The effectiveness and cost-effectiveness of cochlear implants for severe to profound deafness in children and adults: a systematic review and economic model

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Executive summary

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Executive summary: Cochlear implants for severe to profound deafness in children and adults

Objectives
To assess the clinical effectiveness and cost-effectiveness of cochlear implants for children and adults with severe to profound sensorineural hearing loss by answering the following questions:

1. For severely to profoundly deaf people (either using or not using hearing aids), is it effective and cost-effective to provide a first (i.e. unilateral) cochlear implant?
2. For severely to profoundly deaf people with a single cochlear implant (either unilateral or unilateral with a hearing aid), is it effective and cost-effective to provide a second (i.e. bilateral) cochlear implant?

Methods
These questions were addressed using the following criteria:

Intervention Multichannel cochlear implants using whole-speech processing coding strategies, e.g. ACE, SPEAK, CIS and SMP, i.e. devices that are the same as, or comparable with, those currently available on the NHS.

Comparators In the review of clinical effectiveness, multichannel implants were compared with non-technological support (no devices of any kind) and acoustic hearing aids, and unilateral implants were compared with bilateral implants, and bilateral implants with unilateral implants plus acoustic hearing aids. In the cost-effectiveness analysis the following comparisons were made: no implant versus unilateral implantation; simultaneous bilateral versus unilateral implantation; and sequential bilateral versus unilateral implantation.

Population Children and adults with severe to profound deafness. People with severe loss of hearing cannot detect tones on average at a level below 70–94 decibels hearing level (dB HL) in their better-hearing ear, whereas those with profound hearing loss cannot detect tones below 95 dB HL in their better-hearing ear.

Main outcomes Measures of sensitivity to sound (hearing), speech perception, speech production, adverse effects of treatment, health-related quality of life, and educational outcomes.

Main databases searched Limited to English language papers only but no restriction on publication date. The bibliographies of retrieved references were checked for additional publications. All initial searches were carried out in October 2006 and the update searches were rerun in July 2007. Databases searched included MEDLINE; EMBASE; Cochrane Database of Systematic Reviews; CENTRAL; NHS EED; DARE; HTA (NHS-CRD); EconLit; National Research Register; and ClinicalTrials.gov.

Study selection Studies were included if they were randomised controlled trials (RCTs), quasi-RCTs, or pre-/post, cross-sectional or non-randomised controlled studies. They were excluded if they used either single channel implants or feature extraction or compressed analogue coding strategies, which are not comparable with current NHS practice, or if they were narrative reviews (including literature reviews), preclinical or technical studies, uncontrolled studies, conference abstracts, animal studies, or not relevant to the UK or otherwise outside the criteria for this assessment. Included studies were critically appraised for internal and external validity. For each comparison sufficient studies were included for 75% of the total population of that comparison to be in the assessment. Relevant data were extracted and narrative reviews undertaken, but meta-analyses of the clinical data were not carried out as the data were too heterogeneous to pool. The manufacturers’ submissions to the National Institute for Health and Clinical Excellence were searched for additional evidence.

PenTAG cost-utility model We developed a state-transition (Markov) model of the main care pathways deaf people might follow and the main complications and device failures. The costs (2006 prices) of assessing candidacy, implantation, training and maintenance are included.
Results

Summary of clinical effectiveness

The systematic search produced 1581 abstracts/titles, from which 1436 items were excluded. The evaluation of the 145 papers retrieved left 33 papers in the clinical effectiveness review. These studies, only two of which were RCTs, used 62 different outcome measures. Although there were some notable exceptions (principally those conducted in the UK), overall the studies were of moderate to poor quality with some weaknesses in design and internal validity.

Children

There is considerable heterogeneity in the studies of one cochlear implant versus non-technological support; therefore, pooling of data was not possible. However, there was a large total number of participants \( n = 848 \) and the design of most of the studies was prospective. All studies reported gains on all reported outcome measures, some demonstrating greater gain from earlier implantation. Measures of hearing showed that clear gains were made 6 months post activation onwards, with hearing thresholds ranging from 32 to 44 dB HL post implantation. The results for speech perception and production show a 50% improvement in understanding speech in noise \( \text{Hearing in Noise Test for Children (HINT-C): before implantation, 11\% \pm 21\%; 6 months after, 61\% \pm 37\%} \).

