool for all modelling.

5.2 Model description

We developed a Markov cohort model as the framework for a CEA. Following CI at <12 months, a cohort of individuals enter a 4-state Markov model at age 3. Following consultations with experts at Rikshospitalet, Oslo University Hospital, and the department of special needs education at University of Oslo we identified 3 functional language health states ("Very low-", "Low" and "Normal+") (Figure 5.1). We added dead as an additional health state to reflect the impact of background mortality. Each year, individuals could move between all the language states during each cycle until 18 years of age after which they remained in the health state they had at that age., Costs and QALYs accrued in each cycle all through life. At the end of the lifelong model all individuals reached the absorbing state of death.

[2] QALY weights were based on PedSQL and KINDL

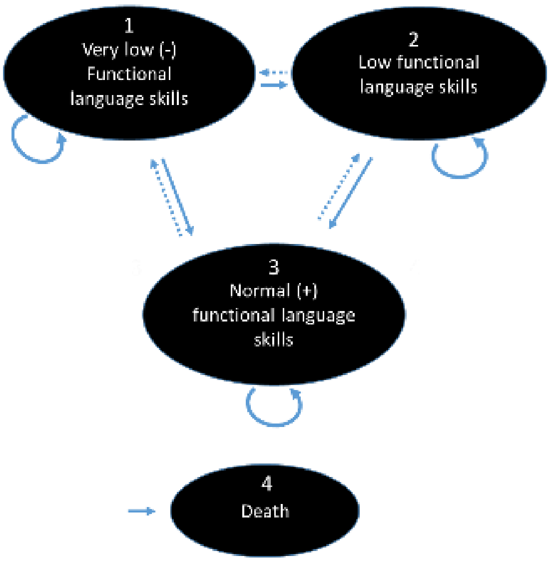

Figure 5.1 Markov model with 3 health states related to language skills and a state of death

An annual cycle length was selected as the children's development of language skills is gradual. Although the development is faster the first years of life, the cycle length was set to be the same for the whole time-horizon due to simplicity. Adverse events as infections or tinnitus after operation were considered mild and seldom and were not included in the analysis (L.R. Opheim, personal communication, 20-Aug-2021).

Deterministic and probabilistic analyses were performed for both undiscounted and discounted outcomes. In the probabilistic analysis, the number of simulations were set to 1000. The probabilistic value was calculated as the average value from the 1000 simulations and 90% credible bounds were given for the values. The gamma distribution was used for costs, the beta distribution for HRQoL and the Dirichlet distribution for the transition probabilities.

5.3 Model inputs and sources

We relied on data from two Norwegian studies (one cross-sectional and one longitudinal study) to inform our health state definitions, as well as the age- and state-specific transition probabilities and health-related quality of life. For resource use, we relied on a combination of expert opinion, national guidelines, and national tariffs.

5.3.1 Cross-sectional study - The 500-project

The national study titled "Speech Perception, Language, and Quality of Life in People Who Have Received CIs as Children in Norway ", included the first children receiving cochlear implantation in Norway. These children had received CI between 1998 and 2015. The study was cross-sectional and captured data about language, cognition, and hearing as well as HRQoL. A total of 496 participants were included, and data collections were performed between 2013 and 2016. Due to heterogeneity of the participants, a sample of 127 of the 496 children were considered eligible for our analysis. This sample was used to define the health states by scores achieved in language tests. Data from this project also informed the HRQoL associated with the language skills (the health states). Because the study involved 500 children, it is subsequently referenced as the *500-project*.

5.3.2 Definition of health states

We defined language skills as Normal or higher ("Normal+"), Low and Very Low or worse ("Very low-) based on scores achieved in language tests in the cross-sectional study (500-project). Individuals with a score of no more than 1 SD below the mean were considered Normal+, individuals with a score of 1-2 SDs below the mean were considered Low, and individuals with a score of more than 2 SDs below the mean were considered Very Low-. Several instruments had been used to test different types of language skills and varied with age. We based the health states on the results from the BPVS-instrument. An expert from OUH with access to raw data grouped the children by the scores. Table 5.1.

Table 5.1 Categorization of scores of alternative language tests according to model health state

Model health states	MSEL Mean 50, SD = 10	CELF-4 Mean = 100, SD = 15	CCC-2 Mean = 100, SD = 15	**BPVS Mean = 100, SD = 15**	**Source**
Normal+ (No more than 1 SD below the mean)	≥ 40	≥ 85	≥ 85	**≥ 85**	500-project
Low (1-2 SD below the mean	< 30-39	< 70-84	< 70-84	**< 70-84**	
Very Low – (More than 2 SD below the mean)	< 30	< 70	< 70	**< 70**	

Abbreviations: **MSEL** = Mullen Scale of Early Learning, **CELF-4** = Clinical Evaluation of Language Fundamentals, **CCC-2:** Child Communication Checklist, **BPVS** = British Picture Vocabulary Scale

The health states were also described with words to make it easier to understand what was meant with being in each of the health states. The translation was performed by another expert at University of Oslo who translated the scores from the BPVS language test using standard deviations into scores as percentiles. Table 5.2.

Table 5.2 Verbal description of health states by percentile range

Percentile range	**Verbal description of health states translated from scores on BPVS**
Percentile range >= 16	"Normal+"; the child performed as well as or higher than approximately 16% of same-age children in the test's normative sample.
Percentile range < 16	"Low"; the child performed lower than approximately 16% of same-age children in the test's normative sample.
Percentile range < 2	"Very Low-"; the child performed lower than approximately 2% of same-age children in the test's normative sample.

Source: V. Diamanti, personal communication, 13. Jan 2023.

5.3.3 The longitudinal 6-years follow-up study

Transition probabilities were derived from a longitudinal Norwegian study by Wie et al. (2020). This study included the first 21 children who received the combination of early and simultaneous implantation in Norway. The group was compared to 21 children with normal hearing matched on age, gender, and maternal education. Both groups performed a variety of language tests at 10 different time points during a 6 years' follow-up period.

5.3.4 Transitions probabilities

Conditional transition probabilities were calculated based on scores on the BPVS for 15 children ages 3 to 6 years from the longitudinal data. Scores from other language tests as MSEL and MCDI were available, but these had only been performed until 4 years after implantation. Due to lack of data for BPVS after 6 years, we held the transition probabilities constant until 17 years of age. From 18 years, individuals remained in their current health state until death. Together with clinician experts, this assumption was justified as no great changes in language skills are expected due to the gradually declining plasticity of the brain when getting older (T. Busch, personal communication, 03-May-2022). Transition probabilities through all ages are summarized in Table 5.3.

Table 5.3 Transition probabilities based on BPVS in the longitudinal 6 years follow-up study

Health state transitions	Age 3 to 4	Age 4 to 5	Age 5 to 6	Age 6 to 17*	Age 18 to 106**	Distribution	Source
Normal to Normal	0,88	0,64	0,63	0,63	1,00	Dirichlet	Wie et al. (2020)
Normal to Low	0,13	0,27	0,25	0,25	0,00	Dirichlet	Wie et al. (2020)
Normal to Very Low	0,00	0,09	0,13	0,13	0,00	Dirichlet	Wie et al. (2020)
Low to Low	0,29	0,33	0,25	0,25	1,00	Dirichlet	Wie et al. (2020)
Low to Normal	0,57	0,33	0,25	0,25	0,00	Dirichlet	Wie et al. (2020)
Low to Very Low	0,14	0,33	0,50	0,50	0,00	Dirichlet	Wie et al. (2020)
Very Low to Very Low	0,00	1,00	0,33	0,33	1,00	Dirichlet	Wie et al. (2020)
Very Low to Low	0,00	0,00	0,67	0,67	0,00	Dirichlet	Wie et al. (2020)
Very Low to Normal	0,00	0,00	0,00	0,00	0,00	Dirichlet	Wie et al. (2020)

*Transition probabilities set to the same as for age 5 to 6 due to lacking data. **Transition probabilities set to be stable by assumption

5.3.5 Health related quality of life

HRQoL-data was available from the same study (the 500-project), as was used to define the health states. By using the same data, scores from language tests used to define the health state and HRQoL could be linked. The average scores for HRQoL were calculated, and each health state (language skills) was assigned a level of HRQoL. Available HRQoL-data from this dataset were mainly from study participants up to 18 years of age. Because of limited data, the HRQoL from 19 years were set to be the same as for those of 18 years and below. The HRQoL-questionnaires used in the 500-project were KINDL, Pediatric Quality of Life Inventory (PedSQL) and Satisfaction with Life Scale (SWLS). Of the three questionnaires, KINDL and PedSQL were used for our analysis.

