

Systematic review of the scientific literature on the economic evaluation of cochlear implants in paediatric patients

Revisione sistematica della letteratura scientifica sulla valutazione economica degli impianti cocleari in pazienti in età pediatrica

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SUMMARY

The aim of the study consists in a systematic review concerning the economic evaluation of cochlear implant (CI) in children by searching the main international clinical and economic electronic databases. All primary studies published in English from January 2000 to May 2010 were included. The types of studies selected concerned partial economic evaluation, including direct and indirect costs of cochlear implantation; complete economic evaluation, including minimization of costs, cost-effectiveness analysis (CEA), cost-utility analysis (CUA) and cost-benefit analysis (CBA) performed through observational and experimental studies. A total of 68 articles were obtained from the database research. Of these, 54 did not meet the inclusion criteria and were eliminated. After reading the abstracts of the 14 articles selected, 11 were considered eligible. The articles were then read in full text. Furthermore, 5 articles identified by bibliography research were added manually. After reading 16 of the selected articles, 9 were included in the review. With regard to the studies included, countries examined, objectives, study design, methodology, prospect of analysis adopted, temporal horizon, the cost categories analyzed strongly differ from one study to another. Cost analysis, cost-effectiveness analysis and an analysis of educational costs associated with cochlear implants were performed. Regarding the cost analysis, only two articles reported both direct cost and indirect costs. The direct cost ranged between € 39,507 and € 68,235 (2011 values). The studies related to cost-effectiveness analysis were not easily comparable: one study reported a cost per QALY ranging between \$ 5197 and \$ 9209; another referred a cost of \$ 2154 for QALY if benefits were not discounted, and \$ 16,546 if discounted. Educational costs are significant, and increase with the level of hearing loss and type of school attended. This systematic review shows that the healthcare costs are high, but savings in terms of indirect and quality of life costs are also significant. Cochlear implantation in a paediatric age is cost-effective. The exiguity and heterogeneity of studies did not allow detailed comparative analysis of the studies included in the review.

KEY WORDS: Hearing loss • Cochlear implant • Cost-analysis • Cost-effectiveness • Education cost

RIASSUNTO

L'obiettivo dello studio consiste nella revisione sistematica della letteratura scientifica avente come oggetto la valutazione economica degli impianti cocleari nei bambini. La revisione sistematica è stata condotta compiendo una ricerca nelle principali banche dati internazionali in ambito clinico ed economico. Sono stati inclusi tutti gli studi primari pubblicati in inglese dal gennaio 2000 al maggio 2010. Le tipologie di studio incluse sono le seguenti: analisi di valutazione economica parziale, quale analisi dei costi diretti e indiretti dell'impianto cocleare; analisi di valutazione economica completa, quali analisi di minimizzazione dei costi, analisi di costo efficacia (ACE), analisi di costo utilità (ACU) e analisi di costo beneficio (ACB) effettuate attraverso studi osservazionali e studi sperimentali. Dalla ricerca nelle banche dati sono risultati 68 articoli. Di questi, 54 non incontrano i criteri di inclusione quindi sono stati eliminati. Dopo la lettura degli abstract dei 14 articoli selezionati, ne sono stati inclusi 11, ritenuti eleggibili. Si è proceduto, dunque, alla lettura degli articoli in full text copy. Sono stati, inoltre, identificati dalla ricerca nelle bibliografie 5 articoli che sono stati aggiunti manualmente. Dopo la lettura dei 16 articoli selezionati, ne sono stati inclusi 9. I paesi studiati, gli obiettivi, il disegno dello studio, la metodologia, la prospettiva di analisi adottata, l'orizzonte temporale, le categorie di costo analizzate differiscono molto da studio a studio. Nella revisione è stata effettuata un'analisi dei costi, della costo-efficacia e dei costi educativi. Rispetto all'analisi dei costi, soltanto due articoli riportano sia i costi diretti che indiretti. I costi diretti variano tra € 39,507 e € 68,235 (€ 2011). Gli studi relativi all'analisi di costo-efficacia non sono facilmente comparabili: uno studio riporta un costo per QALY che oscilla tra \$ 5,197 e \$ 9,209; un altro autore riferisce un costo di \$ 2,154 per QALY se i benefici non sono scontati e di \$ 16,546 se i benefici sono scontati. I costi educativi sono significativi, aumentano con il livello di perdita di udito e il tipo di scuola frequentata. La revisione sistematica mostra che i costi sanitari sono più alti ma i risparmi in termini di costi indiretti e di qualità di vita sono significativi. L'impianto cocleare, dunque, nell'età pediatrica è costo-efficace. L'esiguità e l'eterogeneità degli studi non permette di condurre un'analisi comparativa degli studi inclusi nella review.

