

Hearing-Impaired Children in the United Kingdom, III: Cochlear Implantation and the Economic Costs Incurred by Families

Garry R. Barton, Heather M. Fortnum, Paula C. Stacey, and A. Quentin Summerfield

Objectives: This article addresses two questions. First, are there differences in the economic costs incurred by families of hearing-impaired children depending on whether or not children have cochlear implants? Second, are these differences important when assessed from the perspective of society?

Methods: In a cross-sectional survey, parents of a representative sample of hearing-impaired children provided data about annual resources used by the family because of their child's hearing impairment. The data yielded estimates of two variables: out-of-pocket expenditure and time away from normal activities by parents. The economic cost of the two variables was estimated in euros (€) at 2001/2 price levels, and summed to estimate the overall economic cost incurred by the family. Linear regression was used to estimate the association between costs and implantation, while controlling for average (unaided, preoperative) hearing level, age at onset of hearing impairment, age, gender, the number of additional disabilities, parental occupational skill level, ethnicity, and parental hearing status. The cumulative economic cost incurred by the families of implanted children, between implantation and age 16 yr, was calculated from estimates of the overall economic cost associated with implantation and compared with estimates of the incremental health-sector cost of implantation.

Results: Data were provided by the families of 2858 children, 468 of whom had received a cochlear implant. Compared with the families of nonimplanted children, out-of-pocket expenditure was estimated to be significantly higher for families when children were implanted before the age of 5 yr and had used their implant for less than 2 yr, as was lost productivity when children had used their implants for less than 2 yr. Overall economic cost was estimated to be significantly higher for the families of implanted children who had used their implants for less than 2 yr. The cumulative economic cost was estimated to be €3355 for a family whose child was

implanted at age 3 yr and €949 for a child implanted at age 6 yr. These costs correspond, at most, to 3% of the incremental health-sector costs of implantation.

Conclusions: Compared with families of nonimplanted children, families of implanted children incur additional costs in the 2 yr after implantation. These costs are small in relation to the health-sector costs of providing implantation.

(*Ear & Hearing* 2006;27:563–574)

Several studies have compared the costs of pediatric cochlear implantation with the costs of alternative treatments for profound hearing impairment. Those studies have shown that implantation is associated with increased costs in the health sector (e.g., Barton, Bloor, Marshall, & Summerfield, 2003; Barton, Bloor, Marshall, & Summerfield, 2004; Carter & Hailey, 1999; Cheng, Rubin, Powe, Mellon, Francis, & Niparko, 2000; O'Neill, O'Donoghue, Archbold, & Normand, 2000; Severens, Brokx, & van den Brock, 1997; Summerfield & Marshall, 1995), but with reduced costs in the education sector (Barton, Stacey, Fortnum, & Summerfield, 2006a; Francis, Koch, Wyatt, & Niparko, 1999; O'Neill et al., 2000; Schulze-Gattermann, Illg, Schoenermark, Lenarz, & Lesinski-Schiedat, 2002). The present study investigated the relation between implantation and the economic costs incurred by the families of hearing-impaired children, a potentially important area in which less research has been conducted.

In the United Kingdom, hearing-impaired children receive audiological, surgical, and rehabilitative services from hospitals and clinics in the National Health Service. Within this system, children can receive acoustic hearing aids and cochlear implants. Provision can include assessment, treatment, rehabilitation, and long-term maintenance of devices. These health sector services are supported out of general taxation and are provided at no cost to the families of children. Families, however, must meet the incidental costs of attending hospital/clinic appointments and other costs that arise from having a hearing-impaired child. We use the phrase "costs incurred by families" to refer to the economic value of these incidental resources. Our aims were to identify and measure such resources,

MRC Institute of Hearing Research, University Park, Nottingham, UK (G.R.B., P.C.S., H.M.F., A.Q.S.). Now at the School of Economics, University of Nottingham, Nottingham, UK (G.R.B.); the Department of Psychology, University of York, Heslington, York, UK (P.C.S., A.Q.S.); and the Trent Research and Development Support Unit, University of Nottingham, Nottingham, UK (H.M.F.).

to assign a monetary value (cost) to them, and to isolate the component of cost that is associated with implantation.

COMPONENTS OF ECONOMIC COSTS INCURRED BY THE FAMILY

We considered two main cost components: (i) out-of-pocket expenditure incurred directly by families, for example, in attending hospital/clinic appointments; and (ii) lost productivity, which arose from parents taking time away from work and from leisure activities (See Footnote 1). The level of government benefits received by families was also estimated (Footnote 2). The term "government benefits" refers to the financial support provided by the government to families who qualify for assistance. In the United Kingdom, these benefits are funded through general taxation. Economists classify them as "transfer payments" because they constitute a transfer from one party in society to another for which no economic service is provided in return (Begg, Fischer, & Dornbusch, 1994). Accordingly, it is sometimes argued that government benefits should be excluded from economic evaluations (Brouwer, Rutten, & Koopmanschap, 2001; Drummond, O'Brien, Stoddart & Torrance, 1997), and for this reason we do not include them in our estimate of the economic costs incurred by the family. However, from the government perspective, benefits entail an opportunity cost because the money could be used in other ways (Palmer & Raftery, 1999), and cochlear implantation may change the level of government benefits provided to families. For those reasons, we assess whether there is an association between the level of government benefits and cochlear implantation.

AIMS OF THE ANALYSES

Our first aim was to test the hypothesis that implantation lowers family expenditure by reducing the need for special equipment and special services (Cheng et al., 2000; Hutton, Politi, & Seeger, 1995; Summerfield, Marshall, & Archbold, 1997). Special

equipment includes devices such as vibrating pillow alarms, flashing lights that supplement door-bells and telephone signals, and televisions with teletext. Special services include sign-language translation, lip-speaking (where someone accurately repeats a speaker's message to a lip-reader), and video subtitling. The hypothesis that implantation reduces these costs is a particular instance of the more general hypothesis that the health sector costs of pediatric cochlear implantation are partly offset by savings in other domains.