When unilateral cochlear implantation was compared with acoustic hearing aids the results indicate greater gains in all outcomes with cochlear implants. In one study, on a 4-point scale measuring ability to identify everyday sounds, children with cochlear implants had mean scores 1.6 points above those of children with acoustic hearing aids. The speech perception outcomes ranged from a minimal difference in understanding of spoken language of 0.1 points at 24 months post implant to 56.5 points on picture identification tasks.

Comparing unilateral implantation with bilateral implantation the strongest evidence for an advantage from the latter was for understanding speech in noisy conditions, with bilateral implantation giving a mean improvement of 13.2% \( (p < 0.0001) \). Age at second implant was found to affect the speed of improvement and final gain; those receiving their second implant earlier made greater gains.

The comparison of bilateral implants with unilateral cochlear implants plus an acoustic hearing aid was compromised by small sample sizes \( \text{range 10–30} \) and poor reporting. The psychoacoustic results give the strongest evidence: improvement was seen in the ability to detect the direction of sound \( \text{minimal audible angle: bilateral = 28.0\%; unilateral + hearing aid = 44.4\%; } p < 0.05 \). Speech perception was better in children with two cochlear implants. The degree of benefit ranged from a mean difference of 4.0 for the Children’s Realistic Intelligibility and Speech Perception (CRISP) test of matching pictures to spoken words to 25.0 for the Multisyllabic Lexical Neighbourhood Test (MLNT) of recognising spoken words, both in quiet conditions.

None of the studies of children reviewed reported health-related quality of life or educational outcomes. Therefore the searches were screened again for studies with broader inclusion criteria. Six quality of life and seven educational outcome studies were found. Compared with before implantation, cochlear implants improved children’s quality of life. The educational studies showed that children who are implanted before they attend school are more likely to do well academically and attend mainstream education than those implanted after school age. Profoundly deaf children with cochlear implants performed at levels similar to moderately or severely deaf children without implants.

Adults

Comparing unilateral implantation with non-technological support, results for speech perception demonstrated a greater benefit from cochlear implants in all studies. Measures were taken before and post implantation at intervals, with participants acting as their own controls. The overall results indicate an improvement in quality of life from cochlear implant use with a Health Utilities Index 3 \( (\text{HUI-3}) \) gain for traditional candidates of 0.22 \( (95\% \text{ CI 0.19–0.24}) \) and for marginal hearing aid users of 0.15 \( (95\% \text{ CI 0.11–0.19}) \). There is some indication that increased age at implantation may reduce effectiveness \( \text{[normalised index of audiovisual gain (AVGN): } r = 0.164, p < 0.01] \), and also a negative correlation between duration of deafness and effectiveness \( (r = -0.203, p < 0.01) \), with people who had been profoundly deaf for more than 30 years before implantation not showing any significant benefit.

Six studies compared unilateral implantation with acoustic hearing aids. Speech perception measures all showed benefits for cochlear implants, in particular a mean increase in score of 37 points for cochlear implants compared with acoustic hearing aids in noisy conditions \( (p < 0.001) \) with BKB sentences. However, prelingually deafened adults benefited less, with mean change scores of 20% compared with 62% for the postlingually deafened. When participants were asked to rate
functional performance and the effects of cochlear implants on their quality of life, cochlear implants were given a higher functional performance rating (59%) than hearing aids (40%). Another study found commensurate gains in quality of life, with 84% of participants quite or very positive about the impact of cochlear implants on their lives.

The comparison of unilateral with bilateral cochlear implantation demonstrated hearing advantages from bilateral implantation: mean difference for spatial hearing 0.71 (95% CI 0.08–1.33, p < 0.01), quality of hearing 0.7 (95% CI 0.2–1.2, p < 0.05), hearing for speech 9.00 (95% CI 3.00–5.00, p < 0.01) measured on the Speech Hearing, Spatial Hearing and Qualities of Hearing Questionnaire, and for detection of sound direction 24° azimuth (p < 0.001). Benefits in speech perception were significant in noisy conditions on all measures. These ranged from 12.6 for City University of New York (CUNY) sentences (p < 0.001) to 76% for HINT sentences (p < 0.0001). There were particular advantages from the head shadow effect (–3.5, p < 0.0001). However, not all measures showed significant gains.