Several different types of language tests were performed in the 500-project (Table 5.1) and there were available data for HRQoL associated with the health states based on these different types of language tests. The HRQoL associated with each health state varied somewhat with the language tests used when defining the health state. The HRQoL by state also differed between parent reported and self-reported tests. In the base case analysis, we used the scores from the PedSQL when the health states had been based on scores in the BPVS language test. Further, the parent reported versions were used. HRQoL associated with each of the 3 health states are listed in Table 5.4. HRQoL was set to be the same for each state during the whole time-horizon.

Table 5.4 Health-related quality of life by health state based on PedSQL

Health state	PedSQL/BPVS	SE	Distribution	Source
Normal+	86.4	0.021	Beta	500-project
Low	81.5	0.024	Beta	500-project
Very Low-	74.7	0.022	Beta	500-project

Abbreviations: Pediatric Quality of Life Inventory, SE: standard error

5.3.6 Age-related adjustment of utility

QALY weights (health state utility values) for the general population were included to adjust the QALYs related to the health states by age-related utility. We used inputs consistent with the Norwegian guidelines (NiPH, 2021) to reflect quality of life by age. Research has shown that quality of life decreases with age, independently of any condition or disease.

5.3.7 Mortality

The risk of death due to re-implantation, or health-state specific mortality was considered zero; therefore, individuals were at risk of death due to background causes only. We used population mortality data from Statistics Norway (2021). The proportion of the population transitioning to death at each cycle was calculating by multiplying the proportion of the population alive in each cycle by the probability of death in each cycle.

5.3.8 Costs

We identified, quantified, and valued costs according to best practice (Drummond et al., 2015) and stratified by health care sector, education sector and production loss to enable analyses from different perspectives. Both costs common for all patients and health state specific costs were included. Resource use in the specialist health care sector was discussed and collected in meetings with experts at Rikshospitalet OUH and from follow-up correspondence from these meetings. Resource use for support and follow-up in kindergarten and school were discussed in a meeting with the Public special education service (Statped) and in correspondence with the Education and psychological counselling service (PPT), who both are involved in the supportive care of these children. Production loss, included as societal costs, was discussed in a meeting with the expert team. Data on production loss for the group studied are not currently available and had to be based on assumptions. However, some support from the discussion with the expert team was taken when assuming values.

Unit costs in 2021 NOK were based on the national primary care reimbursement tariffs (Normaltariffen) and the unit cost database of the Norwegian Medicines Agency, The Norwegian Pharmaceutical Product Compendium (Felleskatalogen) and DRG-lists for somatic hospitals. The costs of time for different types of involved personnel were valued using the average monthly wages published by Statistics Norway (Statistics Norway, 2021).

Standard errors for all costs were set to 10% of the mean value by assumption. The gamma distribution was used in the probabilistic analysis.

5.3.9 Costs in Health care sector

Costs from the health care sector were equal across all health states. These costs were divided into direct medical costs and non-medical costs. Direct medical health care costs were directly related to the health care system such as assessments in hospital, surgical operations, CI sound onset, other initial CI costs and running costs of the specialist health care sector (Rikshospitalet OUH). Non-medical costs included transportation costs, use of caregivers or patient's time and costs of hotel stays in connection with care in the specialist health care sector. Direct medical and non-medical costs and their sources are presented in Table 5.5 and Table 5.6. Costs were counted from 0 years, to capture the costs including assessments and operation between ages 0 to 3 years.

Table 5.5 Direct medical costs in the specialist health care sector (NOK 2021)

Type of cost	**Cost**	**SE***	**Distribution**	**Description**	**Sources**
Initial and first year costs: Assessments, CI operation, routine controls etc. (age 0)	251 076	25 108	Gamma	Assessment at local institutions Assessment at Oslo University Hospital (OUH) Surgery, device and sound onset Routine controls at 3, 6, 9 and 12 months Parent's out-of-pocket costs for batteries, cables, and other accessories	Expert advice OUH, Unit database NoMA, Norwegian Pharmaceutical Compendium, Normaltariffen, DRG-list, code 803U Statistics Norway: Tables 11418, 11419, 12897 HELFO and Out-patient regulations
Assistive Listening Devices (age 1-18 years)	10 000	1000	Gamma	Provision of assistive listening devices (remote microphones, FM systems, telecoil, Bluetooth streaming dongle, neck loop etc.) at start in kindergarten, primary and secondary school, and high school	Expert advice OUH Assumption
Routine controls (age 1)	5 141	514	Gamma	Two routine controls 2nd year by physicist and pedagogue (18 and 24 months)	Expert advice OUH Statistics Norway: Table 11418
Hospital Case Manager Annual administration (age 1 -106)	500	50	Gamma	Annual administration costs from 2nd year: Setting up appointments etc.	Assumption
Routine controls (age 2-18)	2 571	257	Gamma	Annual controls from Year 3 until 18 years by physicist and pedagogue	Expert advice OUH Statistics Norway: Table 11418
Technical accessories (age 1-106)	1 000	100	Gamma	Parent's (or the patient's from age 18) yearly out-of-pocket costs for batteries, cables, and other accessories	Expert advice OUH Assumption
Processor upgrades every 6 years (age 6- 106)	96 250	963	Gamma	Processor upgrade (every 6th year from Year 6)	DRG-list, code 49C
Re-implantation (age 40)	224 937	22 494	Gamma	Once during lifetime (at 40 years)	Expert advice OUH

*Standard errors were not available and was set by assumption to 10% of the mean value. **Abbreviations**: **DRG:** Diagnosis Related Groups, **HELFO:** The Norwegian Health Economics Administration, **OUH:** Oslo University Hospital.

Table 5.6 Non-medical costs: Time, travel and hotel costs ages 0 -106

Type of cost	Cost	SE*	Distribution	Description	Source
Initial and 1st year costs for caregivers and patient (travel, time and hotel) (age 0)	64 833	6483	Gamma	Travel related to assessment OUS, implantation, fitting of hearing aid, sound onset and routine controls (28 return trips) Use of time related to assessment OUS, implantation, fitting of HA, sound onset and routine controls. 3 nights in hotel in connection with surgery (1 night) and sound onset (2 nights)	Expert advice OUH Statistics Norway, Table 11419 and Table 12897
Caregiver's and patient's travel to routine controls (age 1)	5808	581	Gamma	2 return trips for one caregiver + child	NoMA unit database
Caregiver's time routine controls (age 1)	3 810	381	Gamma	Use of time for one caregiver - Two routine controls	Statistics Norway: Table 11418
Caregiver's and patient's travel to routine controls (age 2-18)	2904	290	Gamma	1 return trip for one caregiver + child	NoMA unit database
Caregiver's time routine controls (age 2-18)	3810	381	Gamma	Use of time for one caregiver - 2 routine controls	Statistics Norway: Table 11418
Travel by processor upgrade (age 6-18)	2904	290	Gamma	Travel to OUH by processor upgrade - One caregiver + child - 1 return trip every 6th year	NoMA unit database
Caregiver's time processor upgrade (age 6-18)	1905	191	Gamma	Use of time for one caregiver in connection with processor upgrade every 6th year at OUH	Statistics Norway: Table 11418
Patient's travel by processor upgrade (age 19-106)	1452	145	Gamma	Travel patient to OUH by processor upgrade - 1 return trip every 6th year	NoMA unit database
Patient's time by processor upgrade (age 19-106)	1905	191	Gamma	Use of time for patient in connection with processor upgrade every 6th year at OUH	Statistics Norway: Table 11418
1-time patient travel for re-implantation (age 40)	1452	145	Gamma	Patient's travel by re-implantation – once in life	NoMA unit database
Patient's time re-implantation (age 40)	3810	381	Gamma	Patient's use of time by re-implantation – once in life	Statistics Norway: Table 11418
Hotel re-implantation (age 40)	865	87	Gamma	Hotel stay one night – once in life	Statistics Norway: Table 12897

*Standard errors were not available and was set by assumption to 10% of the mean value.

