Hearing-Impaired Children in the United Kingdom, IV: Cost-Effectiveness of Pediatric Cochlear Implantation

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Objective: **To estimate the cost-effectiveness of pediatric cochlear implantation by conducting a cost-utility analysis from a societal perspective.**

Design: **In a cross-sectional survey, the parents of a representative sample of hearing-impaired children assessed the health utility of their child using a revised version of the Health Utilities Index Mark III questionnaire. Linear regression was used to estimate the gain in health utility associated with implantation while controlling for eight potentially confounding variables: average (4-frequency, unaided, preoperative) hearing level (AHL), age at onset of hearing-impairment, age, gender, number of additional disabilities, parental occupational skill level, ethnicity, and parental hearing status. The gain in health utility was accumulated to estimate the number of quality-adjusted life years (QALYs) that would be gained from implantation over 15 yr and over a child's lifetime. The incremental societal cost of implantation, calculated in euros (€) at 2001/2 levels, was estimated by summing the incremental costs of implantation that are incurred in the health sector, in the education sector, and by the child's family. The cost-effectiveness of cochlear implantation was estimated by calculating the incremental societal cost per QALY gained and was compared with an upper limit of acceptability of €50,000 per QALY.**

Results: **The parents of 403 implanted children, and 1863 nonimplanted children, completed the health utility questionnaire. Higher health utility was associated with a more favorable AHL, an older age at onset of hearing impairment, female gender, having fewer additional disabilities, having parents with a greater occupational skill level, white ethnicity, and implantation. The gain in health utility associated with implantation was estimated to be higher for children with a worse preoperative AHL and who were implanted when younger. Over 15 yr, for a child implanted at age 6 with a preoperative loss of 115 dB, 2.23 QALYs were estimated to be gained, compared with a mean incremental societal cost of €57,359, yielding a mean cost per QALY of €25,629. Cost-effectiveness was more favorable: (1) when estimated over a child's lifetime rather than 15 yr, (2) for children with a worse preoperative AHL, and (3) for children who were implanted when younger.**

Conclusions: **The mean cost of gaining a QALY for the children in the present sample falls within acceptable limits. The strategy of giving highest priority for implantation to children with the greatest loss of hearing, and who are younger, maximizes benefit for a given cost.**

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Resources in health services are scarce. Accordingly, the cost-effectiveness of health care interventions should be scrutinized to enable resources to be allocated so as to maximize effectiveness (i.e., the benefit received by patients) for a given cost. The effectiveness of health-care interventions can be measured in the common metric of health utility, where more preferable states of health have higher values, with a value of zero corresponding to death and a value of one corresponding to perfect health (Drummond, O'Brien, Stoddart & Torrance, 1997; Gold, Siegel, Russell, & Weinstein, 1996). This generic approach enables the benefits of interventions for different conditions to be compared on the same scale. By accumulating the gain in health utility associated with an intervention over time, the number of quality adjusted life years (QALYs) that will be received from the intervention over that time period can be estimated (Drummond et al., 1997; Torrance & Feeny, 1989). Cost-effectiveness can then be calculated as the cost of gaining a QALY (cost/QALY). Interventions which gain QALYs at lower cost are judged to be more cost-effective than interventions which gain QALYs at higher cost.

The primary aim of the study reported in this paper was to estimate the cost-effectiveness of providing cochlear implants to children. A secondary aim was to examine differences in cost-effectiveness between different groups of candidates for implantation. Justification for this secondary aim is provided by evidence that the clinical and demographic characteristics of children influence many outcomes from implantation, including speech perception (e.g., Miyamoto, Osberger, Robbins, Myres, & Kessler, 1993; Stacey, Fortnum, Barton, & Summerfield, 2006; Tyler,

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Fryauf–Bertschy, Gantz, Kelsay, & Woodworth, 1997), spoken language, auditory performance, speech intelligibility, academic abilities (Stacey, et al., 2006), and the cost of education (Barton, Stacey, Fortnum, & Summerfield, 2006a; Schulze–Gattermann, Illg, Schoenermark, Lenarz, & Lesinski–Schiedat, 2002). It is possible therefore that the cost-effectiveness of implantation also varies according to the characteristics of the child. We focus on three characteristics which have previously been shown to exert a positive influence on the benefit associated with implantation: a younger age at implantation (Kileny, Zwolan, & Ashbaugh, 2001; Kirk, Miyamoto, Lento, Ying, O'Neill, & Fears, 2002; Miyamoto, et al., 1993; Nikolopoulos, O'Donoghue, & Archbold, 1999; Sharma, Dorman, & Spahr, 2002; Stacey, et al., 2006; Svirsky, Teoh, & Neuburger, 2004; Tyler, et al., 1997), a greater duration of implant use (Stacey, et al., 2006; Tyler, et al., 1997), and a poorer preoperative average hearing level (AHL) (Stacey, et al., 2006; Tyler, et al., 1997). If the cost-effectiveness of implantation varied with any of these variables, then it could be more cost-effective to implant some groups of children than others (e.g., younger children in preference to older children, or children with less favorable AHLs in preference to children with more favorable AHLs). Given that the number of implants funded each year is restricted both in the United Kingdom and elsewhere (Summerfield, Stacey, Roberts, Fortnum, & Barton, 2003), evidence of cost-effectiveness could guide the allocation of resources among candidates for cochlear implants. In this way, priority could be given to candidates who are expected to receive the greatest benefit for a given cost.

Several studies have used measures of health utility to assess the cost-effectiveness of pediatric cochlear implantation (Cheng, Rubin, Powe, Mellon, Francis, & Niparko, 2000; Hutton, Politi, & Seeger 1995; O'Neill, O'Donoghue, Archbold, & Normand, 2000; Summerfield & Marshall, 1995; Summerfield, Marshall, & Archbold, 1997; Wyatt, Niparko, Rothman, & De Lissovoy, 1995). All of these studies concluded that pediatric implantation was acceptably cost-effective, but none estimated how cost-effectiveness varied among candidates. We estimated this variation, and were guided by two studies of speech-perception outcomes from implantation. The first suggests that the cost-effectiveness of implantation is likely to be better when children are implanted younger (O'Neill, O'Donoghue, Archbold, Nikolopoulos, & Sach, 2002) and, the second, that cost-effectiveness improves with time after implantation (Sach, O'Neill, Whynes, Archbold, & O'Donoghue, 2003).