Quality of life was measured with generic and disease-specific instruments. Two measures showed benefits from bilateral implantation: the Glasgow Health Status Inventory (2.00; 95% CI 1.00–7.00, p < 0.05) and Abbreviated Profile of Hearing Aid Benefit (communication 5.7; SF 0.2, p < 0.0001). However, in another study neutral and negative results came from the HUI-3 (–0.01; 95% CI –0.1 to 0.08, NS), visual analogue scale (VAS; –0.06; 95% CI 0.12–0.00, NS) and EuroQol 5 dimensions (EQ-5D; –0.045; 95% CI –0.12 to 0.03, p < 0.05), although multiple regression indicated that the negative results might have been primarily due to the worsening tinnitus following the second implant.

The qualitative study found that all 17 interviewees easily in social situations. The head shadow effect (–3.5, p < 0.0001). There were particular advantages from the head shadow effect (–3.5, p < 0.0001). However, not all measures showed significant gains.

**Summary of cost-effectiveness**

As there were no data for bilateral implantation in children, estimates of the utility gain were assumed to be the same as for adults. Therefore the incremental cost-effectiveness ratios (ICERs) for bilateral implantation in children are highly speculative.

The PenTAG Markov model base-case analysis (over a lifetime) estimated that, for prelingually profoundly deaf children, in comparison with no cochlear implant use, unilateral implantation conferred an additional 4.48 quality-adjusted life-years (QALYs) for an additional £60,070 per person, giving an estimated ICER of £13,413 per QALY. Simultaneous bilateral implantation conferred an estimated additional 0.67 QALYs for an additional £27,105 per person compared with unilateral implantation, giving an estimated ICER of £40,410 per QALY. Sequential bilateral implantation versus unilateral implantation conferred an estimated additional 0.60 QALYs for an additional £32,657 per person, giving an estimated ICER of £54,098 per QALY.

The PenTAG model base-case analysis estimated that, for postlingually sensorineurally profoundly deaf adults, in comparison with no cochlear implant, unilateral implantation conferred an additional 2.40 QALYs for an additional £33,959 per person, giving an ICER of £14,163 per QALY. Simultaneous bilateral implantation versus unilateral implantation conferred an additional 0.38 QALYs for an additional £19,048 per person, giving an ICER of £49,559 per QALY. Sequential bilateral implantation conferred an additional 0.33 QALYs over unilateral implantation for an additional £19,678 per person, giving an ICER of £54,098 per QALY.

Deterministic one-way sensitivity analyses showed that the cost–utility results were sensitive to changes in discount rates, the time horizon used in the analysis, and the long-term utility gain associated with unilateral implant use compared with not using cochlear implants. Results for bilateral implantation were sensitive to the discount offered on the cost of a second implant system and extremely sensitive to the incremental utility associated with bilateral cochlear implant use as opposed to unilateral implant use.

Probabilistic threshold analyses suggest that, when measured on a lifetime horizon, and compared with either non-technological support or acoustic hearing aids, unilateral cochlear implants are highly likely to be cost-effective for adults and children at willingness to pay thresholds of £20,000 or £30,000 per QALY. There are likely to be overall additional benefits from bilateral implantation, enabling children and adults to hold conversations more easily in social situations.
Children

Probabilistic sensitivity analysis based on 1000 simulated trials showed that, at an assumed maximum willingness to pay threshold of £30,000 (or £20,000) per QALY, unilateral implantation conferred greater net benefit over no implantation in 100% (99.9%) of simulations and was dominated (fewer QALYs for greater cost) in 0%. Again, assuming that the mean incremental utility gain associated with bilateral cochlear implant use is the same in children as in adults, simultaneous bilateral implantation conferred greater net benefit over unilateral implantation in 34.9% (16.6%) of simulations and was dominated in 16.9%. Comparing sequential bilateral implantation and unilateral implantation, the former conferred greater net benefit in 21.3% (5.5%) of simulations and was dominated in 16.2%. However, any changes to the central estimate would have a potentially large impact on any decision uncertainty and could alter these results considerably.