5.3.10 Societal costs

Societal costs included in the model were costs in education sector and production loss due to unemployment. These costs were assumed to be health state specific.

Health-state specific costs in the education sector included costs of municipality level in kindergarten and school. As this was a new field of costing analysis, there were time concerns getting a complete overview of costs related to supportive care and the association to the different health states, and these costs are based on assumptions. Even if we were not able to get an overview of the costs, communication with Statped and PPT implied assistance in making assumptions. The costing of these services covered the period from kindergarten at age 1 to high school at age 18. We assumed that resource use was highest for children in health state Very Low-, somewhat lower in health state Low and lowest in health state Normal+.

The costs in education sector reflect supportive care in kindergarten and school facilitated by the Public special education service (Statped, 2022) and the Education and psychological counselling service (Utdanningsdirektoratet, 2022). Table 5.7.

Caregivers use of time was also added to the costs in this sector. Travel was not added as most activities were considered to take place close to their home. Table 5.8.

Table 5.7 Costs of the education sector by age and health state

Type of cost	Normal	Low	Very Low	Description	Source
Kindergarten					
Age 1 Supportive care	51 759	51 759	90 590	First observation of child Administrative work after first observation First guidance meeting with personnel Facilitation for listening and groups, Contact with and guidance to caregiver(s) Physical follow-up meetings Digital follow-up meetings Social worker assistant Speech therapist	Assumption Statped*
Age 2 Supportive care	27 223	46 638	66 054	Contact with and guidance to caregiver Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped
Ages 3-5 Supportive care	13 612	28 907	44 202	Contact with and guidance to caregiver Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped

Primary school					
Age 6 Supportive care	32 363	51 778	71 194	First observation of child in school Administrative work after first observation Meeting with teacher and parents after first observation Facilitation for listening, group rooms and playgroups Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped
Age 7-12 Supportive care	18 501	37 916	57 331	Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped
Secondary school					
Age 13 Supportive care	20 497	39 912	59 328	Facilitation Meeting with teacher and parents Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped
Age 14-15 Supportive care	18 501	37 916	57 331	Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped
High school					
Age 16 Supportive care	20 534	39 949	59 364	Facilitation Meeting with teacher and parents Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped
Age 17-18 Supportive care	18 501	37 916	57 331	Physical follow-up meetings Digital follow-up meetings Speech therapist Social worker assistant	Assumption Statped

*Statped: Public special education service

Table 5.8 Cost of caregivers' use of time related to education services

Type of cost	Normal+	Low	Very Low-	Description	Source
Kindergarten					
Age 1 Time Caregiver	13 220	13 220	13 220	1 hour/week guidance with Senior Advisor	Assumption
Age 2 Time Caregiver	12 204	2 204	12 204	2 hours/month guidance with Senior Advisor	Assumption
Age 3-5 Time Caregiver	3 051	6 102	9 153	1 hour/month Normal 2 hours/month Low 3 hours/month Very Low in connection with Speech therapist and Assistant	Assumption

Primary school					
Age 6 Time Caregiver	12 204	15 254	18 305	2 hours/month Normal 2,5 hour/month Low 3 hours/month Very Low	Assumption
Age 7-12 Time Caregiver	3 051	6 102	9 153	1 hour/month Normal 2 hours/month Low 3 hours/month Very Low	Assumption
Secondary school					
Age 13 Time Caregiver	12 204	15 254	18 305	2 hours/month Normal 2,5 hour/month Low 3 hours/month Very Low	Assumption
Age 14-15 Time Caregiver	3 051	6 102	9 153	1 hour/month Normal 2 hours/month Low, 3 hours/month Very Low	Assumption
High school					
Age 16 Time Caregiver	12 204	12 204	18 305	2 hours/month Normal 2,5 hour/month Low 3 hours/month Very Low	Assumption
Age 17-18 Time Caregiver	3 051	6 102	9 153	1 hour/month Normal 2 hours/month Low 3 hours/month Very Low	Assumption

We used the human capital method (Grossman, 2000) to estimate production loss. Limited data on unemployment are available for the population studied, and assumptions for the level of unemployment were done to explore these costs.

Published data for the average national wage rate and the proportions of the general population employed by age was used in the calculation of production loss (SSB, 2021). Average wage and proportions employed were multiplied with a factor of 1,38 to include payroll tax and social costs. We assumed that unemployment was higher among CI patients: 15% increased unemployment in health state Low and 25% in health state Very Low-. Table 5.9.

Table 5.9 Inputs for calculation of production loss (NOK 2021)

Description of inputs for calculation	Number	Source
Average monthly wage (NOK)	50 790	Statistics Norway, table 11418
Payroll and Social costs factor	1.38	NoMA's unit data base
Proportion employed 15-19 years (%)	38.6	Statistics Norway, table 06445
Proportion employed 20-24 years (%)	67.1	Statistics Norway, table 06445
Proportion employed 25-39 years (%)	81.1	Statistics Norway, table 06445
Proportion employed 40-54 years (%)	86.0	Statistics Norway, table 06445
Proportion employed 55-66 years (%)	67.9	Statistics Norway, table 06445
Proportion employed 67-74 years (%)	19.7	Statistics Norway, table 06445
Proportion unemployed in Low (%)	15.0	Assumption
Proportion unemployed in Very Low (%)	25.0	Assumption

5.4 Model validation and transparency

The structure of the model including the number and the definition of health states were decided in close collaboration with experts in the field of hearing loss. Resource use was collected from Rikshospitalet OUH from experts working closely with these patients and with high knowledge about the procedures. The setup of the model was presented at meetings during the process so that the expert team could comment or suggest adjustments. Further insight on how these patients are followed-up in the municipality was received in a meeting with Statped and from correspondence with both Statped and PPT.

A column checking that the proportions in each of the health states summed to 1 was added in the Model-sheet to decrease the probability for mistakes or wrongly written transition formulas. All transitions probabilities in the parameter-sheet were summed and checked to be equal to 1. All formulas in the Parameter and Model-sheet were looked through for any discrepancies or mistakes.

A detailed list with description of all costs is given in the appendices as well as calculations for some of the units. Details for the input parameters for HRQoL and transition probabilities and referrals to their sources are given in the methods chapter.

5.5 Exploratory analysis of Auditory Verbal Therapy (AVT)

Under base case assumptions we included the potential health and economics impacts of AVT in an exploratory analysis. We assumed AVT is given for one after implantation as the this is practiced in Norway today. Because the children entered the model at three years and after AVT had been given, the effect of the AVT was assigned after training completion. The proportion entering the model with language skills in the Normal+ state was assumed to be 11% higher than the group modelled without AVT and the proportion in Low 11% lower (Table 5.10). The effect of AVT was assumed to continue until age 17. Language skills obtained early in life have shown to be predictive of outcomes in early adulthood (Castellanos et al., 2014; Geers et al., 2011; Hunter et al., 2017), but the duration of the effect of AVT is unknown. A relative risk of 1.20 was assumed for transitions from state Low to Normal+ and from state Very Low- to Low and was applied from age 4 until age 17 (Table 5.11).