PAROLE CHIAVE: Perdita di udito • Impianto cocleare • Analisi dei costi • Costo-efficacia • Costo educativo

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Introduction and aim

A systematic review of the literature was performed to summarize the results of studies on the economic evaluation of cochlear implants in paediatric patients.

Method

This systematic review of the literature was conducted with explicit and reproducible methodology to minimize any possible distortions, biases or erroneous conclusions due to the lack of inclusion of important studies, according to the criteria of The Cochrane Collaboration and others^{1,2}.

Research strategy

The systematic review of the literature was performed in September 2009 and updated on 31 May 2010, interrogating international electronic databases both in the clinical and economic sectors: "PubMed MEDLINE" and the "Centre for Reviews and Dissemination", which includes the Economic Evaluation Database (NHS EED), the Database of Abstracts of Reviews of Effects (DARE) and the Health Technology Assessment (HTA) Database. The search was made using keywords, Boolean operators and Mesh descriptor. The results obtained from database query were imported by RefWorks Version 6.0, a software for the management of bibliographic data that allows removal of duplicate records. After having identified the publications, two reviewers, working independently, reviewed the titles and the abstracts, applying the below-described inclusion and exclusion criteria. Any differing opinions were resolved by discussion.

Criteria of study selection

Inclusion criteria

All primary studies published in English from 2000 up to the moment of the search were eligible for inclusion (May 2010).

Types of studies

- partial economic evaluation, including direct, indirect and quality of life costs of cochlear implantation;
- complete economic evaluation, including minimization of costs, cost-effectiveness, cost-utility and cost-benefit evaluation performed through observational and experimental studies.

Outcomes

In particular, we considered the following outcomes:

- efficacy unit cost measured in physical units through the incremental cost-effectiveness ratio (ICER);
- cost of quality adjusted life years (QALY);

- benefit unit cost measured in monetary units;
- direct and indirect costs of cochlear implantation.

The articles presenting the costs of the years before 2000 were also included in the review.

Exclusion criteria

Studies not meeting the inclusion criteria were excluded, and in particular unreviewed conference papers, systematic reviews, case reports, letters and commentaries were not included. Non-English studies published before 2000 were not considered.

The selected studies were considered eligible as well as articles selected through the consultation of bibliographies of publications, either pertinent or recommended by professionals engaged in this field, and responding to the inclusion criteria defined, in order to minimize any possible distortions, biases, or erroneous conclusions due to the lack of inclusion of important studies on the issue. After identification, two reviewers, working independently, reviewed the titles and the abstracts, applying the eligibility criteria previously defined. Eligible studies were carefully examined in full text, assessing coherence, consistency and methodological quality, using the available tools to evaluate both internal and external validity, including the criteria of Evers³.

Results

A total of 68 articles were obtained from the database research. Of these, 54 did not meet the inclusion criteria and were eliminated. After reading the abstracts of the remaining 14 articles, 11 were considered eligible. The articles were then read in full text. Furthermore, 5 articles identified by bibliography research were added manually. After reading these 16 articles, 9 were included in the review. Following is a list of the 7 articles excluded, along with the reasons for exclusion.

O'Neill et al.⁴, as the method used for assessment of the costs was not clear.