Adults

Probabilistic sensitivity analysis based on 1000 simulated trials showed that, at £30,000 (or £20,000) per QALY, unilateral implantation conferred greater net benefit than no implantation in 100% (100%) of simulations and was dominated (fewer QALYs for greater cost) in 0%. Simultaneous bilateral implantation conferred greater net benefit over unilateral implantation in 20.7% (30%) of simulations and was dominated in 13.2%. Sequential bilateral implantation conferred greater net benefit over unilateral implantation in 8.9% (0.7%) of simulations and was dominated in 12.8%.

Conclusions

Unilateral cochlear implantation is safe and effective for adults and children. In the latter it seems likely that unilateral implantation improves academic performance and may increase the likelihood of children remaining in mainstream education. Greater benefits are derived from earlier implantation and a shorter duration of deafness before implantation. In profoundly deaf adults and profoundly and prelingually deaf children, unilateral cochlear implants are likely to be cost-effective. Probabilistic threshold analyses suggest that, when measured on a lifetime horizon, and compared with either non-technological support or acoustic hearing aids, unilateral cochlear implants are highly likely to be cost-effective for adults and children at willingness to pay thresholds of £20,000 or £30,000 per QALY. There are likely to be overall additional benefits from bilateral implantation, enabling children and adults to hold conversations more easily in social situations. Any conclusion about the cost-effectiveness of bilateral cochlear implants should take into account the high degree of uncertainty within the PenTAG model regarding the probable utility gain.

Suggested future research questions and priorities

1. Determination of the level of residual hearing remaining before it becomes cost-ineffective to provide an implant rather than an acoustic hearing aid.
2. Definition of the earliest age at which the implantation of a congenitally deaf child is safe and effective.
3. Investigation of the utility gain for children from bilateral compared with unilateral implantation.
4. Studies in children and adults enabling mapping (i.e. reliable prediction) from measures of speech perception and production and hearing to validated generic utility assessment instruments.
5. Studies on employment prospects in adults or children using cochlear implants compared with employment prospects in profoundly/severely deaf people.
6. Larger studies with longer follow-up, using standard measures for outcomes and quality of life impact, and recording full data on known covariates of postimplantation speech and quality of life outcomes. There may be a strong case for a national research registry of all cochlear implantees in the UK.
7. Development of a standard classification system for defining levels of functional hearing.
8. More comparative empirical research into the relative effectiveness of, and patient and clinician preferences for, simultaneous versus sequential bilateral implantation.

Publication

The Health Technology Assessment (HTA) programme, part of the National Institute for Health Research (NIHR), was set up in 1993. It produces high-quality research information on the effectiveness, costs and broader impact of health technologies for those who use, manage and provide care in the NHS. ‘Health technologies’ are broadly defined as all interventions used to promote health, prevent and treat disease, and improve rehabilitation and long-term care.

The research findings from the HTA programme directly influence decision-making bodies such as the National Institute for Health and Clinical Excellence (NICE) and the National Screening Committee (NSC). HTA findings also help to improve the quality of clinical practice in the NHS indirectly in that they form a key component of the ‘National Knowledge Service’.

The HTA programme is needs led in that it fills gaps in the evidence needed by the NHS. There are three routes to the start of projects.

First is the commissioned route. Suggestions for research are actively sought from people working in the NHS, from the public and consumer groups and from professional bodies such as royal colleges and NHS trusts. These suggestions are carefully prioritised by panels of independent experts (including NHS service users). The HTA programme then commissions the research by competitive tender.

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Some HTA research projects, including TARs, may take only months, others need several years. They can cost from as little as £40,000 to over £1 million, and may involve synthesising existing evidence, undertaking a trial, or other research collecting new data to answer a research problem.

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Reports are published in the HTA journal series if (1) they have resulted from work for the HTA programme, and (2) they are of a sufficiently high scientific quality as assessed by the referees and editors.

Reviews in Health Technology Assessment are termed ‘systematic’ when the account of the search, appraisal and synthesis methods (to minimise biases and random errors) would, in theory, permit the replication of the review by others.

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The views expressed in this publication are those of the authors and not necessarily those of the HTA programme or the Department of Health.

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