Table 5.10 Proportions at entry into model for base case analysis and exploratory analysis of AVT

	Proportions at entry – base case*	**Proportions at entry - Exploratory analysis of AVT****
Normal+	0.53	0.59
Low	0.47	0.41
Very Low-	0.00	0.00

*Based on longitudinal study by Wie et al (2020). **Based on assumptions.

Table 5.11 Relative risk of transitions in exploratory analysis of AVT

Ages 4-17	**Exploratory analysis of AVT – relative risk**	**Source**
Transition from Low to Normal+	1.20	Assumption
Transition from Very Low- to Low	1.20	Assumption

AVT may be provided direct in-person or digitally. We assumed direct in-person meeting between caregiver, child, and pedagogue. Travel and time costs were added (Table 5.12).

Table 5.12 Costs for Auditory Verbal Therapy by physical meetings the first year

Type of cost	Cost	SE	Distribution	Description	Source
Auditory Verbal Therapy (AVT)	18 988	1 899	Gamma	1 hour training with pedagogue + 1,5 hour's administration work, 18 times –first year	Expert advice OUH
Travel costs related to AVT	46 464	2 323	Gamma	Travel to AVT - first year – 32 return trips for one parent + child (NOK 726 one-way x 2 x 32)	Assumption NOMA unit database
Time costs for caregiver related to AVT	18 288	1 829	Gamma	Time used by AVT – first year - One caregiver - 18 half days (4 hours)	Assumption

5.6 Inputs for sensitivity analyses

For sensitivity analyses of HRQoL we replaced PedSQL data with KINDL data to explore how it affected the outcomes for the main analysis and the exploratory analysis of AVT. The inputs from KINDL by health state are shown in Table 5.13.

Table 5.13 HRQoL by health state based on KINDL

Health state	KINDL/BPVS Norwegian	SE	Distribution	Source
Normal+	79.3	0.014	Beta	500-project
Low	78.8	0.025	Beta	500-project
Very Low-	68.4	0.033	Beta	500-project

6 Results

6.1 Descriptive analysis of standard of care

From the societal perspective (including all the sectors and production loss), the total discounted expected lifetime cost per CI patient was NOK 2 375 698 (Table 6.1). When only costs in the health care sector were included, the expected discounted lifetime costs decreased with more than half to NOK 1 060 858. When we disaggregated the costs, we found that the expected costs in the education sector alone were NOK 581 683, of which NOK 112 722 were costs for caregiver's use of time and NOK 468 961 were wage costs for personnel giving supportive care in kindergarten and school. Production loss accounted for approximately 30% of the total costs and amounted to NOK 733 050. By dividing the costs in the health care sector into direct medical health care and non-medical costs for time, travel and hotel, costs were estimated at NOK 888 948 and NOK 172 016, respectively.

The total undiscounted costs from the broadest societal perspective amounted to NOK 5 347 950, while the total undiscounted costs in the health care sector made up about 40% of these costs with an estimated value of NOK 2 160 826. The total undiscounted costs in the education sector were NOK 732 867, 14% of total costs, and costs related to production loss made up approximately 46% of the total costs and amounted to NOK 2 454 256.

Table 6.1 Lifetime costs by sector and analytic perspective

	Undiscounted (90% credible bound)	**Discounted** (90% credible bound)
HEALTH CARE SECTOR (Specialist health care only)		
Direct medical specialist health care	**1 949 556** (1 784 828 - 2 114 114)	**888 948** (819 476 - 961 932)
Non-medical costs: Time, travel, and hotel	**211 270** (199 298 - 223 120)	**172 016** (161 487 - 82 873)
Total	**2 160 826** (1 992 003 - 2 324 883)	**1 060 964** (990 478 - 1 135 286)
EDUCATION SECTOR		
Accommodation and follow-up in kindergarten and school	**592 141** (504 659 - 687 275)	**468 961** (408 805 - 530 418)
Caregivers time	**140 727** (126 629 - 157 723)	**112 722** (103 082 - 123 201)
Total	**732 867** (631 908 - 843 469)	**581 683** (512 825 - 652 720)
PRODUCTION LOSS		
Total	**2 454 256** (1 784 828 - 2 114 114)	**733 050** (395 774 - 1 077 050)
Analysis perspective		
Healthcare perspective	**1 949 556**	**888 948**
Extended healthcare perspective	**2 160 826**	**1 060 964**
Societal perspective (all sectors)	**5 347 950**	**2 375 698**

Analysis perspectives: **Health care perspective:** Direct medical costs only. **Extended healthcare perspective**: Direct medical and non-medical costs incl. travel and time. **Societal perspective:** Costs from all sectors incl. education sector and production loss.

Total discounted lifetime QALYS were 22.4 (90% CB 21.7 – 23.0) and undiscounted lifetime QALYs 60.5 (90% CB 58.5 – 62.4). Table 6.2.

Table 6.2 Total undiscounted and discounted lifetime QALYs

	Undiscounted (90% credible bound)	Discounted (90% credible bound)
Total lifetime QALYs	**60.5** (58.5 – 62.4)	**22.4** (21.7 – 22.9)

6.2 Exploratory analysis of AVT

Table 6.3 presents the results from the exploratory analysis of AVT and includes incremental costs and incremental QALYs and the corresponding ICERs and NMBs. From the societal perspective, the discounted ICER indicated that AVT may be considered cost saving. The discounted ICER for the extended health care perspective, including the travel, time and hotel costs associated with the health care sector, was NOK 731 036 per QALY and would not be considered cost-effective by a WTP threshold of NOK 500 000/QALY. When only medical costs were included, taking the health care perspective, the ICER was NOK 163 823 per QALY and cost-effective at a threshold of 300 000 NOK per QALY. The NMB corresponds with the ICERs with positive values indicating cost-effectiveness.

The same trends, as for the discounted values, were seen for the undiscounted. The health care and the extended health care perspectives had ICERs indicating cost-effectiveness being under a threshold of NOK 300 000 per QALY and the societal perspective was cost saving. In the extended health care perspective, the ICER increased, but less than for the discounted values, to 254 980 NOK per QALY. The health care perspective with exclusion of the non-medical costs, had an ICER of NOK 57 708 per QALY.

Table 6.3 ICER and NMB by perspectives

Perspective	Current costs	Costs AVT	Increment costs	Current QALYs	QALYs AVT	Increment QALYs	ICER Δ costs/ Δ QALYs	NMB Threshold 300 000	NMB Threshold 500 000
Undiscounted									
Health care	1 949 556	1 968 529	18 973	60.5	60.8	0.33	**57 708**	79 659	145 414
Extended health care	2 160 826	2 244 657	83 831	60.5	60.8	0.33	**254 980**	14 801	80 556
Societal	5 347 950	5 171 164	-176 786	60.5	60.8	0.33	**Cost saving**	275 418	341 173
Discounted									
Health care	888 948	907 836	18 888	22.4	22.5	0,12	**163 823**	15 701	38 760
Extended health care	1 060 964	1 145 249	84 285	22.4	22.5	0.12	**731 036**	-49 696	-26 637
Societal	2 354 857	2 363 685	8 828	22.4	22.5	0.12	**76 569**	25 761	48 820

6.3 Sensitivity analysis

A separate analysis was run by replacing the PedsQL with the KINDL-questionnaire as input for HRQoL, while no changes were made to any other input parameters. There were no significant changes in the cost outcomes compared with the base case main descriptive analysis as expected when only changing the HRQoL-instrument (Table 6.4). The discounted lifetime QALYs were similar while the undiscounted QALYs decreased slightly from 60.5 to 56.5 QALYs (Table 6.5).