Sach et al.⁵, because the article deals with cost-effectiveness, although the costs are only briefly mentioned and not reported exhaustively.

Copeland et al.⁶, as it was too generic, and does not perform a cost analysis.

Taylor et al.⁷, because it was not sufficiently clear with regard to the content in the article. An economic evaluation was undertaken, but the Authors only report on other cost-effectiveness studies (which in any case are treated too superficially). The costs for cochlear implants are not provided, and only an estimate of the potential needs is performed, reporting the aggregate costs for children and adults.

Manrique et al.⁸, due to the absence of a method for identification of the healthcare resources consumed and valorization of cost units. The authors mention cochlear

implantation costs in the abstract only, and do not deal with the problem in the main text. Definition of the target population, follow-up and study design are unclear.

Smith-Olinde et al.⁹, as the authors did not perform an analysis of costs, but only describe the quality of life of children with permanent hearing impairment.

Raine et al.¹⁰, since it is focused only on the absence of use of the cochlear implant, and therefore the reported costs only concern non-usage of the device, while the aim of the present review is to assess cost-effectiveness of implantation.

With regard to the studies included, countries examined, objectives, study design, methodology, prospect of analysis adopted and temporal horizon, the cost categories analyzed strongly differ from one another.

We performed an analysis of costs, a cost-effectiveness analysis and an educational cost analysis associated with cochlear implants. The following is a summary of some of the features of the studies included:

- 7 studies were conducted in Europe (5 in Great Britain^{4,12,17-19}, 1 in Germany¹⁵ and 1 in France¹¹), 1 in the United States¹³ and 1 in Canada¹⁶;
- 2 multicentre studies^{11,12}, 3 single-centre studies^{13,5,6};
- 3 prospective studies¹¹⁻¹³, 3 retrospective studies¹⁴⁻¹⁶, 3 cross-sectional studies¹⁷⁻¹⁹.

The studies had different objectives:

Cheng¹³ aimed to determine the quality of life and cost of cochlear implantation in children with hearing impairment. O'Neill¹⁴ investigated the cost-utility of cochlear implants, and includes cost savings associated with education. Schulze-Gattermann¹⁵ analyzed, from the point of view of the payer, the cost-benefits of cochlear implantation in children with congenital deafness and perilingual deafness, comparing them with children using hearing aids. Barton¹² evaluated the costs of 12 programmes adopted for CI in the UK, using the micro-costing method. Barton¹⁷ has two objectives: the former consisted in evaluating out-of-pocket expenses by the families of hearing-impaired children; the latter attempted to verify the types of differences when assessment is performed from a societal point of view. Barton¹⁸ assessed the cost-effectiveness of cochlear implants from a societal point of view. Barton¹⁹ aimed to evaluate the impact on costs on the education of hearing-impaired children. Fitzpatrick¹⁶ aimed to identify direct and indirect CI costs, and at defining the benefits for children with prelingual deafness. Molinier¹¹ evaluated the direct medical costs of CI from the payer's point of view.

The 9 articles included in the review used different methods for assessment of the economic impact of cochlear implantation in children.

Cheng¹³ and O'Neill¹⁴ conducted a cost-utility study as well as an evaluation of CI costs. Cheng¹³ reported the di-

rect and indirect costs of cochlear implantation; O'Neill¹⁴ analyzed the total direct costs and educational costs for paediatric patients. Schulze-Gattermann¹⁵ performed a cost-benefit study relative to the evaluation of direct and indirect healthcare costs of cochlear implant *vs.* hearing aid. Furthermore, the author analyzed the costs relative to Nursery and Primary Schools. Barton¹² estimated the average cost of CI and the impact of the total cost on the English Healthcare System. Barton¹⁷ studied out-of-pocket expenses made by families of a sample of children with and without cochlear implantation.