Our second aim was to estimate costs in three areas that are directly relevant to the families of hearing-impaired children: (i) expenditure that families elect to incur, such as the cost of special equipment and special services; (ii) the cost of resources that families are required to incur, such as the cost of time away from normal activities to accompany a child to a hospital or clinic; and (iii) the financial support that families may receive from government because they include a hearing-impaired child. In these three respects, it may be useful for families of children recently diagnosed as hearing-impaired to learn about the past practice and experience of other similar families.

Our third aim was to estimate the cost to society of the resources used by families as a result of the family including a hearing-impaired child. Economists are urged to adopt a societal perspective, in which costs and effects in all domains are taken into account, when estimating the cost-effectiveness of interventions in health care (Drummond et al., 1997; Gold, Siegel, Russell, & Weinstein, 1996). A societal perspective is particularly relevant where interventions entail repeated or extended visits to hospitals or clinics, requiring patients or their caregivers to take time away from their usual activities. Pediatric implantation is an example of such an intervention. Judgments of the cost-effectiveness of some interventions hinge on whether economic costs incurred by patients and their families are taken into account (Greenberg, Peiser, Peterburg, & Pliskin, 2001; Moore, Speroff, Grogan, Poulouse, & Holzman, 2005). In a separate analysis (Barton, Stacey, Fortnum, and Summerfield, 2006b), we combined the economic costs incurred by the family, as estimated in this paper, with estimates of the incremental health-sector costs of implantation and of the change in the costs of compulsory education (Barton et al., 2006a), to estimate the incremental cost-effectiveness of pediatric cochlear implantation from a societal perspective.

A further reason for adopting a societal perspective is that results are more likely to generalize across countries. For example, in the United Kingdom, the cost of special equipment and special ser-

¹We use the term "parent" to refer to a child's parent(s) and/or their caregiver(s).

²In the United Kingdom, the main government benefit to which families of hearing-impaired children may be entitled is the Disability Living Allowance. Families qualify for this benefit if their child is either aged 3 mo or over and needs extra help or care, or is aged 3 yr or over and has severe difficulty walking, or is aged 5 yr or over and needs extra help getting around out of doors (UK Department for Work and Pensions, Reference Note 1). Each family is entitled to make one claim for Disability Living Allowance per child, with the level of award ranging between €23 and €88 per week. Entitlement is not dependent on family income.

TABLE 1. Variables tabulated for the child in each family

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	(i) At birth, (ii) Between the ages of 0 and 3 yr, (iii) After 3 yr of age		
3. Age	Age in years on date questionnaire was returned		
4. Gender	(i) Male, (ii) Female		
5. Number of additional disabilities	(i) None, (ii) One, (iii) Two or more		
6. Parental occupational skill level	Classification of the skill level in the parent's job, lowest to highest: (i) Level 1, (ii) Level 2, . . . , (iv) Level 4 (Office for National Statistics, 2000)		
7. Ethnicity	(i) White, (ii) Other (non-white)		
8. Parental hearing status	(i) No hearing difficulties, (ii) At least some difficulties		
9. 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

vices is often borne by families themselves, whereas in other countries it may be funded by other agencies, such as insurance companies in the United States. Thus, it is more informative to compare costs incurred by families between countries when those costs are assessed from the perspective of society.

PREVIOUS STUDIES

Two previous studies have estimated costs incurred by the families of implanted children (Cheng et al., 2000; Sach & Whynes, 2003). In the United States, Cheng et al. (2000) estimated the cost-effectiveness of pediatric cochlear implantation and, in so doing, estimated the changes associated with implantation in four variables: the cost of time off work by parents (lost productivity), costs of travel and parking incurred in attending hospital/clinic appointments, the future earnings of the child, and the cost of special equipment. Estimates were based on informed assumptions by the authors about resources used by the families of implanted children.

Sach and Whynes (2003) used the example of pediatric cochlear implantation to demonstrate how different assumptions about the value assigned to non-health sector resources, such as time away from usual activities, can result in radically different estimates of the non-health sector costs associated with an intervention in health care. In so doing, they collected data on parental time away from work and leisure activities to attend hospital/clinic appointments, and on associated out-of-pocket expenses. Data were obtained in face-to-face interviews with the families of children who had received implants in one pediatric cochlear implant center in the United Kingdom. No comparisons were made

between implanted children and nonimplanted control subjects.

EXTENDING PREVIOUS RESEARCH

We extended the two previous studies by comparing the families of implanted children with the families of nonimplanted children to estimate the differences in the economic costs incurred by families that are associated with implantation. We made the comparison while controlling the first eight variables listed in Table 1: average (unaided, preoperative) hearing level (AHL) (Footnote 3), age at onset of hearing impairment, age, gender, the number of additional disabilities, parental occupational skill level, ethnicity, and parental hearing status. It was important to control these potentially confounding variables because children with implants in the United Kingdom have a later average age at the onset of hearing impairment, are less likely to suffer from disabilities in addition to their hearing impairment, and come from more affluent families, than the population of nonimplanted children (Fortnum, Marshall, & Summerfield, 2002). If these variables are not controlled, there is a risk of attributing effects to implantation that should more properly be

³The abbreviation AHL is used in Table 1 and elsewhere 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 2 kHz by the equation: $PTA = -0.86 + 0.98 \times AHL$. Thus, AHLs between 95 and 125 dB HL correspond to PTAs between 93 and 122 dB HL.

attributed to other variables. Additionally, because outcomes from implantation vary according to age at implantation (Baumgartner, Pok, Egelierler, Franz, Gstoettner, & Hamazavi, 2002), duration of implant use (Fryauf-Bertschy, Tyler, Kelsay, & Gantz, 1992; Miyamoto, Osberger, Robbins, Myres, & Kessler, 1993), and preoperative hearing level (Stacey, Fortnum, Barton, & Summerfield, 2006), we assessed how the economic costs incurred by families vary according to these three variables. Finally, we conducted analyses of uncertainty (Briggs & Gray, 1999) to assess the robustness of conclusions to changes in key assumptions.