Table 6.4 Lifetime costs by sector and analytic perspective using KINDL as HRQoL-instrument

	Undiscounted (90% credible bound)	**Discounted** (90% credible bound)
DIRECT HEALTH CARE (Specialist health care only)		
Direct medical specialist health care	**1 946 016** (1 778 030 - 2 110 573)	**888 777** (820 615 - 953 911)
Direct non-medical specialist health care: Time, travel, and hotel	**210 840** (199 043- 222 429)	**164 352** (153 412 - 175 544)
Total	**2 156 856** (1 988 033 - 2 325 337)	**1 053 129** (984 098 - 1 121 550)
EDUCATION SECTOR		
Accommodation and follow-up in kindergarten and school	**594 868** (510 963 - 681 081)	**470 698** (411 142 - 534 482)
Caregivers time	**140 901** (126 054- 156 297)	**113 316** (102 886 - 124 468)
Total	**735 769** (638 570 - 836 462)	**584 014** (513 989 - 657 953)
PRODUCTION LOSS		
Total	**2 234 374** (1 307 642 - 3 701 150)	**747 424** (402 711 -1 122 972)
Analysis perspective		
Healthcare perspective	**1 946 016**	**888 777**
Extended healthcare perspective	**2 156 856**	**1 053 129**
Societal perspective (all sectors)	**5 364 325**	**2 384 568**

Analysis perspectives: **Health care perspective:** Direct medical costs only. **Extended healthcare perspective**: Direct medical and non-medical costs incl. travel and time. **Societal perspective:** Costs from all sectors incl. education sector and production loss.

Table 6.5 Total undiscounted and discounted QALYs using KINDL as HRQoL-instrument

	Undiscounted (90% credible bound)	**Discounted (90% credible bound)**
Total lifetime QALYs	**56.5** (54.9 – 58.1)	**22.4** (21.7 – 23.0)

The same trends for the ICERs as in the base case analysis were seen for the exploratory analysis of AVT (Table 6.6). The discounted ICER of the societal perspective indicated AVT as cost saving. In comparison the discounted ICER in the base case analysis had an ICER of NOK 76 569 per QALY for the societal perspective indicating some extra costs for implementing AVT. From the extended health care perspective, the discounted ICER was NOK 818 541 and not cost-effective equivalent to the base case. As in the base case AVT could be considered cost-effective from the health care perspective (including only medical costs) with an ICER of NOK 185 688 per QALY. The ICERs from the undiscounted values were comparable to the base case and were cost saving from the societal perspective, NOK 129 704 for the extended health care perspective, and NOK 29 524 for the health care perspective.

Table 6.6 ICER and NMB by perspectives using KINDL as HRQoL-instrument

Perspective	Current costs	Costs AVT	Increment costs	Base case QALYs	QALYs AVT	Increment QALYs	ICER Δ costs/ Δ QALYs	NMB Threshold 300 000	NMB Threshold 500 000
Undiscounted									
Health care	1 946 016	1 965 078	19 062	56.5	57.2	0.65	**29 524**	174 632	303 761
Extended health care	2 156 856	2 240 599	83 744	56.5	57.2	0.65	**129 704**	109 951	239 080
Societal	5 364 325	5 184 715	-179 610	56.5	57.2	0.65	**Cost saving**	373 304	502 433
Discounted									
Health care	886 342	907 751	21 409	22.4	22.5	0.12	**185 688**	13 180	36 239
Extended health care	1 051 061	1 145 435	94 374	22.4	22.5	0.12	**818 541**	-59 785	-36 726
Societal perspective	2 409 868	2 362 795	-47 073	22.4	22.5	0.12	**Cost saving**	81 662	104 721

7 Discussion

7.1 General

A decision-analytical model was developed to estimate the lifetime costs and effects of children receiving CI. This analysis has provided insight into, and an overview of lifetime costs related to cochlear implantation, not only for costs incurring in specialist care, but also for costs required for follow-up and supportive care in other parts of the society and for production loss.

The estimated discounted costs in the main descriptive analysis differed substantially by perspective and were estimated to NOK 888 948 in the health care perspective, to NOK 1 060 964 in the extended health care perspective and to NOK 2 375 698 in the societal perspective. The analysis indicates that non-health care costs related to CI may be high relative to health care costs. Interestingly the costs in the education sector and for production loss, totally NOK 1 314 733, accounted for 55.3% of all discounted costs through lifetime. The total discounted costs in the education sector accounted for 24.5% of the total costs, and out of these 19.7% were related to supportive care by personnel in kindergarten and school and the remaining 4.8% to caregiver's use of time. Production loss assuming unemployment of 15% in health state low and 25% in Very Low- was NOK 733 050 and made up 30% of the total lifetime costs. The non-medical costs of travel, caregiver's time and hotel related to appointments in the health care sector were NOK 172 016 accounting for 16.2% of the total costs estimated in the health care sector. In comparison with the expected remaining QALYs in the general population at age 3 estimated to 68.3 (NiPH, 2021), the lifetime QALYs of 60.5 in our population was considerably lower.

Our exploratory analysis of AVT was motivated by 1) current data indicating better improvements in language skills with AVT compared to other types of language training and 2) previous studies indicating an association between language skills and quality of life (Fortunato-Tavares et al., 2012; Haukedal et al., 2018). We explored how systematic training like AVT could affect resource use and HRQoL in the long term, and how investment in training early in life could affect societal aspects like unemployment later in life when dividing a group of children by different health states. We explored if there was a gain for the society as whole and how it could be seen from different perspectives.

7.1.1 The cost-effectiveness in exploratory analysis of AVT by perspective.

The ICERs from the societal perspective from the discounted values indicated AVT as cost-effective. Compared with the societal perspective, the discounted ICER in the extended health care perspective was higher and estimated at NOK 731 036 per QALY, not being cost-effective. The ICER decreased to NOK 163 823 per QALY in the health care perspective indicating cost-effectiveness.

In both the health care and extended health care perspective, the costs were the same for all the health states and we could not expect to see any costs savings by AVT. In the societal perspective, however, the costs and HRQoL differed between the health state and higher language skills, due to AVT, resulted in lower costs and higher HRQoL. We demonstrated that adding a broader perspective can give important information on gains for the society as whole.

Currently, societal costs are not considered when interventions for health improvements are evaluated for reimbursement. There is however an ongoing discussion whether a societal perspective should be included in decision making and if any recommendations should be added in the revised upcoming "Stortingsmelding om prioritering" (Kalveland, 2023; Legemiddelindustrien (LMI), 2023). The proportion of people in employment may become more and more important in the future as the population is aging and the proportion of elderly not working are increasing. Being part of the work force and contribute to the society may also influence the quality of life in a positive direction.

We can find support for our results from the AVT analysis in a socio-economic analysis of the effect of AVT on employment recently performed by a Danish group. This analysis has been summarized in their AVT impact report (Hallstrøm, 2022). This group based their analysis on proportions of the children reaching language skills equal to their age-equivalent peers at school start, with and without AVT. They used numbers from Danish and international studies with estimates of 80% of children receiving AVT reaching age-equivalent levels of language skills at school age and only 30% of the children not receiving AVT. They further assumed that reaching normal language levels at school age give these children prerequisites to participate in education and being part of the work force. When projecting the effect into higher employment in the future, the found that AVT can give a societal gain of DKK 372 million per class year. The cost of 3 years of AVT per class year was calculated to DKK 7,4 million.

7.2 Strengths and limitations

A main strength of this analysis is that the model structure and its parameter assumptions were developed in close collaboration with experts at University of Oslo and Rikshospitalet OUH with thorough knowledge about, and insight into CI and hearing loss. Access to these experts made it possible to get extensive data for resource use in the health care sector. This team of experts also had access to raw data collected from patients included in Norwegian studies, on which we derived the transition probabilities and HRQoL.

The project has provided new detailed information about procedures and costs related to CI in the specialist health care sector in Norway. Setting up a model with a lifelong time-horizon and with a societal perspective may fill some of the knowledge gaps in this patient group and provide background for further analyses in the field of hearing loss.