The study by Barton¹⁸ analyzed the cost-effectiveness of cochlear implantation from the point of view of CI social costs and "health utility". Barton¹⁹ evaluated CI impact in terms of average cost per child according to the type of school and average yearly costs for the education of implanted children and children with different levels of hearing loss. Using a retrospective study, Fitzpatrick¹⁶ reported on the direct healthcare costs for CI children. Molinier¹¹ analyzed direct healthcare costs in a multicentre prospective study.

The article by Schulze-Gattermann¹⁵ is the only one that compared a group of cochlear implanted children *vs.* a group of children with hearing aid. The other studies do not perform any such comparison. In some cases, comparisons are performed between samples of children who use cochlear implant *vs.* children who do not.

The articles selected by Barton et al.¹⁷⁻¹⁹ refer to the study by Fortnum²⁰ who consider children born from 1980 to 1995, residing in the UK in 1998, with severe permanent hearing loss for a total of 17,600 children.

Seven articles take into account a sample of patients, while the other 2 refer to the costs of CI programs^{12,14}. Molinier¹¹ is the only author who considered both adult and paediatric patients in the same study.

The utilities analyzed were different according to the generic tool employed: time-trade-off (TTO), visual analogue scale (VAS), health utilities index-mark III (HUI)¹³, HUI-3^{18 a}. Applied discount rate: none^{11,16}, 3%^{13,18,19}, 6%^{12,14,5}.

Cost analysis

The studies performing an analysis of direct and indirect costs are Cheng¹³ and Schulze-Gattermann¹⁵. O'Neill¹⁴, Barton¹², Fitzpatrick¹⁶ and Molinier¹¹ only performed an analysis of the direct costs.

According to Cheng¹³, the direct costs for the use of cochlear implants relative to 1999 are \$ 60,228, while indirect costs are \$113,426. Thus, CI implies cost savings of \$ 53,198 per child. The direct costs are estimated using the Medicare Resource-Based Relative-Value Scale (RBRVS) with regard to health benefits provided during the pre-operative, operative and post-operative stages. The

^a An explanation of these quality of life assessment instruments is provided in the paragraph relative to cost-effectiveness.

costs for the device, guarantee and batteries are estimated as the mean costs of the most commonly used implants.

Indirect costs include lost working hours and changes in the fields of education and income. Lost working hours in families were calculated considering 4 hours per visit and average income calculated according to type of work and sex. Three working days of leave were considered at the time of the intervention. Changes in the costs of education are based on differences in educational placements before and after receiving the implant.

Schulze-Gattermann¹⁵ analyzed direct and indirect costs in four groups of patients (group 1: CI before 1 year and 9 months with congenital and prelingual deafness; group 2: CI from 2 to 3 years and 9 months with prelingual or perilingual deafness; group 3: CI from 4 to 6 years and 9 months with prelingual or perilingual deafness; group 4: children with hearing aid). The total costs for the three groups of children using CI varied from € 138,000 to € 177,000 vs. € 160,000 for prosthesis users.

Direct and indirect discounted healthcare costs were € 57,000 for group 1, € 52,000 for group 2, and € 46,000 for group 3. Healthcare costs for children using prostheses were € 18,000. The author reported the aggregate costs, and therefore it is impossible to assess direct and indirect costs separately.

O'Neill¹⁴ evaluated the direct costs of cochlear implants obtained from the Nottingham Paediatric Cochlear Implant Programme. The total cost after the first 4 years was \$ 60,480 and was distributed as follows: the assessment and implant costs were \$ 44,000 (first year), rehabilitation and maintenance \$ 6,400 (second and third year), maintenance \$ 3,680.

Barton¹² defined the average costs relative to the different stages of the implant which were: € 3,743 for assessment, € 27,863 for implantation, € 12,044 for mapping, € 6,209 for the first year of maintenance and € 2,497 for the following years, € 6,569 for updating of the sound processor (every 10 years). The average cost, discounted at 1 year, was € 42,972, after 15 years was € 73,763 and € 95,034 after 73 years.