METHODS

Participants and Procedures

Fortnum, Summerfield, Marshall, Davis, and Bamford (2001) described the national UK ascertainment of 17,160 hearing-impaired children, which was a precursor to this study. The parents of a stratified sample of 8876 families of these children were invited to give consent for them, their child's teacher, and their child's audiologist to participate in this study. The sample included all children with cochlear implants, all children with severe or profound hearing impairment, and a sample of approximately one in nine of the moderately impaired children, stratified by age, gender, and hearing level to be representative of the population of children with moderate impairments (41 to 70 dB). Further details of the methods used to obtain the sample and to confirm the representativeness of those who participated were described by Stacey et al. (2006) and Fortnum, Stacey, and Summerfield (in press), respectively.

During 2000/2001, parents reported data that were used to estimate the economic costs incurred by the family, as described below. Information on nine other variables (Table 1) that have either the potential to explain variation in the economic costs incurred by the family, or are potential confounding variables, were also collected, as described by Stacey et al. (2006).

Measuring Economic Costs Incurred by the Family

Parents were sent a questionnaire (MRC Institute of Hearing Research, Reference Note 2), within which they were asked to report data which allowed estimates of the four measures that are enumerated as (i) to (iv), below. These measures were valued and combined to provide estimates of the two components of the overall economic cost incurred by families (enumerated as (A) and (B) in Table 2).

TABLE 2. Methods used to estimate the two components of the overall economic cost incurred by families (A and B), their total (C), and the level of government benefits (D)

Cost variable	Measures	Valuation
(A) Out-of-pocket expenditure	Sum of expenditure by family in the previous 12 mo on: (i) Special equipment (ii) Special services (iii) Hospital/clinic attendance	Cash value of summed expenditure
(B) Lost productivity	(iv) Number of days spent away from normal activities by parents in the previous 12 mo to accompany child to hospital/clinic appointments	Estimate of loss of gender-weighted productivity for each day spent away from work or leisure
(C) Overall economic cost	Sum of (A) and (B)	
(D) Government benefits	Government benefits received by family because of child's hearing impairment in the previous 12 mo	Sum of cash value of payments to family

The first three measures were the expenditures incurred by the family in the previous 12 mo because of their child's hearing impairment: (i) special equipment, (ii) special services, and (iii) hospital/clinic attendance. These measures were summed to estimate Cost A, the overall out-of-pocket expenditure incurred by the family because of their child's hearing impairment in the previous 12 mo (referred to henceforth as "out-of-pocket expenditure"). Expenditure was reported by parents in UK pounds sterling (£) and converted into euros (€) (Footnote 4).

Parents also reported (iv) the number of days which they themselves had spent away from normal activities in the previous 12 mo to attend hospital/clinic appointments because of their child's hearing impairment. To assign a monetary value to one day away from normal activities, we used estimates from Sach and Whynes (2003) of the proportion of time off work, the proportion of time off leisure activities, and the cost of taking time off work for parents of each gender. These estimates were that the male parent took time off work for 54% of appointments attended and time away from leisure activities for 46% of appointments attended, whereas the corre-

⁴UK Sterling was converted to euros (€) at a rate of €1.00 = UK £0.65. 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 3).

sponding percentages for the female parent were 23% and 77%. Using the human capital approach (Gold et al., 1996; Mohr, Feldman, Dunbar, McConkey-Robbins, Niparko, Rittenhouse, & Skinner, 2000), we estimated the value of time away from work was €102 per day for males and €76 per day for females (mean gross weekly wage in the United Kingdom, excluding tax, pension and national insurance contributions, at 2002 price levels) (Sach & Whynes, 2003). We further assumed that leisure time is valued at zero, and that a child is equally likely to be accompanied by the mother, as by the father, when attending hospital/clinic appointments. Using these assumptions, the average monetary value of one day away from normal activities to accompany a child to a hospital/clinic appointment was estimated to be €36. This value was multiplied by the number of days which parents reported. The result provided an estimate of Cost B, the lost productivity associated with attending hospital/clinic appointments in the previous 12 mo (referred to as "lost productivity").

Finally, we estimated Cost C, the overall economic cost incurred by the family in the previous 12 mo (referred to as "overall economic cost") by summing the values of the two component costs: (A) out-of-pocket expenditure and (B) lost productivity. To aid comparisons with costs in other sectors (Barton et al., 2003; Barton et al., 2006a), all costs were inflated to 2001/2002 levels, using the GDP deflator (HM Treasury 2004, Reference Note 4).

Government Benefits

In the aforementioned questionnaire (MRC Institute of Hearing Research, Reference Note 2) parents were also asked to indicate which government benefits they received because their family included a hearing-impaired child. A list of potential government benefits that may be received because a family includes a hearing-impaired child (UK Department for Work and Pensions, Reference Note 1) was provided, and parents were also asked to add any others which were not included in the list. Weekly unit costs (UK Department for Work and Pensions, Reference Note 1) were assigned to each benefit that was reported. These costs were summed and multiplied by fifty two to estimate Cost D, the value of the government benefits received by each family in the previous 12 mo because their child had a hearing impairment (referred to as "government benefits").