Only one study (Cheng et al., 2000) based the measure of effectiveness on health-utility data for children. Other studies (e.g., Hutton et al., 1995; O'Neill et al., 2000; Summerfield, & Marshall, 1995; Summerfield et al., 1997) inferred the gain in utility for children from the gain measured with adults, which may be inappropriate because the effect of implantation for prelingually deafened children may differ in kind and degree from the effect for postlingually deafened adults (Summerfield & Marshall, 1995; Cheng et al., 2000). Cheng et al. (2000) asked the parents of 78 children who had received implants in a single hospital in the United States to judge, retrospectively, what their children's health utility had been before implantation and to judge how it was currently after implantation. The gain in utility associated with implantation was estimated by subtracting the retrospective score from the current score. Three methods were used to measure health utility: (i) the Health Utilities Index Mark III (HUI3) questionnaire (Feeny, Furlong, Boyle, & Torrance, 1995; Feeny, Furlong, Torrance, Goldsmith, Zhu, De Pauw, Denton, & Boyle, 2002), (ii) a visual analogue scale (Torrance, Feeny, & Furlong, 2001), and (iii) the time trade-off (TTO) technique (Torrance, 1986). The mean changes in utility were (i) +0.39, (ii) +0.27, and (iii) +0.22. After discounting future benefits (as discussed later in this paper), the number of QALYs that would be gained from cochlear implantation over a child's lifetime was estimated to be (i) 11.59, (ii) 8.03, and (iii) 6.54. When assessed in relation to medical reimbursement costs in the United States, the cost/QALY was (i) US $5200, (ii) US $7400, and (iii) US $9000 (1999 price levels, Footnote 1).

These values were judged to be strongly competitive when compared with the cost/QALY of other health-care interventions. Using educational cost data reported by Francis, Koch, Wyatt, and Niparko (1999), Cheng et al. (2000) estimated that implantation would reduce the cost of school education by US $65,588. This saving, coupled with other costs outside the health service, including an estimated increase in future earnings of US $55,574, meant that pediatric cochlear implantation was predicted to result in a net cost saving for US society of US $53,198 per child implanted. Accordingly, it was judged that pediatric cochlear implantation was a cost-effective intervention both when assessed in the health domain, and when assessed from a broader societal perspective.

[1] The average exchange rate between the US dollar and the euro (€) was US $1.00 = €0.8851 in the calendar year 2003 (Bank of England 2004, Reference Note 1). To aid comparisons, we report costs estimated in previous studies in the currency in which they were originally reported and state the year on which cost analyses were based.

The study reported by Cheng et al. (2000) had three potential limitations. First, estimates of the gain in health utility were collected retrospectively and thus may have been subject to recall bias (Dawson, Kanim, Sra, Dorey, Goldstein, Delamarter, & Sandhu, 2002). Second, estimates were based on a small number of children (up to 78 in the health sector, and 27 in the education sector) who were implanted in the same hospital. As such, it is not clear how far the results can be generalized (Dans, Dans, Guyatt, & Richardson, 1998; Johnston, Buxton, Jones, & Fitzpatrick, 1999). In addition, the small sample size meant it was not possible to assess how the cost-effectiveness of implantation varied between children with different characteristics. Third, the health-service costs of implantation were based on reimbursement levels rather than actual costs. Reimbursement levels do not always provide a good estimate of the true cost of the resources consumed in providing heath care (Beck, Beecham, Mandalia, Griffith, Walters, Boulton, & Miller, 1999), and there is evidence that the level of public reimbursement for cochlear implantation in the United States is often below cost (Cheng & Niparko, 1999; Garber, Ridgely, Bradley & Chin, 2002).

We addressed these limitations by undertaking a cross-sectional survey in which data for a large representative sample of children were collected (Fortnum, Stacey, & Summerfield, in press). We estimated the gain in health utility associated with cochlear implantation by comparing the implanted and nonimplanted members of the sample, while controlling the influence of other variables that differ between implanted and nonimplanted children (i.e., while controlling potentially confounding influences on health utility). We accumulated the gain in health utility to estimate the number of QALYs that would be obtained from implantation. This number was assessed in relation to estimates of the incremental cost to society of providing implantation. The incremental cost is the additional cost of providing implants over and above the cost of management with acoustic hearing aids. To obtain a societal perspective, the incremental cost was estimated as the combination of components incurred in the health sector (Barton, Bloor, Marshall, & Summerfield, 2003), the education sector (Barton, et al., 2006a), and by the child's family (Barton, Fortnum, Stacey, & Summerfield, 2006b). The third component was composed of out-of-pocket expenditure and lost productivity associated with time away from normal activities by parents.

In the UK, the National Institute for Health and Clinical Excellence (NICE) takes cost-effectiveness data into account in issuing mandatory guidance on which health technologies should, and which should not, be provided in the publicly funded health sector. Initial interpretations of NICE's judgments (e.g. Raftery, 2001) concluded that interventions that gained QALYs for more than about €50,000 were unlikely to be approved. More recent interpretations have concluded that cost-effectiveness is only one of several variables that predict whether an intervention will be approved (Dakin, Devlin, & Odeyemi, 2006) and that the cost-effectiveness boundary is gradual rather than abrupt, such that the threshold beyond which interventions are unlikely to be funded is in the range from €38,000/QALY to €77,000/QALY (Devlin & Parkin, 2004). Nonetheless, for purposes of discussion it is useful to adopt categorical criteria. We infer that interventions that cost less than about €15,000 per QALY offer good value for money, that interventions that cost between €15,000 and €50,000 per QALY offer acceptable value for money, and that interventions that cost more than about €50,000 per QALY offer questionable value for money. We assessed the cost-effectiveness of cochlear implantation for different groups of children in relation to these criteria.

The cost-effectiveness of cochlear implantation was assessed over two time periods: (i) 15 yr, and (ii) the child's lifetime. In line with Barton et al. (2003), we judged that 15 yr is the minimum length of time for which implants will be used by children before being superseded by new interventions. A child's lifetime is the maximum period over which the costs and benefits of cochlear implantation could accrue, and is the period incorporated in previous estimates of the cost-effectiveness of pediatric cochlear implantation (Cheng et al., 2000; Hutton et al., 1995; O'Neill et al., 2000; Summerfield & Marshall, 1995; Summerfield et al., 1997; Wyatt et al., 1995).

METHODS

Overview

The cost-effectiveness of pediatric implantation was estimated using two values. The first was the incremental societal cost of implantation, which is the difference in cost to society between implantation and alternative interventions. The second was the gain in health utility associated with implantation compared with alternative interventions, which in turn allowed the number of QALYs gained from implantation to be estimated.