In the period 2000/2001, the total cost from the perspective of the Healthcare System for CI in 237 children mapping or maintenance in 1290 previously implanted children was estimated to be € 14.2 million. Average lifelong healthcare costs for implantation and maintenance of an implanted child in the UK were estimated to be € 95,000. Fitzpatrick¹⁶ estimated the average direct healthcare costs for each child at \$ 64,171.46 (Canadian dollars), and distributed as follows: 4.8% for assessment, 44.5% for hardware, 2.8% for surgery, 5.4% for hospitalization, 0.1% for post-surgery follow up; follow-up at 1st year absorbs 19.8%, follow-up at 2nd year 12.8% and follow-up at 3rd year accounting for 10.3% of costs.

Molinier¹¹ analyzed direct healthcare costs in a multi-centre prospective study conducted on 268 children in 19

French university hospitals. The average cost per child was € 34,686, of which 64.4% is represented by device costs. Costs were distributed as follows: € 814 for pre-operative assessment (65.7% of costs for tests and outpatient visits, 34.3% for hospitalization), € 24,498 for implantation (8.8% for hospitalization and 91.2% for the device), € 6,743 for rehabilitation (88.5% for tests and outpatient visits, 11.5% for hospitalization) and € 2,631 for travel expenses.

Table I shows a comparison relative to the direct costs reported by the selected authors. The costs were first inflated to the year 2011 and then converted in Euros in case of different currencies. In order to inflate until the year 2011, the Consumer Price Index and the Gross Domestic Product Deflator Index for the euro zone were used for the dollar and euro, respectively. Conversions from the USA dollar to euro (\$1 ± € 0.70) were performed up to 14 June 2011.

From the table, it can be seen that the direct costs range between € 39,507¹¹ and € 68,235¹². The device represents

Table I. Direct costs of cochlear implants in children (Euro, 2011).

Cheng ¹³	€ 56,611 total direct costs
	<ul style="list-style-type: none"> • 4.8% pre-operative costs • 40.6% operative costs • 54.6% post-operative costs
O'Neill ¹⁴	€ 58,104 total direct costs (after the first 4 years)
	<ul style="list-style-type: none"> • 72.7% evaluation and implant • 21.2% rehabilitation and maintenance • 6.1% maintenance
Barton ¹²	€ 68,235 average total direct costs
	<ul style="list-style-type: none"> • 6.5% evaluation • 48.8% implant • 21.1% mapping • 10.9% first year of maintenance • 8.4% second year of maintenance • 4.4% following years
Fitzpatrick ¹⁶	€ 53,197 total direct healthcare costs
	<ul style="list-style-type: none"> • 4.8% evaluation • 52.3% implant • 19.8% post-implant follow-up (1st year) • 12.8% post-implant follow-up (2nd year) • 10.3% post-implant follow-up (3rd year)
Molinier ¹¹	€ 39,507 average direct healthcare costs
	<ul style="list-style-type: none"> • 2.4% pre-operative costs • 70.6% implant • 19.4% rehabilitation • 7.6% travel

Note: Schulze-Gattermann¹⁵ was not included in the review as aggregate direct and indirect costs were reported.

one of the most significant costs: 32% of the total direct costs in Cheng¹³, 72.7% of the total (estimate-associated cost) in O'Neill¹⁴, 48.8% in Barton¹², 52.3% in Fitzpatrick¹⁶ and 70.6% in Molinier¹¹. The percentage relative to post-operative costs was 54.6% of the total costs in Cheng¹³, 42.9% in Fitzpatrick¹⁶, 27.3% in O'Neill¹⁴, 23.7% in Barton¹² and 19.4% in Molinier¹¹.

Cost-effectiveness assessment

The articles assessing cost-effectiveness are Cheng¹³, O'Neill¹⁴ and Barton¹⁸. Cheng¹³ evaluates the cost-utility of cochlear implantation from a social perspective using three different quality of life assessment instruments: visual analogue scale (VAS), time trade off (TTO) and health utility index (HUI). The VAS consists in a scale ranging from 0 (death) to 1 (perfect health) in which respondents indicate a number corresponding to their perception of the quality of life. In TTO, the respondents have two alternatives: alternative 1 is the current state of health for time t (remaining life expectancy); alternative 2 corresponds to an excellent state of health for time x. HUI is a questionnaire organized according to the following health sectors: hearing, language, sight, feeling, pain, deambulation, cognition and personal care.