Linear Regression Analyses

Linear regression analyses (Strube, 2003) were conducted to estimate the strength of association between each explanatory variable listed in Table 1

and the seven dependent variables which were estimated in monetary terms ((i) special equipment, (ii) special services, (iii) hospital/clinic attendance, (A) out-of-pocket expenditure, (B) lost productivity, (C) overall economic cost, and (D) government benefits), while controlling the influence of the other explanatory variables. An additional analysis was undertaken to assess whether the overall economic cost associated with implantation varied according to preoperative AHL.

We report results in the form of the parameters of the equation that predicts each dependent variable as the weighted sum of the explanatory variables. Where the 95% confidence interval (95% CI) of a parameter does not include zero, the explanatory variable makes a significant independent contribution to explaining variance in the dependent variable. Before each analysis, the continuous explanatory variables, age and AHL, were transformed mathematically (Footnote 5) to meet the requirement of linear regression analysis that continuous explanatory variables vary linearly with the dependent variable. The transformations were made using methods described by Stacey et al. (2006). Analyses were performed with the SAS system (Freund & Littell, 2000).

Cumulative Economic Costs Incurred by the Families of Implanted Children

We used the results of the linear regression analyses to estimate the cumulative economic cost incurred by the families of implanted children from the time of implantation up to the age of 16 yr (referred to as "cumulative economic cost"). The mean difference between the overall economic cost incurred by the families of children implanted for less than 2 yr and nonimplanted children, was taken as the estimate of the economic cost incurred by the families of implanted children in the first and second years after implantation. The corresponding difference for families of children who had been implanted for between two and four years was taken as the economic cost incurred by the families of implanted children in the third and fourth year after implantation. The corresponding difference for the families of children implanted more than four years

⁵AHL was transformed when analyzing special equipment ($176/(1 + e^{-(AHL - 72)/8.4})$), hospital/clinic attendance ($44/(1 + e^{-(AHL - 69)/4.2})$), lost productivity ($3.26/(1 + e^{-(AHL - 120)/13.2})$), the overall economic cost incurred by the family ($2738.45/(1 + e^{-(AHL - 74.90)/10.32})$), and government benefits ($41.56/(1 + e^{-(AHL - 73.98)/10.92})$). Age was transformed when analyzing out-of-pocket expenditure ($108/(1 + e^{-(AGE - 10)/0.46})$), lost productivity ($-6.26/(1 + e^{-(AGE - 6.66)/0.64})$), the overall economic cost incurred by the family ($-1220.73 + 1217.97/((1 + e^{-(AGE - 13.18)/-0.67})^{17.44})$), and government benefits ($52 \times (-19.27 + 21.25/((1 + e^{-(AGE - 10.92)/-0.08})^{0.73}))$).

TABLE 3. Components of out-of-pocket expenditure incurred by families in the previous 12 mo

	AHL 41–70 dB	AHL 71–95 dB	AHL 96–105 dB	AHL >105 dB	Implanted
Special equipment	€151	€260	€263	€295	€219
(95% CI)	(€89–€213)	(€207–€313)	(€204–€323)	(€234–€355)	(€170–€269)
Range (minimum to maximum)	(€0–€5708)	(€0–€9785)	(€0–€3262)	(€0–€3800)	(€0–€2944)
Special services	€34	€64	€103	€59	€137
(95% CI)	(€0–€77)	(€23–€105)	(€19–€187)	(€37–€81)	(€73–€201)
Range (minimum to maximum)	(€0–€7338)	(€0–€8154)	(€0–€11415)	(€0–€1631)	(€0–€5708)
Hospital and clinic appointments	€45	€64	€107	€67	€239
(95% CI)	(€34–€56)	(€47–€80)	(€59–€155)	(€46–€89)	(€176–€303)
Range (minimum to maximum)	(€0–€1060)	(€0–€3262)	(€0–€4892)	(€0–€2038)	(€0–€8562)
N	340	568	318	348	346

Values are expressed in euros (Footnote 3), at 2001/2002 price levels (Footnote 4) and averaged for families of children with an AHL between 41 and 70 dB (moderate hearing impairment), an AHL between 71 and 95 dB (severe hearing impairment), an AHL between 96 and 105 dB, an AHL >105 dB, and for families of children with implants. N denotes the number of families for whom data were obtained.

was taken as the economic cost incurred by the families of implanted children in the fifth year and each subsequent year after implantation until the child reached 16 yr of age. We then estimated the cumulative economic cost by summing the estimates of the economic cost incurred by the families of implanted children in each year after implantation up to the age of 16 yr. In accumulating costs, costs incurred in future years were discounted at 3% per annum (Footnote 6). Estimates of the cumulative economic cost were then compared with estimates of the incremental lifetime (Footnote 7) health sector costs of implantation (Barton et al., 2006b).

Analysis of Uncertainty

Different methods for assigning a cost to time away from work and leisure activities can result in a 10-fold difference in the estimated annual level of productivity lost by parents of implanted children (Sach & Whynes, 2003). Accordingly, we reestimated the cost of lost productivity by using the assumption that leisure time is valued at 150% of a person's wage rate (Drummond et al., 1997) rather than 0%, with time off work valued at the same rate as in the baseline analysis. This change meant that, on average, one day away from work and leisure activities was valued at €115, compared with the baseline estimate of €36. This revised estimate of lost productivity resulted in a revised estimate of the

overall economic cost incurred by the family, and in turn, a revised estimate of the cumulative economic cost.