Participants and Procedures

Fortnum, Summerfield, Marshall, Davis, and Bamford (2001) ascertained the population of children in the United Kingdom with permanent bilat-

TABLE 1. Explanatory variables tabulated for each child

Variable	Values		
1. Average unaided (preoperative) hearing level (AHL)	Unaided pure-tone air-conduction thresholds in the better-hearing ear averaged across the four frequencies 0.5, 1, 2, and 4 kHz.		
2. Age at onset of hearing impairment (AONS)	(i) At birth, (ii) Between the ages of 0 and 3 yr, (iii) After 3 yr of age.		
3. Age (AGE)	Age in years on date questionnaire was returned.		
4. Gender (GEND)	(i) Male, (ii) Female.		
5. Number of additional disabilities (NDIS)	(i) None, (ii) One, (iii) Two or more.		
6. Parental occupational skill level (POSL)	Classification of the level of skill entailed in the parent's job, ranging from (lowest to highest) (i) Level 1, (ii) Level 2, (iii) Level 3, (iv) Level 4.		
7. Ethnicity (ETHN)	(i) White, (ii) Other.		
8. Parental hearing status (PHS)	(i) No hearing difficulties, (ii) At least some difficulties.		
9. Cochlear implantation (CI) (categorized according to age at implantation and duration of implant use)	Group	Age at implantation	Duration of use
	(1)	<5 yr	≥4 yr
	(2)	≥5 yr	≥4 yr
	(3)	<5 yr	≥2, <4 yr
	(4)	≥5 yr	≥2, <4 yr
	(5)	<5 yr	<2 yr
	(6)	≥5 yr	<2 yr
	(7)	nonimplanted	nonimplanted

eral hearing impairment >40 dB HL in the better hearing ear. They identified 17,160 children. We undertook a cross-sectional survey, in which the parents of a sample of 8876 of these children were invited to consent for themselves, their child's school teacher, and their child's audiologist to participate in the present study by completing postal questionnaires about their child. The sample included all children with cochlear implants (N = 993), all other profoundly impaired (AHL >95 dB) children (N = 3288), all severely impaired (AHL 71–95 dB) children (N = 3580), and a stratified random sample of approximately one in nine of the moderately impaired (AHL 41–70 dB) children (N = 1015). Subsampling the moderately impaired children was desirable for practical reasons, given the large number of children with a moderate hearing impairment. The parents' questionnaire obtained data which enabled the health utility of children to be estimated, as described below. Values of nine other variables (Table 1) with the potential to explain variation in a child's health utility were obtained from the questionnaires completed by parents, teachers, and audiologists, as described by Stacey et al. (2006).

Health Utility

Self-completion questionnaires, in which a person describes their status in different health domains, are the most commonly used method for estimating health utility scores (Brazier, Deverill, Green, Harper, & Booth, 1999). Such questionnaires are designed to obtain a description of a person's health state. A utility score is then assigned to that state, based on preferences previously elicited from members of the general public (Feeny et al., 2002).

In the present study, estimates of the health utility of children were obtained by proxy from parents with a modified version of the HUI3 questionnaire. The HUI3 questionnaire (Feeny et al., 1995; Feeny et al., 2002) measures a person's capacity to function in eight domains related to vision, hearing, speaking (and being understood), mobility, dexterity, emotion, cognition, and pain. Some questions in the standard HUI3 questionnaire are conceptually and linguistically complex. For example, in the section on pain, the response options combine three concepts: the frequency and intensity of pain and discomfort; the degree of disruption to normal activities; and the extent to which discomfort is relieved by drugs. After obtaining feedback from parents, we changed the wording of some questions so that they would be more easily understood by English speakers in the United Kingdom, and also simplified some questions by decomposing the issues which they addressed into separate questions. This revised version of the HUI3 questionnaire was embedded within the parents' questionnaire (MRC Institute of Hearing Research, Reference Note 2). The revised response options permitted a straightforward mapping onto the response options in the original questionnaire. Thus, the standard HUI3 scoring algorithm (Feeny et al., 2002) could be used to estimate the health utility of each child. The mean health utility, and associated 95% confidence interval (95% CI), was calculated for nonimplanted children with different hearing losses and for implanted children.

Gain in Health Utility • The gain in health utility associated with implantation was estimated using linear regression. This form of analysis predicts a dependent variable using a weighted sum of the values of a set of explanatory variables. Each weight

(coefficient) is an estimate of the association between the dependent variable and an explanatory variable, while controlling the strength of association between the dependent variable and each of the other explanatory variables (e.g. Strube, 2003). Where the 95% CI of a coefficient does not embrace zero, the corresponding explanatory variable makes a significant independent contribution to explaining variance in the dependent variable (i.e., is statistically significant).

We used linear regression to estimate the extent to which health utility could be predicted by the nine explanatory variables in Table 1: AHL (Footnote 2), age at onset of hearing-impairment, age, gender, number of additional disabilities, parental occupational skill level, ethnicity, and parental hearing status and cochlear implantation. The latter variable was a seven-valued factor that distinguished non-implanted children from six groups of implanted children formed by the intersection of two ages at implantation, <5 yr, and ≥5 yr, and three durations of use of implants, <2 yr, 2 to <4 yr, and ≥4yr. The coefficients for each group of implanted children provided an estimate of the gain in utility for that group. It is an assumption of linear regression analysis that continuous variables vary linearly with the dependent variable. Accordingly, before conducting the analysis, age and AHL were transformed to vary linearly with utility using the methods described by Stacey et al. (2006) (Footnote 3). Two other continuous measures, age at onset of hearing impairment and the number of additional disabilities, had skewed distributions. These measures were converted into categorical variables.

Two analyses were performed. The first was descriptive. It determined whether there was a significant association between implantation and health utility when the other explanatory variables in Table 1 were controlled. The second analysis established how the relationship between implantation and health utility differed according to the AHL of implanted children. This second analysis included an interaction term between AHL and cochlear implantation (i.e., the coefficient for AHL was allowed to differ, depending on whether children were implanted or nonimplanted) (Footnote 4). Further details of this approach, and examples of its use, are provided by Stacey et al. (2006) and Barton et al. (2006a). Analyses were performed with the SAS system (Freund & Littell, 2000).