However, there are several limitations to this analysis. Sufficient data for HRQoL for individuals above 18 years of age were not available in the Norwegian dataset, and the HRQoL was set to be the same during the whole time-horizon. The HRQoL associated with the three health states may possibly vary through life. The majority of HRQoL- data in the published literature for the adult population, have been collected before and after CI in previously hearing persons and were not considered relevant for our population of pre-lingually deaf children (Dixon et al., 2023).

We did not have good available data for language tests reflecting functional language. Some data for tests like MSEL could reflect functional language better, but for the period from 3 years after CI and onwards there was only one data collection point for this test. The BPVS testing receptive language can give an indication of how the children will perform on other dimensions of language but may not be the best measure for predicting skills through life. The BPVS test has been normed for Norwegian language, but do not have a sufficient range and a floor effect exist for children with CI. The test also has a ceiling effect as norms only was obtained for persons up to 16 years (Halaas-Lyster et al., 2010).

The transition probabilities for this analysis were derived from a Norwegian longitudinal study following the children the first 6 years after CI. Transition probabilities after 6 years were based on the same values as from the 6 first years. We need more knowledge about language skills for these children later in life and in adulthood to get more robust data. The children in the Norwegian study are still followed up and some data were collected after 13 years. These have been analysed but were not included in the calculations of transitions probabilities.

In contrast to specialist care, we have limited or no information about other sectors such as primary health care, education sector and social services. Furthermore, we do not know how these costs relate to levels of language skills achieved after CI. The inclusion of resource use in the education sector was mainly based on assumptions. In our limited timeframe, we were not able to identify all resources or to collect cost data from registers or databases or to receive enough information from persons involved in the follow-up and supportive care of these children.

Similarly, we did not have complete information about the production loss related to language skills in patients with cochlear implantation. Children receiving early simultaneous bilateral CI with the most recently technologically updated devices have not yet reached the relevant age for work life. Production loss was therefore based on assumptions and input from experts. According to an expert team, it is difficult to give any numbers for unemployment due to impaired hearing, but a suggestion of 40-70% unemployment for this group was given. The Norwegian job market may be better compared to other countries, and not all jobs require good hearing or language.

There are many types of relevant resource use not captured in this analysis. Examples are costs for special school, special classrooms, sign language interpreter, note taker for pupils and students with hearing loss, social services, potential increased use of primary health care, parent courses and extra follow-up of parents and children from the specialist health care. The societal perspective defined in this analysis may have been too narrow by terminology as few types of costs were included.

In the education sector, costs for speech therapist, social worker and increased caregiver's use of time were added, but no effect of this supportive care on language skills was added to the model. Further, the effect and costs of AVT were added in this analysis, but not effects and costs for the more unsystematic language training given today. The costs for such training could outweigh some of the costs for AVT. This could also apply for the effect. Because of lack of data, the magnitude of the effect of AVT on transitions from Low to Normal+ and Very Low- to Low was based purely on assumptions. The duration of the effect of AVT is unknown, and the effect was set by assumption to last until 17 years of age. No relative risks were added to other transitions as for example the probability of staying in the Normal+ state or to the transitions going to a lower level of health state. A probabilistic analysis was performed for the exploratory analysis of AVT with the purpose to account for uncertainty in the parameters. This type of analysis may not have been relevant as most of the inputs for the health state specific costs were based on assumptions including the effect of AVT, but performing this analysis would still account for uncertainty in the transition probabilities and HRQoL.

The 500-project used to define the health states and the HRQoL associated with these did not have the exact population of patients as we studied in this project, i.e, some individuals had received CI later than their first life year and sequential, and not simultaneous bilateral CI. This is due changes in the practice of CI over the years and the fact that patients in this study received CI before all today's standard of care was introduced.

7.3 Policy implications and future research

Our analyses give insight into the lifetime costs of cochlear implantation but has also revealed areas for further study. In particular, we have very limited information of how CI implanted children impacts long term language skills, educational achievement and employment over the lifetime.

Our analysis illustrates how AVT in early life can affect resource use and HRQoL later in life. The results indicate that systematic training like AVT should be offered to all CI children.

8 Conclusion

With the help of clinical experts in the field, for the first time in Norway, we developed a model to capture the long-term health and economic consequences for children receiving bilateral CI. Costs from different sectors were included, and we have obtained insights into the types of costs associated with cochlear implantation. The conceptualized model will be able to formally evaluate AVT programs once data become available.

Core parameters of the CI model were uncertain, but the results indicate considerable lifetime costs of CI and lower HRQoL than in the general population. Of the total discounted societal lifetime costs of NOK 2 375 698, 44.7% fell on the health care sector (including time, travel and hotel), 24.5% on the education sector (including use of time) and 30% of the costs stemmed from production loss.

By dividing the children into health states by language skills, we were able to explore possible outcomes of AVT in the long-term. The exploratory analysis of AVT indicates that effective, early educational interventions may be cost-effective or even cost-saving.

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10 Appendices

10.1 Appendix I: Detailed cost list for health care sector

Medical costs health care sector						
Assessment at local institutions	**Unit**	**Unit cost**	**No. of units**	**Cost**	**Description**	**Source**
Assessment at local institution	Visit	1 000	1.0	1 000	Local hearing centre AABR (automated auditory brainstem response) of audiographer without physician	Expert advice from OUH
Consultation ENT-specialist	Visit	116	1.0	1 116	Consultation ENT-specialist	Normaltariffen 1. July 2022: 3ad (387) + 3bd (171) * 2 = 1116
Assessment at local hospital	Visit	2 482	1.0	2 482	Initial auditory tests at local hospital day unit ABR (auditory brainstem response) /ASSR (auditory steady-state response) Day unit at local hospital - with physician, physicist and audiographer Physician consultation - DRG803U, weight 0,026-Natural sleep/dexdor (nasal spray narcosis) at day unit with monitoring/surveillance	Expert advice from OUH
Assessment at Oslo University Hospital						
Assessment by physicist	Visit	2 482	1.0	2 482	Initial auditory tests by physicist at day unit OUH (brain stem)	Expert advice from OUH
Assessment by physician	Visit	1 430	1.0	1 430	Otomicroscopic assessment by physician (DRG803U)	DRG-list, Code 803U
Assessment by pedagogue	Hour	422	12.25	5 169	Conversations, tests and paperwork by pedagogue, Conversations and tests: Observations, Ling Sounds, Mullen (1 hour) Paperwork (1,5 days = 11.25 hours)	Expert advice from OUH
Assessment by audiographer	Hour	459	1.0	459	Auditory tests by audiographer, OAE (otoacoustic emissions), VRA (Visual Reinforcement Audiometry), play audiometry, tympanometry	Expert advice from OUH
Cross Functional Meeting	Hour	2 515	0.5	1 258	Cross functional meeting with physician, pedagogue, physicist and audiographer, Approx 30 minutes	Expert advice from OUH
Summary conversation	Hour	1 385	0.5	692	Summary meeting with caregivers by physician and pedagogue. Approx 30 minutes	Expert advice from OUH
MRI temporal bone	Examination	1 124	1.0	1 124	MRI temporal bone	HELFO and Out-patient regulations (poliklinikkforskriften)
CT temporal bone	Examination	1 062	1.0	1 062	CT temporal bone	HELFO and Out-patient regulations (poliklinikkforskriften)
Fitting of hearing aid	Visit	1 918	1.0	1 918	Fitting of hearing aid(s) and cost of device	Expert advice from University of Oslo
Medical check by physician	Hour	963	0.5	481	Consultation and check with physician before surgery, consultation with doctor at "junior" level going through allergies, heart etc. preparing for surgery	Expert advice from OUH
Meningitis vaccination	Vaccine + visit	919	1.0	919	Meningitis vaccination	Norwegian Pharmaceutical Compendium, Normaltariffen 2ad
Administrative work by coordinator	Hour	358	1.0	358	Administrative work by coordinator (setting up time for operation etc)	Expert advice from OUH