RESULTS

Response Rate

Consent to participate was received from the parents of 3274 children (37% of those invited to participate), 527 of whom had a cochlear implant. Questionnaires were returned by the parents of 2858 children (88% of those who consented), 468 of whom had received a cochlear implant. Because some respondents did not answer all questions, we report the number of families included in each analysis.

Unadjusted Effects of Average Hearing Level

The mean annual amount spent on special equipment was €244 (95% CI, €223 to €267), compared with €92 (95% CI, €63 to €122) for special services, and €95 (95% CI, €81 to €108) for hospital/clinic attendance. These values sum to a mean out-of-pocket expenditure of €431 (95% CI, €390 to €473). Results in Tables 3 and 4 show that expenditure was generally higher for families with children with less favorable AHLs. Compared with families of nonimplanted children with AHLs \geq 105 dB, families of implanted children incurred lower levels of expenditure on special equipment, but higher levels on special services and hospital/clinic appointments, resulting in higher levels of out-of-pocket expenditure.

Parents reported that they, themselves, had spent an average of 6.7 d (95% CI, 6.2 to 7.2) away from normal activities in the previous 12 months because of their child's hearing impairment. The number of days increased for the parents of children with less favorable AHLs, and for the parents of implanted children (Table 4).

⁶Future costs were discounted to reflect the preference to consume resources now rather than in the future (Drummond et al., 1997). In line with UK government recommendations, a discount rate of 3% per annum was used (HM Treasury 2003, Reference Note 5). Thus a cost of x euros that would be incurred in y years time was valued as $x/(1.03)^y$ euros.

⁷Using age at implantation together with data from the UK Government Actuary's Department (Government Actuary Department 2004, Reference Note 6), we calculated life expectancy following implantation. Lifetime costs were estimated by summing costs over a child's remaining life expectancy.

TABLE 4. Components of the overall economic cost incurred by families in the previous 12 mo

	AHL 41–70 dB	AHL 71–95 dB	AHL 96–105 dB	AHL >105 dB	Implanted
Out-of-pocket expenditure	€230	€389	€474	€421	€596
(95% CI)	(€149–€311)	(€305–€472)	(€355–€592)	(€348–€494)	(€473–€719)
Range	(€0–€7347)	(€0–€18036)	(€0–€11415)	(€0–€4118)	(€0–€15150)
N (total = 1920)	340	568	318	348	346
Time away from normal activities	5.7	5.7	5.8	5.9	10
(95% CI)	(4.6–6.8)	(5.1–6.4)	(4.9–6.8)	(4.7–7.1)	(8.5–11.5)
Range	(0–88.0)	(0–48.0)	(0–42.0)	(0–80.0)	(0–100.0)
N (total = 1379)	252	417	201	217	292
Government benefits	€3065	€4060	€4125	€4719	€4283
(95% CI)	(€2715–€3416)	(€3811–€4309)	(€3811–€4439)	(€4424–€5014)	(€4024–€4542)
Range	(€0–€14351)	(€0–€15918)	(€0–€14164)	(€0–€14351)	(€0–€14351)
N (total = 1921)	344	589	318	370	348

Values are averaged for families of children with moderate an AHL between 41 and 70 dB (moderate hearing impairment), an AHL between 71 and 95 dB (severe hearing impairment), an AHL between 96 and 105 dB, an AHL >105 dB, and for families of children with implants. N denotes the number of families for whom data were obtained.

The mean value of government benefits received in the previous 12 mo was €4081 (95% CI, €3969 to €4193), though 456 families (16.2%) reported that they received no government benefits. Government benefits tended to be higher for the families of children with less favorable AHLs (Table 4). The mean level of government benefits was higher for the families of implanted children than for the families of nonimplanted children with an AHL <105 dB, but lower than the mean level for nonimplanted children with an AHL ≥105 dB.

Linear Regression Analyses

The parameters of the regression equations are listed in Table 5 for the three components of out-of-

pocket expenditure: (i) special equipment, (ii) special services, and (iii) hospital/clinic attendance. Table 6 shows the overall linear regression equations for Cost A (out-of-pocket expenditure), Cost B (lost productivity), Cost C (overall economic cost), and Cost D (government benefits). The percentages of the variance explained in the analyses are low (ranging from 1% for expenditure on special services to 20% for government benefits). Nonetheless, many of the explanatory variables display a systematic relation with the dependent variables and the overall pattern shown in Table 6 can be interpreted.

AHL • The less favorable a child’s AHL, the higher was the overall economic cost incurred by families. This association arose from higher levels of out-of-

TABLE 5. Parameter values (95% confidence intervals) of the variables used to predict expenditure on special equipment (adjusted $r^2 = 0.03$), special services (adjusted $r^2 = 0.01$), and hospital/clinic attendance (adjusted $r^2 = 0.06$)