Incremental Costs

Health Sector • Barton et al. (2003) measured the costs incurred by the UK National Health Service in providing implants to children in 12 pediatric cochlear implant programs. The incremental health sector cost of implantation was estimated by deducting an estimate of the costs that would have been entailed in providing acoustic hearing aids to the children. The analysis yielded an estimate of the average incremental cost of managing a child in each "year of care," starting with the year leading up to and including implantation. Barton et al. (2003) did not investigate whether costs vary according to age at implantation or preoperative AHL. Accordingly, the average incremental cost was assumed to apply to all implanted children. Costs, which were reported by Barton et al. (2003) at 2000/1 levels,

[2] The abbreviation, AHL, is used in this article to refer to the average of pure-tone air-conduction thresholds at the frequencies 0.5, 1, 2, and 4 kHz in the better-hearing ear. In the case of nonimplanted children, AHL refers to unaided hearing levels. In the case of implanted children, AHL refers to preoperative unaided hearing levels. In the children in the present study, AHL is related to the three-frequency pure-tone average (PTA) computed at 500 Hz, 1 kHz, and 2kHz by the equation: PTA = −0.86 + 0.98 × AHL. Thus, AHLs between 95 and 125 dB HL correspond to PTAs between 93 and 122 dB HL.

[3] The explanatory variables in Table 1 include covariates (the continuous variables, AGE and AHL) and factors (the other variables which are categorical). Utility scores did not vary systematically with AGE. Therefore, AGE was not transformed. Utility scores did vary with AHL. After exploring several functions, an exponential function was chosen to transform AHL to vary linearly with health utility:

$$AHL' = -0.38/(1 + e^{(-(AHL - 98.79)/9.32)}).$$

[4] The linear equation obtained from the first descriptive regression analysis took the form:

$$U = w_0 + w_1(\text{if GEND} = \text{"Female"}) + w_2 \times AGE + w3(\text{if ETHN} = \text{"White"}) + w4(\text{if PHS} = \text{"No hearing difficulties"}) + w_5 \times AHL' + w_{6,1}(\text{if AONS} = \text{"Between the ages of 0 and 3 yr"}) + w_{6,2}(\text{if AONS} = \text{"After 3 yr of age"}) + w_{7,1}(\text{if NDIS} = \text{"One"}) + w_{7,2}(\text{if NDIS} = \text{"Two or more"}) + w_{8,1}(\text{if POSL} = \text{"Level 4"}) + w_{8,2}(\text{if POSL} = \text{"Level 3"}) + w_{8,3}(\text{if POSL} = \text{"Level 2"}) + w_{9,1}(\text{if CI} = \text{"Group 1"}) + w_{9,2}(\text{if CI} = \text{"Group 2"}) + w_{9,3}(\text{if CI} = \text{"Group 3"}) + w_{9,4}(\text{if CI} = \text{"Group 4"}) + w_{9,5}(\text{if CI} = \text{"Group 5"}) + w_{9,6}(\text{if CI} = \text{"Group 6"})$$

where U is health utility, w_0 to w_9 are (constant) coefficients, and the strings of upper-case letters are the abbreviations for the explanatory variables defined in Table 1. Coefficients are interpreted differently depending on whether they weight a covariate or a factor. w_2 and w_5 are the differences in health utility associated with a unit change in AGE and AHL' (the transformed value of AHL). The other coefficients are the differences in health utility associated with possession of one value of a factor rather than another. Thus, for example, $w_{9,1}$ is the difference in health utility between a child with an implant in Group 1 and a child who is similar in respect of all other explanatory variables but who does not have an implant.

The linear equation obtained from the second regression analysis had the same form as the equation above, with the additional term:

$$[w_{10,1} \times AHL'(\text{if CI} = \text{"Group 1"}) + w_{10,2} \times AHL'(\text{if CI} = \text{"Group 2"}) + w_{10,3} \times AHL'(\text{if CI} = \text{"Group 3"}) + w_{10,4} \times AHL'(\text{if CI} = \text{"Group 4"}) + w_{10,5} \times AHL'(\text{if CI} = \text{"Group 5"}) + w_{10,6} \times AHL'(\text{if CI} = \text{"Group 6"})].$$

were inflated to 2001/2 levels using the Hospital and Community Health Services Pay and Prices Index (National Health Service Executive, 2004) to ensure compatibility with the cost levels for education and the family.

Education Sector • The teachers' questionnaire, used in the present study, obtained the school placement of each child and the level of support received by the child in that placement because of the child's impaired hearing. Barton et al. (2006a) combined these data with the costs of placing and supporting hearing-impaired children in different educational settings to estimate the annual cost of education of each child.

Economic Costs Incurred by the Family • The parents' questionnaire, used in the present study, obtained data on annual resources used by the family because of their child's impaired hearing in two areas: (i) out-of-pocket expenditure, and (ii) time away from usual activities by parents in accompanying children to clinic and hospital appointments. Barton et al. (submitted) summed the cost of each of these two resources to obtain an estimate of the overall economic cost incurred by each child's family.

The data for the annual education cost and overall economic cost incurred by the family were analyzed using linear regression, in the same style as the analyses of health utility, describe above. The analyses yielded estimates of the annual education cost and of the overall economic cost incurred by families associated with each variable in Table 1. The cumulative education cost associated with implantation (up to the age of 16 yr) and the cumulative economic cost incurred by the families of implanted children (up to the age of 16 yr) were also calculated.

Accumulating Utility and Costs

The number of QALYs gained from implantation was calculated by summing the gain in health utility associated with implantation in each year of care. The gain for children who had been implanted for less than 2 yr was taken as the estimate of the gain in the first year and second year after implantation. The corresponding gain for children who had been implanted for between 2 and 4 yr was taken as the estimate of the gain in the third year and fourth year after implantation. The corresponding gain for children who had been implanted for more than 4 yr was taken as the estimate of the gain in the fifth year and each subsequent year after implantation.

The incremental health sector costs of implantation were estimated by deducting the cost of alternative treatments (as estimated by Summerfield, Marshall, Barton, & Bloor, 2002) from the health sector costs of pediatric cochlear implantation (as estimated by Barton et al., 2003). The incremental costs of implantation incurred in the health sector, in the education sector (Barton et al., 2006a), and by the families of implanted children (Barton et al., 2006b) were summed to estimate the incremental societal cost of implantation. The gains in utility and incremental costs associated with implantation were assigned to the year of care in which they arose. They were then accumulated for two periods of time after implantation: (i) 15 yr and (ii) children's remaining life expectancy (Footnote 5).

Discounting • In line with recommendations for economic analyses in the United Kingdom (Treasury, Reference Note 4), future costs and benefits were discounted at 3% per annum to reflect the fact that people generally prefer to consume resources now rather than in the future but prefer to defer expenditure into the future rather than incurring it now (Drummond et al., 1997). The year leading up to, and including, implantation was enumerated as the 0th year of care. No benefits were considered to be obtained in that year. Benefits obtained in the first year of care were discounted. Thus, the number of QALYs gained from implantation was calculated as $\sum_{i=1}^{n} \frac{u}{1.03^i}$, where n is the total number of years over which benefits were accumulated and u is the difference in health utility between implanted and nonimplanted children.