The mean score reported by VAS respondents reaches 0.27 (along a 0-1 scale) passing from 0.59 before surgery to 0.86 after implantation. The mean score of TTO ranges from 0.75 (immediately before implantation) to 0.97 after implantation, with an increase of 0.22. The HUI score ranges from 0.25 before surgery to 0.64 after implantation, with an increase of 0.39. Direct health costs for QALY are \$ 9,029 using TTO, \$ 7,500 using the VAS and \$ 5,197 using the HUI.

Considering cost-utility, O'Neill¹⁴ reported a cost of \$ 2,154 for QALY if benefits were not discounted and \$ 16,546 if benefits were discounted. The author claimed that if we consider a child implanted at the age of 4 with a life expectancy of 74 years, the QALY costs gained are 16.33.

Barton¹⁸ demonstrated increased health saving costs in children with worse pre-implantation hearing loss, when relating pre-operative hearing loss and an age at 3 and 6 years at the time of the implant. Higher QALY cost savings were associated with worse hearing loss, younger age at time of implantation and longer duration of implant usage. Barton performed an analysis of the QALY costs according to the different prospects: social, sanitary, aggregate sanitary and education costs. Health/QALY costs relative to life duration were estimated to vary between € 15,410 (for a child implanted at 3 years, with pre-operative hearing loss of 125 dB) and € 47,723 (for a child implanted at 6 years with hearing loss of 105 dB). In conclusion, cost-effectiveness is more favourable when life duration is estimated instead of 15-year-age, for children with worse level of hearing loss and for children implanted at a younger age.

Analysis of education costs

O'Neill¹⁴ compared the costs of education in a child with hearing loss between 70 and 95 dB and a child with hearing loss of 95 dB. The costs refer to the 4th year of education (an 8-year-old child at primary school) and 7th year of education (an 11-year 3-month old child at middle school).

Schulze-Gattermann¹⁵ claimed that education costs covered by the public healthcare system depend on the different types of schools. In the base scenario, the costs for deaf children's nursery schools are € 28,820 per year vs. € 7,810 for traditional schools. Primary schools for deaf children cost € 16,410 per year vs. € 4,450 for traditional schools. Furthermore, education costs are higher than healthcare costs. In the UK, education is compulsory from the age of 4 to 16: primary school goes from 4 to 11 years and middle school from 11 to 16 years. In group 1 (0-1.9 years), 58.7% of total costs are represented by education costs and 41.3% by healthcare costs, in group 2 (2-3.9 years) 69.4% is absorbed by education costs and 30.6% by healthcare costs, in group 3 (4-6.9 years) 74% and 26% are education and healthcare costs, respectively, and in group 4 (children using hearing aids) 88.7% of costs are related to education costs and 11.3% to medical costs.

Barton¹⁹ distributed the sample according to the level of hearing loss (moderate, severe, profound 96-105 dB,

Table II. Educational costs for children (Euro, 2011).

	<i>Yearly average costs</i>
O'Neill ¹⁴	Hearing loss of 95 dB
	- child at 4 th yr of education: € 15,507
	- child at 7 th yr of education: € 18,852
	Hearing loss 70-95 dB
	- child at 4 th yr of education: € 11,931
	- child at 7 th yr of education: € 12,114
Schulze-Gattermann ¹⁵	<i>Discounted aggregate educational costs</i>
	- Group 1 (CI before 21 mths): € 100,521
	- Group 2 (CI between 2 yrs and 3 yrs 9 mths): € 146,438
	- Group 3 (CI between 4 yrs and 6 yrs 9 mths): € 162,571
	- Group 4 (children with acoustic prosthesis): € 176,222
Barton ¹⁹	<i>Yearly total educational costs</i>
	- Children with moderate AHL: € 18,338
	- Children with severe AHL: € 26,155
	- Children with profound AHL (96-105 dB): € 31,776
	- Children with profound AHL (> 105 dB): € 37,910
	- Children with implant: € 32,331

profound > 105 dB and implanted) and of type of school attended. The annual costs for education varied between € 15,745 in the group with moderate hearing loss and € 28,059 for implanted children.