Variable	Special equipment	Special services	Hospital/clinic attendance
Constant	–€198 (–€363 to –€33)*	–€62 (–€235 to €111)	€57 (–€42 to €155)
Average hearing level:	€1.10 (€0.58 to €1.63) [‡]	€0.77 (–€0.45 to €1.99)	€1.08 (–€0.07 to €2.23)
Age at onset of hearing impairment:			
≥3 yr vs birth	–€58 (–€178 to €62)	€7 (–€103 to €116)	–€12 (–€84 to €60)
0 to 3 yr vs birth	–€18 (–€91 to €55)	–€19 (–€86 to €48)	–€1 (–€45 to €43)
Age:	€15 (€8 to €22) [‡]	€1 (–€5 to €8)	–€4 (–€9 to €0)*
Gender: Female vs male	–€27 (–€80 to €25)	€20 (–€28 to €68)	€17 (–€14 to €49)
Disabilities:			
Two plus vs none	–€49 (–€121 to €22)	–€36 (–€101 to €29)	€10 (–€33 to €53)
One vs none	€38 (–€34 to €109)	–€34 (–€99 to €31)	€34 (–€8 to €77)
POSL: 4 (“highest”) vs 1 (“lowest”)	€163 (€71 to €255) [‡]	€122 (€38 to €206) [‡]	€65 (–€10 to €120)
Ethnicity: White vs other	€42 (–€46 to €130)	€0 (–€80 to €80)	–€41 (–€94 to €12)
Parental hearing:			
No problems vs some problems	€18 (–€65 to €101)	€5 (–€71 to €81)	€2 (–€48 to €52)
Cochlear implantation:			
Group 1 vs no implant	–€60 (–€192 to €72)	€49 (–€75 to €173)	€74 (–€5 to €153)
Group 2 vs no implant	€29 (–€127 to €185)	€55 (–€90 to €201)	€78 (–€15 to €171)
Group 3 vs no implant	–€63 (–€200 to €75)	–€26 (–€154 to €103)	€176 (€94 to €258) [‡]
Group 4 vs no implant	–€14 (–€191 to €163)	–€70 (–€234 to €94)	€98 (–€8 to €203)
Group 5 vs no implant	–€82 (–€291 to €126)	€230 (€39 to €422)*	€481 (€356 to €606) [‡]
Group 6 vs no implant	€1 (–€206 to €208)	€74 (–€117 to €264)	€152 (–€28 to €276)

Data from 1843 families were included in each analysis. (*p < 0.05, †p < 0.01, ‡p < 0.001). See Footnote 5 for details of the transformations made to average hearing level and age.

TABLE 6. Parameter values (95 % confidence intervals) of the variables used to predict out-of-pocket expenditure (N = 1843, adjusted $r^2 = 0.03$), lost productivity (N = 1324, adjusted $r^2 = 0.11$), overall economic cost (N = 1324, adjusted $r^2 = 0.19$), and government benefits (N = 1843, adjusted $r^2 = 0.20$)

Variable	Out-of-pocket expenditure (€)	Lost productivity (€)	Overall economic cost (€)	Government benefits (€)
Constant	-€119 (-€372 to €133)	€409 (€303 to €514)‡	€159 (-€228 to €545)	€3005 (€2309 to €3700)‡
Average hearing level (transformed):	€0.97 (€0.49 to €1.44)‡	€51 (€16 to €86)†	€0.16 (€0.08 to €0.25)‡	€1.10 (€0.89 to €1.31)‡
Age at onset of hearing impairment:				
≥3 yr vs birth	-€63 (-€264 to €138)	-€3 (-€79 to €73)	-€192 (-€472 to €89)	-€771 (-€1328 to -€215)†
0 to 3 yr vs birth	-€37 (-€160 to €85)	€9 (-€38 to €56)	-€54 (-€228 to €120)	-€401 (-€741 to -€62)*
Age (transformed):	€1.12 (€0.17 to €2.08)*	€36 (€26 to €45)‡	-€0.09 (-€0.21 to €0.03)	€0.94 (€0.70 to €1.18)‡
Gender: Female vs male	€13 (-€75 to €101)	-€13 (-€47 to €21)	€5 (-€118 to €129)	-€77 (-€321 to €167)
Disabilities:				
Two or more vs none	-€72 (-€192 to €48)	€24 (-€21 to €69)	-€62 (-€229 to €105)	€2546 (€2214 to €2878)‡
One vs none	€36 (-€83 to €155)	-€1 (-€47 to €45)	€51 (-€118 to €221)	€691 (€360 to €1021)‡
POSL: 4 ("highest") vs 1 ("lowest")	€353 (€199 to €507)‡	-€23 (-€88 to €42)	€278 (€38 to €518)*	-€1593 (-€2020 to -€1166)‡
Ethnicity: White vs other	-€3 (-€151 to €144)	-€20 (-€79 to €40)	-€63 (-€281 to €156)	€453 (€45 to €862)*
Parental hearing:				
No problems vs some problems	€24 (-€116 to €163)	€1 (-€77 to €81)	€31 (-€178 to €240)	-€73 (-€459 to €313)
Cochlear implantation:				
Group 1 vs no implant	€75 (-€146 to €297)	-€8 (-€90 to €75)	€84 (-€208 to €375)	-€457 (-€1075 to €161)
Group 2 vs no implant	€153 (-€109 to €415)	€30 (-€70 to €130)	€12 (-€339 to €363)	€88 (-€640 to €816)
Group 3 vs no implant	€96 (-€133 to €325)	€29 (-€59 to €118)	€221 (-€79 to €521)	-€169 (-€797 to €460)
Group 4 vs no implant	€22 (-€274 to €319)	€15 (-€93 to €123)	-€14 (-€404 to €376)	-€393 (-€1218 to €433)
Group 5 vs no implant	€622 (€276 to €969)‡	€282 (€151 to €412)‡	€1214 (€754 to €1673)‡	-€595 (-€1552 to €361)
Group 6 vs no implant	€230 (-€117 to €577)	€321 (€203 to €440)‡	€474 (€42 to €905)*	-€253 (-€1217 to €710)

See Footnote 5 for details of the transformations made to average hearing level and age. * $p < 0.05$, † $p < 0.01$, ‡ $p < 0.001$.

pocket expenditure (due to higher levels of expenditure on special equipment) and lost productivity. Government benefits were also estimated to be higher the less favorable a child's AHL.

Age at Onset of Hearing Impairment • The overall economic cost was not significantly related to age at onset of hearing impairment. However, levels of government benefits were estimated to be lower when hearing impairment started after rather than at birth.