Costs incurred in the 0th year of care were not discounted. Thus, the cost of implantation was calculated as $\sum_{i=0}^{n} \frac{c_i}{1.03^i}$, where c_i is the cost incurred in the i^{th} year of care. A tutorial application of these techniques to adult cochlear implantation was reported by the UK Cochlear Implant Study Group (2004).

Cost-Effectiveness Analysis

Cost-effectiveness was estimated by dividing the incremental societal cost of implantation by the number of QALYs gained from implantation to obtain the incremental (societal) cost per QALY gained.

[5] Life-expectancy data were obtained from the UK Government Actuary's Department (Government Actuary Department 2004, Reference Note 3). Average levels of life expectancy, across boys and girls, were calculated for a child implanted at age 3 yr and a child implanted at age 6 yr. On average, both groups of children were expected to live to age 79 yr, hence the average life expectancy of the two groups was estimated to be 76 yr and 73 yr, respectively (boys are estimated to have a life-expectancy 2 yr below the average and girls 2 yr above the average). The life-expectancy of children with cochlear implants does not differ significantly from the life-expectancy of the age- and gender-matched population (Summerfield & Marshall, 2001; Summerfield, Cirstea, Roberts, Barton, Graham, & O'Donoghue, 2005).

TABLE 2. Estimated levels of health utility for five groups of hearing-impaired children

	Group				
	Moderate (AHL 40–70 dB)	Severe (AHL 71–95 dB)	Profound (AHL 96–105 dB)	Profound (AHL >105 dB)	Implanted
HUI3 score	0.677	0.616	0.497	0.353	0.575
(95% CI)	(0.652–0.702)	(0.598–0.634)	(0.469–0.525)	(0.327–0.379)	(0.553–0.598)
N	260	464	259	290	403

Analyses from Alternative Perspectives • To determine whether conclusions were sensitive to the perspective of the analysis, we compared results obtained from the societal perspective with results obtained from the perspectives of the health sector alone, and the health and education sectors together. The second analysis also allowed comparisons with results reported by O'Neill et al. (2000) and Schulze-Gattermann, et al. (2002).

Orientation of the Analyses

The modal age at implantation of children in the sample was 3 yr, and the mean age at implantation was 6 yr (Footnote 5). The average AHL of children with implants was 115 dB (Stacey et al., 2006), with 93% of the sample (395 out of the 425 for whom AHL data were available) having an AHL between 100 dB and 130 dB (inclusive). To cover informative ranges of AHL and age at implantation, we estimated the costs, benefits, and cost-utility ratios associated with implantation for children defined by the combination of three AHLs (105 dB, 115 dB, and 125 dB), with two ages at implantation (3 and 6 yr).

RESULTS

Response Rate

Consent to participate was received from the parents of 3274 children (37% of those invited to participate). Questionnaires were returned by the parents of 2858 of these children (88% of those who consented to participate). The parents of 2266 children (69% of those who consented to participate), 403 of whom had an implant, completed all sections of the revised HUI3 questionnaire. Steps taken to confirm the representativeness of the responding sample were reported by Fortnum et al. (in press).

Health Utility

Average levels of health utility, without adjustment for the effects of other variables, were lower for children with less favorable AHLs (Table 2). The average health utility of children with implants was between that of severely and profoundly impaired nonimplanted children.

Gain in Health Utility • Higher health utility was associated with a more favorable AHL, an older age at the onset of hearing impairment, female gender, having fewer additional disabilities, having parents with a greater occupational skill level and white ethnicity (Table 3). In addition, implantation was associated with a significant gain in health utility for five of the six groups of implanted children. The nonsignificant gain in health utility (for group 5) may be explained by the fact that this group was composed of the smallest number of children ($N = 25$ out of 278 implanted children). The largest gains were shown by children who were implanted before the age of 5 yr who had used their implants for more than 4 yr (Table 3).

When an interaction term between cochlear implantation and preoperative AHL was added to the

TABLE 3. Coefficient values and 95% confidence intervals of the variables used to predict health utility

Variable	Coefficient value (95% CI)
Constant	0.622 (0.563 to 0.680)‡
Transformed average hearing level:	1.001 (0.910 to 1.093)‡
Age at onset of hearing impairment:	
≥3 yr vs birth	0.068 (0.025 to 0.112)†
0 to 3 yr vs birth	0.039 (0.012 to 0.066)†
Age:	
1 yr older	0.002 (–0.001 to 0.005)
Gender:	
Female vs male	0.025 (0.006 to 0.045)*
Disabilities:	
Two plus vs none	–0.281 (–0.312 to –0.250)‡
One vs none	–0.086 (–0.112 to –0.059)‡
POSL:	
4 (highest) vs 1 (lowest)	0.049 (0.014 to 0.084)†
3 vs 1	0.026 (–0.006 to 0.058)
2 vs 1	0.034 (0.000 to 0.067)*
Ethnicity:	
White vs other	0.034 (0.001 to 0.068)*
Parental Hearing:	
No problems vs some problems	0.007 (–0.024 to 0.038)
Cochlear implantation:	
Group 1 vs no implant	0.232 (0.184 to 0.280)‡
Group 2 vs no implant	0.183 (0.126 to 0.239)‡
Group 3 vs no implant	0.212 (0.161 to 0.263)‡
Group 4 vs no implant	0.172 (0.103 to 0.240)‡
Group 5 vs no implant	0.066 (–0.013 to 0.144)
Group 6 vs no implant	0.130 (0.053 to 0.206)‡

Adjusted $r^2 = 0.38$; F-ratio = 47.58 ($p < 0.001$), 1490 children (278 of which had implants) were included in the analysis ($p < 0.05$, † $p < 0.01$, ‡ $p < 0.001$).*

TABLE 4. Gains in health utility and in QALYs associated with cochlear implantation as a function of preoperative AHL, age at implantation, and the time period over which gains are accumulated