Table II shows the educational costs reported by O'Neill¹⁴, Schulze-Gattermann¹⁵ and Barton¹⁹, which were first inflated to 2011 and then converted to Euro as described in the paragraph "Cost analysis". Conversions from USA dollars to Euro (\$ 1 = € 0.70) were performed up to 14 June 2011.

Discussion

The articles examined concern the economic aspects of cochlear implantation according to different approaches: direct and indirect cost analysis, and educational and cost-effectiveness analysis. Cheng¹³ is the only author who reported the direct and indirect costs in detail, clearly specifying the methodology employed for their estimation. Costs are referred to the lifetime of an implanted child. Furthermore, the author conducted a cost-utility analysis with three different utility instruments, stating that cochlear implantation is cost-effective, and generates important health benefits and cost-savings for society. Direct healthcare costs vary from \$ 5,197 to \$ 9,027 for QALY using the three instruments. Schulze-Gattermann¹⁵ deals with direct and indirect, but not aggregate costs, and reports the total costs for the three implanted groups of children compared to the group using cochlear implant. The author reaches the conclusion that cochlear implantation implies a cost-benefit relation that is favourable to implantation *vs.* prosthesis. Educational costs are much higher than healthcare and indirect costs, and increase with the age of the implant.

The article by O'Neill¹⁴ took into account cochlear implantation funding based on full costs, including complications and maintenance with reference to the CI program of the UK. Education costs and cost-utility were also described. The analysis of all types of costs is however too superficial and is unclear.

Barton¹² clearly defined the methodology employed for cost assessment, measuring the resources associated with CI (staff, accommodation, equipment, accessory expenses, hospitalization, device implantation and adverse events), using the micro-costing method^b. The level of use of each resource is reported up to the financial year 1998/1999 and all costs are inflated to 2000/2001.

Mean costs for each implanted child, aggregate expenses for the different phases of the pathway included processor improvement, healthcare costs, and total yearly costs of the healthcare system. Fitzpatrick¹⁶ examined direct and indirect costs; however, the latter are only quoted, but not

evaluated. Direct costs, related to a retrospective analysis of 18 CI children, were referred to the period from pre-implantation assessment to 3 years after implantation. The author quotes the education costs and those incurred by the family, but does not quantify them in monetary terms.

Molinier¹¹ performed a prospective assessment only of the direct costs of CI in France, but the investigation could trigger further cost-effectiveness and cost-utility studies. The economic analysis takes into account all the phases of the patient's pathway with CI, but stops at 1-year of follow up.

The three studies^{13 14 8} perform a cost-effectiveness analysis, but are not easily comparable between themselves; in particular, the analysis made by O'Neill¹⁴ is not as accurate as the ones performed by Cheng¹³ and Barton¹⁸. Cheng¹³ performs a cost-utility analysis according to type of instrument employed, reporting cost per QALY which ranges between \$ 5,197 and \$ 9,209. O'Neill¹⁴ reports cost per QALY which vary from \$ 2,153 when the costs are not discounted, to \$ 16,546 when the discount is applied, concluding that CI is a cost-effective in children with profound hearing-impairment.

Barton¹⁸ carried out an in-depth cost-effectiveness analysis and showed that health utility costs are significant and broad enough to justify costs. Furthermore, he retained that the benefits are greater in children with higher pre-operative hearing loss and in younger children.

In this review, it was decided to also include education costs, which are very significant from an economic point of view, as demonstrated by the authors selected. The costs increase with the level of hearing loss and type of schools attended.