Age • The overall economic cost was not significantly related to age, as lost productivity costs, which were estimated to be lower for older children (reflecting the fact that older children were estimated to visit hospital less frequently), off-set out-of-pocket expenditure, which was estimated to be higher for older children (as expenditure on special equipment was estimated to increase with age). Government benefits were estimated to be lower for the families of older children.

Gender • The overall economic cost was not associated with gender, nor were out-of-pocket expenditure, lost productivity, or government benefits.

Additional Disabilities • The overall economic cost incurred by families was not estimated to vary according to whether a child had additional disabilities in addition to their hearing impairment. Government benefits were, however, estimated to be higher for the

families of children who had disabilities in addition to their hearing impairment.

Parental Occupational Skill Level • Families with parents at the highest level of occupational skill had a higher overall economic cost than families with parents at the lowest level of occupational skill, as they incurred more out-of-pocket expenses—spending more on special equipment and special services. Conversely, families with parents at the highest level of occupational skill had lower levels of government benefits.

Ethnicity • Ethnicity was not associated with the overall economic cost, out-of-pocket expenditure, or lost productivity. The families of white children, however, were estimated to receive higher levels of government benefits compared with the families of other children.

Cochlear Implantation • The overall economic cost incurred by the families of implanted children was estimated to be significantly higher in the first 2 yr of implant use. This result arose because the families of children implanted before the age of 5 yr, who had had their implant for less than 2 yr, were estimated to have higher out-of-pocket expenditure, arising from higher expenditure on special services and hospital/clinic attendance, and because the families of both groups of children implanted for less than 2 yr also had higher

levels of lost productivity, arising from increased parental time away from normal activities. An additional analysis showed that the overall economic cost associated with implantation did not vary according to a child's preoperative AHL. The level of government benefits was not estimated to vary according to whether a child had an implant.

Cumulative Economic Cost Incurred by the Families of Implanted Children

The cumulative economic cost incurred by the families of implanted children between implantation and age 16 yr was estimated to be €3355 for a child implanted at age 3 yr (the modal age at implantation within our sample of implanted children), and €949 for a child implanted at age 6 yr (the mean age at implantation within our sample of implanted children). Barton et al. (2006b) estimated the incremental health sector costs of implantation to be €132,040 for a child implanted at age 3 yr and €131,292 for a child implanted at age 6 yr. The cumulative economic cost therefore corresponds to between 0.7% and 2.5% of the incremental health sector costs of implantation, depending on age at implantation.

Analysis of Uncertainty

When leisure time was valued as 150% of a person's wage rate, rather than 0%, the cumulative economic cost was estimated to be €3787 for children implanted at age 3 yr, rather than €3355, and €2695 for children implanted at age 6 yr, rather than €949. Thus, under these assumptions, cumulative economic costs were estimated to correspond to up to 2.9% of the incremental health-sector costs of implantation.

DISCUSSION

These analyses show that out-of-pocket expenditure by families, time away from normal activities by parents, and hence the overall economic cost incurred by families, vary systematically according to clinical and demographic characteristics of hearing-impaired children and their families. Assessed from a societal perspective, the cumulative economic cost incurred by the families of implanted children was estimated to range between €949 and €3787, depending on a child's age at implantation and on assumptions made about the value of parental time away from normal activities. These values correspond to up to 3% of the incremental health sector costs of implantation. Assessed from the perspective of the family, costs are highest in the 2 yr immediately after implantation. During this time, the mean

annual cost of out-of-pocket expenditure and time away from normal activities amounted to €1214, when children receive implants before the age of 5 yr, and to €474, when children receive implants after the age of 5 yr. The level of government benefits was also shown to vary according to characteristics of hearing-impaired children and their families, though they were not estimated to vary according to whether or not a child had received a cochlear implant.

Families of implanted children spend less each year on special equipment than do families of non-implanted children with AHLs greater than 105 dB (Table 3). However, this difference is not significant when potentially confounding variables are controlled (Table 5). Thus, the results provide only weak support for the hypothesis (Cheng et al., 2000; Hutton et al., 1995; Summerfield et al., 1997) that implantation is associated with a reduction in expenditure by families on special equipment.

Explanations

Implantation is an invasive surgical procedure that entails extensive rehabilitation. After a child has been identified as suitable for implantation and the family has elected for implantation, it is to be expected that parents will increase their expenditure on their child and take additional time away from normal activities to maximize the benefits of implantation. Indeed, it is widely judged that family commitment is critical to maximizing the benefit that a child receives from implantation (e.g. Arts, Garber, & Zwolan, 2002). The finding that the cumulative economic cost was higher for children implanted under the age of 5 yr is likely to reflect the fact that children implanted younger require relatively more hospital visits. Finally, the finding that the level of government benefits received is not related to implantation is compatible with the idea that disbursement of benefits is based more on the physiological status of children than their functional status.

Comparison with Other Studies

This study compared the families of implanted and nonimplanted children to estimate the economic costs incurred by the families of implanted children. The findings complement and extend the results of Sach and Whynes (2003) and Cheng et al. (2000), who considered the families of implanted children but who did not make comparisons with nonimplanted control patients. Sach and Whynes (2003) found that total time off work and leisure activities by parents was lower for the parents of children who had used their implants for longer, varying between a mean of 10 d per annum when children had been implanted for less than 2 yr, and a mean of 3 d per

annum when children had been implanted for more than 4 yr. These levels are lower than our estimates of 17 (95% CI, 14 to 20) d per annum when children have been implanted for less than 2 yr and 7 (95% CI, 4 to 9) d per annum when children have been implanted for more than 4 yr. Sach and Whynes (2003) also estimated the level of out-of-pocket expenditure to be on average €502 per annum, compared with our estimate of €596 (95% CI, €473 to €719). These differences may be explained by the fact that Sach and Whynes (2003) collected data from the parents of children implanted in one UK hospital, where it may be that children receive fewer hospital/clinic appointments but more outreach visits than the average.