Implantation						
Surgery and device	Unit	200 653	1.0	200 653	Bilateral CI - surgery	Expert advice from OUH
Onset of sound	Unit	11 369	1.0	11 369	Onset of sound 4-6 weeks after implantation by physicist and pedagogue	Expert advice from OUH
Recurrent maintenance costs						
Upgrade of processor	Unit	96 250	0.17	16 035	Processor upgrade (Every 6th year from Age 6)	Expert advice, (DRG-liste Kode 49C, points = 1,751)
Annual administration costs	Unit	500	1.0	500	Annual administration costs (from 2nd Year onwards)	Assumption
Technical Accessories (paid by parents/patient)	Unit	1 000	1.0	1 000	Parent's out-of-pocket costs for batteries, cables, and other accessories, patient's out-of-pocket costs after 18 years	Assumption
Routine controls (until 18 years)						
Routine controls Initial (First year)	Visit	6 907	1.0	6 907	Controls every 3 months after operation by physicist and pedagogue (3, 6, 9 and 12 months)	Expert advice from OUH
Routine controls (age 1)	Visit	5 141	1.0	5 141	Controls every 6 months at age 1 - 2 by physicist and pedagogue (18 and 24 months)	Expert advice from OUH
Routine controls (age 2-18)	Visit	2 571	1.0	2 571	Yearly controls from age 2 until 18 years by physicist and pedagogue	Expert advice from OUH
Re-implantation						
Surgery and sound onset	Unit	212 022	1.0	212 022	Once during lifetime (estimated duration of implant 25-30 years +)	Expert advice from OUH
Cross Functional Meeting	Hour	2 515	0.5	1 258	Cross functional meeting with physician, pedagogue, physicist and audiographer, Cross functional meeting with physician, pedagogue, physicist and audiographer. Approx 30 minutes	Expert advice from OUH
Summary conversation	Hour	1 385	0.5	692	Summary meeting with caregivers by physician and pedagogue, Summary meeting with caregivers by physician and pedagogue. Approx 30 minutes.	Expert advice from OUH
MRI temporal bone	Examination	1 000	1.0	1 000	MRI temporal bone	HELFO and Out-patient regulations
CT temporal bone	Examination	9 126	1.0	9 126	CT temporal bone	HELFO and Out-patient regulations
Medical check by physician	Hour	963	0.5	481	Consultation and check with physician before surgery, ENT surgeon consultation, consultation with doctor at "junior" level going through allergies, heart etc preparing for surgery	Expert advice from OUH
Administrative work by coordinator	Hour	358	1.0	358	Administrative work by coordinator (setting up time for operation etc)	Expert advice from OUH
Auditory visual therapy (AVT) – First year after CI						
AVT_physical meeting	Hour	422	45	18 988	Training with pedagogue (1 hour) + 1,5 hours administration work, 18 times first year (every other week)	Expert advice from OUH
AVT_digital meeting	Hour	422	45	18 988	Training with pedagogue (1 hour) + 1,5 hours administration work, 18 times first year (every other week)	Expert advice from OUH

Facilitation in kindergarten and school						
Assistive listening devices	Unit	10 000	1.0	10 000	Provision of assistive listening devices (remote microphones, FM systems, telecoil, neck loop, Bluetooth streaming dongle etc.) Paid by NAV.	Expert advice from OUH

Non-medical costs (health care sector)

Time, travel, and hotel	Cost	Unit cost	No. of units	Description	Source
First year					
Travel costs (age 0)	20 328	20 328	1	Transportation (caregiver(s) + patient) -Fitting of hearing aid (1 trip) -Surgery (1 trip) -Sound onset (1 trip) -Routine controls (4 trips) Usually both parents are present by surgery and sound onset	Unit database NoMA, Patient's travel one-way trip (2021) – Source: Pasientreiser HF
Caregiver's use of time (age 0)	41 910	41 910	1	Unpaid caregiver-time costs (6 days - First assessment (1 day), surgery (1 day), fitting of hearing aid (1 day) and sound onset (3 days))	Statistics Norway, https://www.ssb.no/arbeid-og-lonn/lonn-og-arbeidskraftkostnader/statistikk/lonn
Hotel stays (age 0)	2 595	2 595	1	3 nights (2 nights by sound onset, 1 night by surgery)	Statistics Norway, table 12897 (2021), Assumption
Year 2 and beyond					
Travel routine control (age 1)	5 808	726	8	One caregiver + child - 2 return trips	Expert advice from OUH, Unit database NoMA, Patient's travel one-way trip (2021) – Source: Pasientreiser HF
Travel routine control (age 2-18)	2 904	726	4	One caregiver + child - 1 return trip	
Time caregiver routine control (age 1)	3 810	254	15	Time used in connection with 2 routine controls at OUH	Expert advice from OUH, Unit database NoMA
Time caregiver routine control (age 2-18)	1 905	254	7.5	Time used in connection with 1 routine control at OUH	
Travel Processor Upgrade (age 6-18)	2 904	726	4	Travel in connection with processor upgrade at OUH 6-18 years	Expert advice from OUH, Unit database NoMA, Patient's travel one-way trip (2021) - Source Pasientreiser HF
Time Processor Upgrade (age 6-18)	1 905	254	7.5	Time used in connection with processor upgrade at OUH 6-18 years	Expert advice from OUH, Unit database NoMA
Travel Processor Upgrade (age 19-106)	1 452	726	2	Travel in connection with processor upgrade at OUH 19-106 years	Expert advice from OUH, Unit database NoMA, Patient's travel one-way trip (2021) – Source: Pasientreiser HF
Time Processor Upgrade (age 19-106)	1 905	254	7.5	Time used in connection with processor upgrade at OUH 19-106 years	Expert advice from OUH, Unit database NoMA
Travel re-implantation (age 40)	1 452	726	2	Travel by re-implantation	Statistics Norway, table 12897 (2021), Assumptions
Time re-implantation (age 40)	3 810	254	15	Time used by re-implantation	Expert advice from OUH
Hotel re-implantation (age 40)	865	865	1	Hotel stay one night	Statistics Norway, table 12897 (2021), Assumption
Travel AVT	46 464	726	64	One caregiver + child - 1 return trips	Travel to AVT - yearly cost the first year
Time caregiver AVT	18 288	254	72	One caregiver - 18 times half day (4 hours)	Time used by AVT - yearly cost the first year

10.2 Appendix II: Hourly wages used for cost estimation

Health care sector	Hourly wage	Monthly wage	Yearly wage	Hours per year	Occupation	Source
Physician specialist	963	96 640	1 159 680	166.5	2212 Specialist medical practitioners	Statistics Norway, table 11418, Private sector and public enterprises (2021)
Audiographer	459	48 490	581 880	1750	2266 Audiologists and speech therapists	
Pedagogue	422	44 590	535 080	1750	2352 Special needs teachers	
Physicist	672	71 010	852 120	1750	2111 Physicists and astronomers	
Coordinator	358	37 860	454 320	1750	3256 Medical assistants	
Special Nurse	563	56 480	677 760	1662.5	2221 Nursing professionals	
Radiographer	450	47 530	570 360	1750	3211 Medical imaging and therapeutic equipment technicians	
GP Physician	746	78 790	945 480	1750	2211 Generalist medical practitioners	
Caregiver average wage	254	50 790	609 480	1750	Net wage, SSB 2021 average all sectors (27% tax subtracted)	Statistics Norway, https://www.ssb.no/arbeid-og-lonn/lonn-og-arbeidskraftkostnader/statistikk/lonn
Education sector	**Hourly wage**	**Monthly wage**	**Yearly wage**	**Hours per year**	**Occupation**	**Source**
Senior Advisor	243	48 480	581 760	1750	2633 Advisors/researchers within humanistic fields	Statistics Norway, table 11418, Private sector and public enterprises (2021)
Assistant Kindergarten	171	34 260	411 120	1750	5311 Barnehage- og skolefritidsassistenter mv.	
Pre School-Teacher	223	44 590	535 080	1750	2342 Pre-school teachers	
Speech Therapist	232	46 350	556 200	1750	2266 Audiographer and Speech therapist (logoped)	
Teacher Primary School	244	48 700	584 400	1750	2341 Primary school teachers	
Teacher High School	262	52 390	628 680	1750	2330 Teacher high school (lektor)	
Social Worker	223	44 610	535 320	1750	3412 Social worker within social fields	
Caregiver average wage	254	50 790	609 480	1750	Net wage, SSB 2021 average all sectors (27% tax subtracted)	Statistics Norway, https://www.ssb.no/arbeid-og-lonn/lonn-og-arbeidskraftkostnader/statistikk/lonn