Pre-operative AHL(dB)	Time period	Variable	Gain in variable for children implanted at age 3 yr (95% CI)	Gain in variable for children implanted at age 6 yr (95% CI)
105	<2 yr use	Health utility	0.066 (–0.012 to 0.144)	0.115 (0.212 to 0.208)*
	2–4 yr use	Health utility	0.174 (0.113 to 0.234)‡	0.172 (0.076 to 0.269)‡
	>4 yr use	Health utility	0.171 (0.109 to 0.223)‡	0.138 (0.046 to 0.229)†
	15 yr total	QALYs	1.843	1.661
	Lifetime total	QALYs	4.894	4.073
115	<2 yr use	Health utility	0.125 (0.029 to 0.221)*	0.156 (0.068 to 0.244)†
	2–4 yr use	Health utility	0.249 (0.194 to 0.304)‡	0.185 (0.113 to 0.234)‡
	>4 yr use	Health utility	0.256 (0.207 to 0.305)‡	0.196 (0.139 to 0.252)‡
	15 yr total	QALYs	2.791	2.238
	Lifetime total	QALYs	7.363	5.668
125	<2 yr use	Health utility	0.153 (0.037 to 0.270)†	0.176 (0.056 to 0.295)†
	2–4 yr use	Health utility	0.286 (0.216 to 0.355)‡	0.191 (0.097 to 0.284)‡
	>4 yr use	Health utility	0.297 (0.238 to 0.357)‡	0.224 (0.153 to 0.294)‡
	15 yr total	QALYs	3.254	2.520
	Lifetime total	QALYs	8.569	6.447

*Significant gains in health utility are denoted by * $p < 0.05$, † $p < 0.01$, ‡ $p < 0.001$.*

analysis, implantation was associated with a significantly larger gain in health utility for children with a worse preoperative AHL (Table 4). For example, after adjusting for other variables, the mean gain in health utility (compared with non-implanted children) for a child implanted at age 3 yr, for more than four years, with a preoperative loss of 125 dB was estimated to be 0.297, and 0.171 for a corresponding child with a preoperative loss of 105 dB. This result arose because health utility declined with worsening AHL among children without cochlear implants but was unrelated to AHL among implanted children.

Gain in QALYs

Similar variation according to preoperative AHL, age at implantation, and duration of implant use was found when these gains in health utility were accumulated over time to estimate both the 15-yr and lifetime QALY gains associated with implantation. Because the gain in QALYs associated with implantation was derived from the gain in health utility, larger gains in QALYs were associated with a worse preoperative AHL (e.g., 3.254 QALYs at 125 dB, compared with 1.843 QALYs at 105 dB, for a child implanted at age 3 yr, over a period of 15 yr), a younger age at implantation, and a longer duration of implant use (Table 4).

Incremental Costs

Health Sector • Over 15 yr it was estimated that €1908 would be averted, on average, due to reduced provision of acoustic hearing aids. The mean incremental health sector cost of implantation was thereby estimated to be €79,263 over a period of 15 yr (Table 5). Over a child's lifetime, incremental health sector costs were estimated to be €132,040 for a child implanted at age 3 yr and €131,292 for a child implanted at age 6 yr (Table 5).

Education Sector • Barton et al. (2006a) estimated that implantation is associated with a negative incremental cost (i.e., a cost-saving) in education for children with preoperative AHLs that exceed 98 dB. Cost-savings were estimated to be larger for children with worse preoperative AHLs. The mean cumulative education cost-saving for a child with a preoperative loss of 115 dB was estimated to be €33,022 after implantation at age 3 yr, and €22,853 after implantation at age 6 yr (Table 5).

Economic Costs Incurred by the Family • Barton et al. (2006b) estimated that implantation is associated with an increase in the overall economic cost incurred by the family but that these costs did not vary according to preoperative AHL. The mean cumulative economic cost was estimated to be €3355 for a child implanted at age 3 yr, and €949 for a child implanted at age 6 yr (Table 5).

Societal Costs • When the health sector costs, cost-savings in education, and economic costs incurred by the family were combined, lifetime incremental societal costs were estimated to range between €92,525 (for a child with a preoperative loss of 125 dB, implanted at age 3 yr) and €119,591 (for a child with a preoperative loss of 105 dB, implanted at age 3 yr) (Table 5).

Cost-Effectiveness Analysis

The incremental societal cost of gaining a QALY for a child with the average AHL (115 dB) implanted at the average age of 6 yr was estimated to be €25,629, when costs and benefits are accumulated

TABLE 5. Costs associated with pediatric cochlear implantation

AHL (dB)	Cost sector	Time period	Implanted at age 3 yr	Implanted at age 6 yr
105	Health	15 yr	€79,263	€79,263
	Health	Lifetime	€132,040	€131,292
	Education	15 yr	–€15,804	–€12,676
	Education	Lifetime	–€15,804	–€12,676
	Family	15 yr	€3,355	€949
	Family	Lifetime	€3,355	€949
	Societal	15 yr	€66,814	€67,536
	Societal	Lifetime	€119,591	€119,565
115	Health	15 yr	€79,263	€79,263
	Health	Lifetime	€132,040	€131,292
	Education	15 yr	–€33,022	–€22,853
	Education	Lifetime	–€33,022	–€22,853
	Family	15 yr	€3,355	€949
	Family	Lifetime	€3,355	€949
	Societal	15 yr	€49,596	€57,359
	Societal	Lifetime	€102,373	€109,388
125	Health	15 yr	€79,263	€79,263
	Health	Lifetime	€132,040	€131,292
	Education	15 yr	–€42,870	–€29,669
	Education	Lifetime	–€42,870	–€29,669
	Family	15 yr	€3,355	€949
	Family	Lifetime	€3,355	€949
	Societal	15 yr	€39,748	€50,543
	Societal	Lifetime	€92,525	€102,572

Positive entries are costs incurred. Negative entries are costs averted.

over 15 yr. This figure was calculated by dividing the estimated incremental societal cost (€57,359) in Table 5 by the incremental QALY gain (2.238) in Table 4. The incremental cost of gaining a QALY is lower the worse the AHL, the younger the age at implantation, and the longer the period of time over which costs and benefits are accumulated (Table 6). The dependency on AHL arises because implanted children with worse AHLs gain more utility and achieve greater cost-savings in education, relative to non-implanted children, than implanted children with better AHLs do. The dependency on age at implantation arises because the younger the age at implantation, the greater the estimated QALY gain associated with implantation, and the greater the number of years over which cost-savings in education can be realized. The dependency on duration of use arises because the QALY gain associated with implantation increases with duration of implant use, whereas the annual costs associated with implantation fall. Together, these effects result in cost/QALY estimates which vary by nearly a factor of four from €10,798/QALY for a lifetime of use by a child with an AHL of 125 dB implanted at age 3 to €40, 660/QALY for 15 yr of use by a child with an AHL of 105 dB implanted at age 6.