In the analysis by Schulze-Gattermann¹⁵, the percentages of children attending traditional schools varied from 69% in the groups of children implanted before 1 year and 9 months to 12% in the group of children using a hearing aid. The costs both of nursery schools (€ 28,820) and primary schools (€ 16,410) for hearing-impaired children are very high compared to the traditional schools (€ 7,810 and € 4,450, respectively). The author claims that the differences in education settings between groups of children of different ages are supported by several studies showing that hearing, language, and education skills are better in children receiving implantation when they are younger.

A drawback we found in present review is the small number of studies that have been published since 2000, making comparisons difficult. Furthermore, the studies performing an estimate of costs are exclusively concentrated on the direct costs; only Cheng¹³ made an in-depth analysis of indirect costs. However, it was possible to provide an economic picture of the impact of CI.

^b The micro costing method, according to which a cost unit is assigned for each resource, is used to estimate the total costs.

It was not possible to compare the different types of implants (monolateral vs. bilateral, bilateral sequential vs. monolateral implantation), and the implants in children with associated disabilities and hearing aid, as they are not reported by the authors included in the review. The comparisons we found concerned children with cochlear implantation vs. children with acoustic prosthesis¹⁵, implanted vs. non-implanted children¹⁷⁻¹⁹.

No quantitative synthesis of the results using a meta-analysis was feasible, as the data emerging from the articles could not be compared, owing to different factors, including scarce number of studies and differences in terms of country, sample, study design, follow-up, utility measures and cost items.

The outcomes of the various studies could not be pooled due to inherent differences as a result of above features. Since meta-analysis is necessary when the same treatment is assessed in a considerable number of studies, in this case meta-analysis of only a few heterogeneous studies would lead to results that would be unreliable and difficult to reproduce.

Conclusion

Cochlear implant represents a significant surgical innovation, and is the first artificial organ of sense able to evoke acoustic feelings by stimulating the inner ear. CI in children represents a controversial issue from an economic, clinical and ethical point of view. The ethical problem derives from the fact that CI implies a permanent lesion of the receptor and total loss of the residual ear, although it is possible to maintain partial or total hearing in some cases.

A number of studies show that the benefits are superior to the risks, and demonstrate an improvement in the quality of life of implanted children. CI has increased considerably over time, even in children aged ≤ 12 months as the introduction and diffusion of screening have led to a considerable reduction in the mean age at diagnosis of congenital hearing loss.

A congenitally hearing-impaired child identified at birth, with hearing aids in the first 6 months and implanted within two years of age, may be able to attend nursery school with no need for educational support²¹.

Schulze-Gattermann¹⁵ claims that, from the payer's point of view, implantation for prelingual hearing-impaired children is strongly recommended for children receiving an implant before two years of age, which involves saving costs of 13% compared to youngsters using the prosthesis until the age of 16 years. Another critical aspect is represented by high costs, as emerges from the studies included in the review, with particular regard to the costs of the device. If on the one hand the healthcare costs are high, the savings in terms of indirect costs and quality of life are considerable. Furthermore, as we have seen, the elevated

school costs have decreased, especially in the countries in which special schools for hearing impaired children are present.

From the clinical studies, it also emerged that there has been an improvement in communication benefits, supporting the hypotheses of those who claim that early intervention ensures optimal results. The impact of CI is positive for the life of the child both from social and educational point of views.

Therefore, unilateral CI in a paediatric age is cost-effective. On the basis of the present review, no studies have examined cost-effectiveness of bilateral CI in a paediatric age. A number of recent international reviews on CI²² confirm that simultaneous bilateral CI procedure is cost-effective in paediatric patients.

The exiguity and heterogeneity of investigations did not allow detailed comparative analysis of the studies included. Therefore, it was necessary to perform an analysis based on three different approaches: cost analysis, cost-effectiveness analysis and analysis of educational costs, which allowed a small cross-section to be obtained concerning the economic impact on cochlear implants in paediatric patients. It is necessary both to consider the high healthcare and education costs, and to make an assessment in terms of benefits for children with CI.

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