In contrast, our empirically based estimates of the cumulative change in economic costs (between implantation and age 16 yr) associated with implantation for the United Kingdom are generally lower than estimates based largely on informed opinion for the United States (Cheng et al., 2000). The US estimates of the lifetime cost of time away from work (lost productivity) and travel expenses were US \$4623 and US \$5419, respectively. Our estimates (calculated using an exchange rate of US \$1.20 = €1.00) of the cumulative cost of lost productivity are US \$638 (implantation age, 3 yr) and US \$969 (implantation age, 6 yr), and, for hospital/clinic attendance, US \$2158 (implantation age, 3 yr) and US \$1079 (implantation age, 6 yr). Also, we estimated that the cumulative expenditure on special equipment to be a saving of US \$725 when a child is implanted at the age of 3 yr, but an increase of US \$137 when a child is implanted at the age of 6 yr, whereas Cheng et al. (2000) assumed a lifetime saving of US \$1012, independent of age at implantation. Differences in methodology between the studies, in salary levels, and in the cost of living between the United States and United Kingdom, may explain these small differences.

It is not clear whether the present analysis should be extended to include an economic cost to society resulting from days off school by children. Lewis, Bruininks, Thurlow, and McGrew (1989) estimated that children with special needs in the United States who receive less special education achieved lower levels of productivity in adulthood, reflected in lower incomes, than children who received more special education. By implication, if implanted children take more time off school than similar nonimplanted children (e.g., due to the time required for surgical implantation and rehabilitation), they might suffer a loss in future earnings. By adopting the same assumptions as Lewis et al. (1989), it is possible to estimate the loss of lifetime earnings that might be associated with the numbers of days off school reported for the

children in the present study. After controlling other variables, implanted children were estimated to take more time off school than nonimplanted children. The estimated loss was €2500 for children implanted at age 3 yr and €5500 for children implanted at age 6 yr. An alternative view is that children's future earnings will be increased as a result of the enhanced educational attainments associated with implantation (Boothroyd & Boothroyd-Turner, 2002; Geers, Nicholas, & Sedey, 2003; Spencer, Tomblin, & Gantz, 1997; Spencer, Barker, & Tomblin, 2003; Stacey et al., 2006; Tomblin, Spencer, & Gantz, 2000). By combining estimates of the earnings of hearing-impaired people (Mohr et al., 2000) with assumptions about the proportion of implanted children who will achieve the earning level of people with normal hearing (Cheng et al., 2000), it is possible to estimate that implantation will lead to an increase in lifetime earnings of the order of €133,000, sufficient to offset completely the health care costs of implantation. Pending controlled measurements of the earnings of adults implanted when children, both views are speculative. For that reason, we did not include the value of time off school by children in estimating the overall economic cost incurred by families.

Limitations

This study has two key limitations. First, the explanatory variables account for only a small percentage of the variance in the measures that contribute to the overall economic cost incurred by families. In contrast, the same variables explain 70% of the variation in children's academic abilities as reported by parents and 39% of the variation in children's reading ages as reported by teachers (Stacey et al., 2006). The limited percentage of the variance explained in overall economic costs may result from noise in our data stemming from the difficulty faced by parents in recalling and enumerating their use of resources, and from differences between families in the need and opportunity to incur costs for reasons unrelated to the explanatory variables that we measured.

The second limitation is that the cross-sectional design required us to infer trends over time by comparing different groups of children. Some protection against reporting spurious effects is provided by the fact that all associations revealed by linear regression, including associations with age and with different ages at implantation and durations of implant use, are made while controlling the strength of association with all other variables. Nonetheless, it would be desirable for further studies to use a longitudinal design.

Implications

Our results imply that relative to the situation that would arise if their child had not been implanted, families with an implanted child in the United Kingdom are likely to incur additional out-of-pocket expenditure if their child is implanted when under the age of 5 yr, to take more time away from work/leisure activities, but to receive similar levels of government benefits. Families can however be reassured that the results of this study indicate that, historically at least, the additional out-of-pocket expenditure has not been great, averaging a few hundred euros per year.

Nor should the policy decision of whether to fund paediatric cochlear implantation hinge on the present results. We estimate that the increase in the mean overall economic cost incurred by families, even when accumulated in the worst case, amounts to only €3787. This total is less than 3% of the estimated incremental health sector costs of implantation (Barton et al., 2006b) and its inclusion does not change the conclusions of an analysis of the cost-effectiveness of paediatric implantation (Barton et al., 2006b).

In conclusion, although the economic impact of implantation may be material for the family of an implanted child, it is small in relation to the societal cost of providing cochlear implants to children.

ACKNOWLEDGMENTS

This study was supported by the Medical Research Council, Defeating Deafness—The Hearing Research Trust, and the National Lottery Charities Board. We thank all children, parents, teachers, and health care professionals who provided data. Ms. D. Betts, Ms. S. Holroyd, and Ms. M. Shaw coded and checked data. Dr. D.H. Marshall, Professor A.C. Davis, Professor J.M. Bamford, and Dr. K.E. Bloor provided advice in formulating the design and methods of the study. Dr. D.H. Marshall programmed the databases that supported the study and guided initial analyses of the data. We thank two anonymous reviewers whose criticisms helped improve this paper.

Address for correspondence: Professor Quentin Summerfield, Department of Psychology, University of York, Heslington, York YO10 5DD, United Kingdom. E-mail: aqs1@york.ac.uk.

Received April 20, 2005; accepted May 13, 2006.

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