TABLE 6. Estimates of the societal cost per QALY associated with pediatric cochlear implantation

AHL (dB)	Time period	Implanted at age 3 yr	Implanted at age 6 yr
105	15 yr	€36,253	€40,660
	Lifetime	€24,436	€29,355
115	15 yr	€17,770	€25,629
	Lifetime	**€13,904**	€19,299
125	15 yr	**€12,215**	€20,057
	Lifetime	**€10,798**	€15,910

Entries in bold are within the inferred limit for good value for money of €15,000 per QALY; all other entries are within inferred acceptable limits of cost-effectiveness (between €15,000 and €50,000 per QALY).

Analyses from Alternative Perspectives

Our main conclusions—that implantation was acceptably cost-effective, and more favorable when estimated over a child's lifetime, for children with a worse preoperative AHL, and for children who were implanted when younger—are not affected by the perspective of the analysis. The same conclusions are reached when cost-effectiveness is assessed from the perspective of the health sector alone (Table 7) or from the combined perspectives of the health and education sectors (Table 8). These comparisons demonstrate that the incremental health sector costs and QALY gains are the main determinants of the cost-effectiveness of pediatric implantation in the United Kingdom.

DISCUSSION

The health utility of hearing-impaired children, estimated with a modified version of the HUI3

TABLE 7. Estimates of the cost per QALY associated with pediatric cochlear implantation from the perspective of the health sector

AHL (dB)	Time period	Implanted at age 3 yr	Implanted at age 6 yr
105	15 yr	€43,008	€47,723
	Lifetime	€26,982	€32,235
115	15 yr	€28,399	€35,413
	Lifetime	€17,933	€23,164
125	15 yr	€24,358	€31,452
	Lifetime	€15,410	€20,366

All entries are within inferred acceptable limits of cost-effectiveness (between €15,000 and €50,000 per QALY).

questionnaire, varies systematically with variables related to the child (average hearing level, age at onset of hearing impairment, gender, and number of additional disabilities) and the family (parental occupational skill level and ethnicity). After adjusting for the effects of these variables, cochlear implantation is associated with a significant gain in health utility. When estimates of the QALY gain associated with implantation are compared with estimates of the incremental societal cost of implantation, it was estimated that implantation was acceptably cost-effective, and more favorable when estimated over a child's lifetime, for children with a worse preoperative AHL, and for children who were implanted when younger. These findings are consistent with the benefits of cochlear implantation that have been found in the domains of speech perception, everyday communication, and educational attainment, for the same sample of children (Stacey et al., 2006).

Comparisons with Other Studies

Health Utility • The gain in health utility associated with implantation was estimated to be higher for children implanted younger, with a worse preoperative AHL, and to increase (though at a marginally decreasing rate) with duration of implant use (3 and 4). For children with the mean AHL of the sample (115 dB), implantation was associated with a mean gain in health utility which varied from +0.13 to +0.26, depending on the age of the child at implantation and the duration of implant use. These values straddle the gain of +0.20, which was measured, also using the HUI3 questionnaire, in two studies of adults (Palmer, Niparko, Wyatt, Rothman, & de Lissovoy, 1999; UK Cochlear Implant Study Group, 2004), and which has been adopted as the starting point in some studies of children (Hutton et al., 1995; O'Neill et al., 2000; Summerfield & Marshall, 1995; Summerfield et al., 1997).

Our estimates are lower, however, than the gain of +0.39 estimated by Cheng et al. (2000) when parents completed the HUI3 questionnaire retrospectively for their children. Possibly, Cheng et al. (2000) recorded a higher estimate because of recall bias associated with retrospective data collection (Dawson et al., 2002). In a meta-analysis of seven studies which estimated the gain in quality of life associated with implantation for adults (Cheng & Niparko, 1999), the four largest gains came from the four studies which used retrospective methods. Additionally, our revisions to the questionnaire may also account for part of the difference. An alternative interpretation is that parents who have witnessed a change in their child's health utility may be sensitized to the states described in the HUI3 questionnaire and may make more accurate judgments as a result.

Societal Costs • Three previous studies (Cheng et al., 2000; O'Neill et al., 2000; Schulze–Gattermann et al., 2002) have estimated the incremental cost of pediatric cochlear implantation in domains outside the health sector. From the viewpoint of the health and education sectors together, the incremental cost has been estimated to be €55,133 (exchange rate US $1.20 = €1.00) in the United Kingdom (O'Neill et al., 2000; 1997/8 prices, 6% discount rate), between €46,000 and €57,000 in Germany (up to age 16 yr) (Schulze–Gattermann et al., 2002; 1999 prices, 6% discount rate), and equivalent to a cost saving of US $5,330 (€4,442) in the United States (Cheng et al., 2000; 1999 prices, 3% discount rate). These estimates are lower than our summation of the lifetime incremental costs in the health and education sectors, which range between €89,170 (for a child implanted at age 3, with a preoperative loss of 125 dB) and €118,616 (for a child implanted at age 6, with a preoperative loss of 105 dB) (Table 5). Part of this difference can be explained by the fact that we used more recent price levels and a lower discount rate (2001/2 and 3%) than O'Neill et al. (2000) (1997/8 and 6%) and Schulze-Gattermann et al. (2002) (1999 and 6%). Additionally, Cheng et al. (2000) and O'Neill et al. (2000) based their estimates of hospital

TABLE 8. Estimates of the cost per QALY associated with pediatric cochlear implantation from the perspectives of the health and education sectors combined

AHL (dB)	Time period	Implanted at age 3 yr	Implanted at age 6 yr
105	15 yr	€34,433	€40,091
	Lifetime	€23,752	€29,123
115	15 yr	€16,568	€25,203
	Lifetime	**€13,448**	€19,132
125	15 yr	**€11,184**	€19,679
	Lifetime	**€10,407**	€15,764

Entries in bold are within the inferred limit for good value for money of €15,000 per QALY, all other entries are within inferred acceptable limits of cost-effectiveness (between €15,000 and €50,000 per QALY).

costs on medical reimbursement levels, which have been argued to underestimate the true hospital costs of implantation (Cheng & Niparko, 1999; Cheng et al., 2000; Garber, et al., 2002; Summerfield et al., 2003). Additionally, Barton et al. (2006a) demonstrated that our estimates of the educational cost-savings associated with implantation may be smaller than those reported by Cheng et al. (2000) and O'Neill et al. (2000) because we controlled for more confounding variables.

Cost-Effectiveness • We estimate that the incremental life-time *health sector* cost/QALY of providing implants to children ranges from €15,410 (for a child implanted at age 3, with a preoperative loss of 125 dB) to €47,723 (for a child implanted at age 6, with a preoperative loss of 105 dB). The only other study which has based estimates of the cost-effectiveness of pediatric cochlear implantation on measures of health utility for children is the US study reported by Cheng et al. (2000). Their estimates of the cost/QALY were lower than ours and ranged between US$5,197 (€4331) and US$9,209 (€7674) (1999 prices). The difference may be explained, in part, by the factors discussed in the previous paragraph.