10.3 Appendix III: Unit calculations

Procedure / assets	Resource	Unit	Unit cost	No. of units	Cost	Notes	Source
Initial auditory tests at OUS: ABR (auditory brainstem respons) /ASSR (auditory steady-state response)							
Drug narcosis	Dexdor	1 ampoule	194	1	194	Dexdor: 1 ampulle 2ml = 200 mikrogram (5 ampuller = 1296,10: 1 ampulle = NOK 259,22, excl. VAT = 0,75 * 259,22 = NOK 194,42 (minste vedlikeholdsdose voksne (ikke indisert hos barn) = 0,2 mikrog/kg/time (infusjon).	Norwegian Pharmaceutical Compendium
Administration of narcosis	Anaesthesia nurse	Hour	563	0,25	141		Assumption
Administration of narcosis	Anaesthetic physician	Hour	963	0,25	241		Assumption
Monitoring of sleep / narcosis	Anaesthesia nurse	Hour	563	1	563		Assumption
Auditory tests (ABR / ASSR)	Audio physicist	Hour	672	2	1344	Repeated testing (required in approx. 50% of children coming from local hospitals): ABR/ASSR (auditory brainstem respons / auditory steady-state response)	Expert advice OUH
<u>Total</u>					**<u>2482</u>**		
MR temporal bone							
Reimbursement HELFO	MR imaging	Unit	295	2	590	Multiply with 2	Out-patient regulations: https://lovdata.no/dokument/SF/forskrift/2007-12-19-1761/KAPITTEL_4#KAPITTEL_4
Deductible	MR imaging	Unit	267	2	534	Multiply with 2	
<u>Total</u>					**<u>1124</u>**		
CT temporal bone							
Reimbursement HELFO	CT imaging	Unit	264	2	528	Multiply with 2	CT temporal bone, Reimbursement category = CT2, Reimbursement HELFO=NOK. 264 , deductible)= NOK 267, Total NOK 531,-. Incl. Framework budget NOK. 531,- * 2 = NOK 1062
Deductible	CT imaging	Unit	267	2	534	Multiply with 2	
<u>Total</u>					**<u>1062</u>**		
Fitting of hearing aid							
Consultations with audiographer	Audiographer	Hour	459	2	918	Fitting of hearing aid(s) and cost of device - 2 consultations with audiographer (2 hours) - HA device: NOK 6000 one side. Duration of use 6 months. NOK 1000, -	Resources (discussed resources at (Teams) costing meeting with Sølvi + Ivar 2nd June 22):
Medical device	Device one side for 6 months	Unit	1000	1	1000		
<u>Total</u>					**<u>1918</u>**		
Meningitis vaccination							
Medication	Nimenrix 2 doses	Unit	291,6	2	583,2		Norwegian Pharmaceutical Compendium, AUP 388,80
General practitioner (GP)	Consultation GP x 2	Unit	168	2	336		Normaltariffen 2ad
<u>Total</u>					**<u>919</u>**		
Bilateral CI-surgery							
Surgeon	Surgeon	Hour	963	4	3850	-1 patient per day per operating room (NOK 5000) -Surgeon: 2 hours per side (total 4 hours) NOK 10000	Bilateral CI - surgery
Narcosis	Drug	Unit	1000	1	1000		Norwegian Pharmaceutical Compendium

Anaesthetic	Anaesthesia nurse	Hour	563	7	3938	-Narcosis: 8.30 -14.00 - 5 h 30 min Drug cost NOK 1 000 -Personnel in room: 8.00 - 14.30 - Anaesthesiologist (7 hours), Anaesthesia nurse (7 hours) NOK 18 900 -Recovery room and 1 day hospitalization NOK 6000 -Medical device cost (NOK 171 000) -One night hospitalization after surgery (inflation adjusted from 2017 NOK)	Expert advice OUH
Anaesthetic	Anaesthesiologist	Hour	963	7	6738		Expert advice OUH
Recovery	Recovery and hospitalization	Unit	9126	1	9126		Expert advice OUH
Medical device	Cochlear device	Unit	171000	1	171000		Expert advice OUH
Operation room	Facility	Unit	5000	1	5000		Assumption
Total					**200 653**		
Onset of sound							
Adjustments over 3 days	Physicist	Hour	672	7,5	5040	Physicist: Adjustments over 3 days (2,5 hours per day x 3) NOK 9000	Expert advice OUH
Sound, guidance, and journal reporting	Pedagogue	Hour	422	15	6329	Pedagogue: 7 hours sound + 2-3 hours guidance, 5 hours journal reporting and writing letters, contacting network (total 15 hours) NOK 18000	Expert advice OUH
Total					**11369**		
Routine controls at OUS							
Pedagogue (age 0)	Pedagogue	Hour	422	2,5	4	3, 6, 9 and 12 months	Expert advice OUH
Physicist (age 0)	Physicist	Hour	672	1,0	4,0	3, 6, 9 and 12 months	Expert advice OUH
Total age 0					**6907**		
Pedagogue (age 1)	Pedagogue	Hour	422	4,5	2,0	18 and 24 months	Expert advice OUH
Physicist (age 1)	Physicist	Hour	672	1,0	2,0	18 and 24 months	Expert advice OUH
Total age 1					**5141**		
Pedagogue from age 2	Pedagogue	Hour	422	4,5	1,0	36 months until 18 years	Expert advice OUH
Physicist from age 2	Physicist	Hour	672	1,0	1,0	36 months until 18 years	Expert advice OUH
Total age 2 onwards					**2571**		
Travel to OUH first year (age 0)							
Travel for assessment	Caregivers and child	Unit	726	6	4356	Both caregivers + child	Expert advice OUH, Unit database NoMA, Patient's travel per trip (2021) - Source Pasientreiser HF
Travel for fitting of HA	Caregivers and child	Unit	726	6	4356	Both caregivers + child	
Travel for surgery	Caregivers and child	Unit	726	6	4356	Both caregivers + child	
Travel for sound onset	Caregivers and child	Unit	726	6	4356	Both caregivers + child	
Travel for routine controls	Caregiver and child	Unit	726	4	2904	One caregiver + child	
					20328		
Travel to OUH beyond first year (age 0)							
Travel for routine controls (age 1)	Caregiver and child	Unit	726	4	2904	One caregiver + child	Expert advice OUH, Unit database NoMA, Patient's travel per trip (2021) - Source Pasientreiser HF
Travel for routine controls (age 2-18)	Caregiver and child	Unit	726	2	1452	One caregiver + child	
Caregivers use of time first year (age 0)							

Assessment	Caregivers	Hour	254	15	3810	Both parents	Expert advice OUH, Enhetskostnadsdatabase NoMA, Patient's travel per trip (2021) - Source Pasientreiser HF
Fitting of HA	Caregivers	Hour	254	15	3810	Both parents	
Surgery	Caregivers	Hour	254	15	3810	Both parents	
Sound onset	Caregivers	Hour	254	90	22860	Both parents	
Routine controls	Caregivers	Hour	254	30	7620	One parent - 4 days	
Total					**41910**		
Hotel stays first year (age 0)							
Surgery	Caregivers and child	Unit	865	1	865	1 room x 1 night	Statistics Norway, table 12897 (2021)
Sound onset	Caregivers and child	Unit	865	2	1730	1 room x 2 nights	
Total					**2595**		