In contrast, our results agree more closely with other UK estimates (Summerfield et al., 1997; O'Neill et al., 2000), provided that analyses conducted in earlier years are brought into alignment by inflating costs to the same year (2001/2) and adopting the same discount rate (3% *per annum*). When we adopt those parameters, we estimate that implantation of a child with a preoperative loss of 115 dB achieves a lifetime health sector cost/QALY of between €17,933 (for implantation at age 3) and €23,164 (for implantation at age 6), compared with €27,745 (Summerfield et al., 1997) and €26,314 (O'Neill et al., 2000).

Limitations

One limitation of the present study is that our estimates of costs and benefits were derived from regression analyses of cross-sectional data, rather than from prospective data for individual children who had been randomized either to receive, or not receive, an implant. Given the challenge of obtaining parental consent to randomization and compliance with randomization, we chose not to undertake a randomized study (Summerfield, 2002). As a result, we cannot be certain that we controlled every variable that influenced whether or not a child had received an implant. Also, our estimates of the gain in utility at different times after implantation are based on the gains displayed by different groups of children at one point in time, rather than the gains displayed by a single group of children at successive points in time. Children implanted at different points in time may receive different implant devices and different care from implant programs, or differ in other ways from children implanted at other time points. Though we controlled many characteristics of the child and family, we did not control all of these variables.

A second limitation arises in our estimation of health utility. Our estimates were obtained by parental proxy, and with a nonstandard version of the HUI3 questionnaire. Our resulting estimate of the gain in health utility associated with implantation is smaller than that of the one study that used the standard HUI3 questionnaire (Cheng et al., 2000), also by parental proxy, albeit with a retrospective design without controls. In our study only parents were asked to estimate the health utility of their child, other measures (e.g., quality of life and educational attainment) were however estimated both by parents and by teachers. Stacey et al. (2006) have shown that there was broad agreement between the estimates of parents and teachers on such measures, thereby suggesting that the method of obtaining health utility estimates by parental proxy is a reliable one.

A third limitation is that we did not assess the impact of all relevant variables on the cost-effectiveness of implantation. Our results are based largely on children who were implanted before the year 2000, and we therefore did not assess, as other papers have (O'Neill et al. 2002; Sach et al., 2003), whether the cost-effectiveness of implantation has improved over time. Similarly, we did not investigate whether the incremental health sector costs of implantation varied according to clinical and demographic characteristics of children. Were they found to do so then this would, in turn, further influence estimates of the cost-effectiveness of implantation. In addition, only 7% of the children in our sample had a preoperative loss that was less than 100 dB (29 out of the 425 children for whom a value of AHL was available), and only 14% were implanted before the age of 3 yr. As a result, we cannot estimate the cost-effectiveness of implantation for children with a preoperative loss of ≤100 dB, nor can we assess the health-economic implications of the recommendation that congenitally hearing-impaired children should, where possible, be implanted before the age of 2 yr (Francis & Niparko, 2003). Trends in our data do suggest however that the results could be less favorable for children with a preoperative loss of ≤100 dB, and more favorable for children implanted before the age of 2 yr. These limitations, coupled with a critical mass of studies which have concluded that pediatric implantation is acceptably cost-effec-

tive (e.g., Cheng et al., 2000; O'Neill et al., 2000; Summerfield et al., 1997), mean that future studies should concentrate on the current point of clinical equipoise by recruiting children who use state-of-the-art implant systems, with AHLs ≤100 dB, and who are implanted before the age of 2 yr.

A fourth limitation is that we have defined candidacy in the unidimensional metric of average unaided preoperative hearing level, whereas the metric used in clinical practice is multidimensional and also includes the ability, or potential, to benefit from acoustic hearing aids. Therefore, the conclusion that estimates of the cost-effectiveness of pediatric cochlear implantation were more favorable for children with a worse preoperative AHL should be treated only as a broad indication of a criterion of candidacy. Failure to take account of other potentially explanatory variables may be why only 38% of the variation in health utility could be explained within our analysis (see Table 5). The same explanatory variables were able to explain 70% of the variation in children's academic abilities (Stacey et al., 2006).

A final limitation stems from the desirability of basing estimates of cost-effectiveness on measures of social preference obtained from the same general population as the one from which patients are drawn (NICE, 2004). The use of the HUI3 in the present study violates this principle. The HUI3 measures the utility of states of health based on preferences expressed by a sample of the population of Ontario, Canada. We applied their preferences to children treated in the United Kingdom. Thus, pending a UK valuation of the health states in the HUI3 (c.f. McCabe, Stevens, Roberts, & Brazier, 2005), we cannot judge whether the absolute values of our estimates of cost-effectiveness are appropriate for the United Kingdom. We can be more confident in the relative values of our estimates, however, because a re-evaluation of the HUI3 for the United Kingdom would be more likely to cause the cost-effectiveness of different implant groups to shift in the same direction, rather than to move relative to one another. More weight can be given, therefore, to the conclusion that implantation is most cost-effective when implants are provided to children with a worse AHL at the youngest age, than to the idea that implantation represents good or poor value for money for any particular group of candidates.

To set against these limitations, the design of the study allowed adjustment for effects of many confounding variables when estimating the association between implantation and health utility. Moreover, a relatively large proportion of the variation in health utility could be explained, compared with that achieved when undertaking similar analyses with two components of the societal cost of implantation: costs of education (Barton et al., 2006a) and the economic costs incurred by the family (Barton et al., 2006b). The credibility of the results is further bolstered by the fact that cost-effectiveness estimates are based on data from a representative sample of UK population of hearing-impaired children.

CONCLUSIONS

The provision of cochlear implants to children has been controversial because of high costs, uncertain outcomes, and doubts about the ethics of performing surgery on a healthy child. This article, and three companion articles (Stacey et al., 2006; Barton et al., 2006a; 2006b), have addressed the first two concerns in controlled comparisons of implanted and nonimplanted children. We have shown that implantation is associated with significant improvements in spoken communication skills and in some aspects of educational achievements and quality of life, provided that children receive implants before the age of 5 yr (Stacey et al., 2006). These benefits are accompanied by reduced costs of education (Barton et al., 2006a), but with modestly increased costs incurred by families (Barton et al., 2006b). The present paper adds to these findings by showing that the health utility benefits of providing implantation to deaf children are significant. In addition, the benefits are estimated to be large enough to have justified the costs, and to be greatest for children with a greater preoperative loss of hearing and for children who are younger.

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