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**Contingent Valuation and Paediatric Cochlear Implantation:  
What does one say about the other?**

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## **Introduction**

Cochlear implantation is a high-cost low-volume intervention that gives profoundly deaf adults and children a sensation of hearing [1,2]. The implantation process involves much multidisciplinary working (audiologists, speech and language therapists, teachers of the deaf, psychologists and surgeons) in order to assess the suitability of candidates for implantation and to work with the child once the device has been surgically fitted [3]. Once fitted the user needs support for as long as they use the device. The first child was implanted in the UK in 1989 at the Nottingham Paediatric Cochlear Implant Programme (NPCIP). By 2001 it was estimated that over 1600 children had been implanted in the UK whilst in the USA 14,000 people (7000 of whom were children) received implants. The total number of implants world-wide for both adults and children has been estimated as around 25,000 [4,5].

The benefits of implantation are numerous from aiding auditory perception [6-8], enhancing speech intelligibility [9,10], and increased likelihood of mainstream schooling [11,12] in addition to a range of psychosocial quality of life benefits [13-15]. The benefits of implantation are expected to be greatest for children rather than adults and it is this age group that this study will focus upon.

In a recently published systematic review of the economic literature on cochlear implantation [16] 21 published and unpublished economic studies of cochlear implantation were identified. Of these 18 were economic evaluations (16 cost-utility analyses and 3 undertook partial cost-benefit analyses, one study attempted both). Since that publication two cost-effectiveness studies have been performed [17,18]. The major conclusions of the review were that cochlear implantation is cost-effective (£8,250 - £12,400 per QALY in the UK [16]) regardless of patient age but that there was a heavy reliance upon assumptions due to gaps in our knowledge base. Cost-utility estimates were most affected by the level of utility gain and the length of device use. The cost-benefit studies undertaken to date have not employed contingent valuation methods rather they measure the monetary benefits in terms of the expected cost savings from education and the increased earning potential of implantees. The cost savings from education should be put into the numerator of the cost-benefit ratio. Clearly, also, the benefits of Paediatric Cochlear Implantation (PCI) are wider than increased future income and therefore, the cost-benefit studies to date have underestimated the benefits of PCI.

Given the importance of outcome in sensitivity analyses, the fact that different utility measures to date have measured wide discrepancies in the gains from cochlear implantation [19], and that cost-benefit studies to date have underestimated the benefits of PCI, another look at outcomes is justified. Not only do different utility measures give different answers but they may not be capturing all the relevant benefits pertaining to PCI [20]. This is because whereas "WTP imposes no restriction on which attributes of a programme people are

allowed to express a value for, ...QALYs...are based on preferences for *health outcomes* only" [21]. It has been suggested elsewhere [16] that a balance sheet approach to cost benefit analysis for cochlear implantation should be undertaken such that all benefits and costs can be stated regardless of whether or not they can be measured. The benefits that are likely to be excluded from present economic evaluations of PCI include:

- The value of information or "process utility" [21];
- Option values;
- Psychosocial benefits such as increased confidence and safety;
- Non-use values/caring externalities such as the benefits for families.

To date, contingent valuation methods (CVM) within the health care setting have largely been applied to low-cost high-volume interventions such as to antenatal screening & care [22, 23], IVF [24, 25, 26], cardiovascular treatments such as hypertension medication [27] and respiratory diseases [28]. The majority of CVM studies evaluate medical interventions (31 or 64% of studies) or pharmaceutical interventions (12 or 25% of studies), only 2 (6%) empirical studies [28] valued surgical interventions. If an outcome measure, as used in economic evaluation, is to be useful in aiding prioritisation of health care interventions it is essential that it can be applied to all interventions regardless of cost and volume.

This paper presents work in progress on the first application of willingness to pay (WTP) and willingness to accept (WTA) in the context of PCI. This paper is structured in three sections. Firstly the methods used will be described, such that it is explicit what and how parental valuations were measured. Secondly, the results section will be divided into the following sections: 1) the nature and scope of differences between WTP and WTA bids elicited; 2) participants reasoning behind WTP and WTA bids; 3) the relationship between WTP and a) changes in health related quality of life as measured by the Visual Analogue Scale (VAS) of the EuroQol, and b) two specific clinical measures CAP (Categories of auditory performance) [29] and SIR (Speech Intelligibility Rating) [9,10], controlling for income; and 4) the implications of various biases in reported preferences for the application of contingent valuation at a societal level. Finally, questions are posed for discussion, future research directions are stated and a conclusion offered. By doing this it is hoped that lessons, both on the application of CVM and its implications for PCI, can be learned.

## **Methods**

At the time of writing 184 face-to-face semi-structured interviews had taken place since July 2001 (136 with parents/guardians of post implanted children, 26 with parents/guardians of preimplanted children, 12 with parents of children found unsuitable for PCI (via phone interviews), and 10 interviews with the general population (convenience sample)). This paper focuses on the 162 pre- (ex-ante user based) and post- (ex-post user based) implanted study

groups (representing around 45% of those implanted or due for implantation at NPCIP), as these interviews have been running for longer. The face-to-face interview format was chosen because it was the elicitation style recommended by National Oceanic and Atmospheric Administration (NOAA), as the most likely to give reliable estimates [30].

All parents received written invitations (approved by the local ethics committee) containing an invitation letter, a patient information sheet and an issues sheet (listing themes to be covered in the interview) a month in advance. Parents were asked to reply using a prepaid envelope and reply slip regarding their willingness to participate in the study. All parents were interviewed either prior/during/or after their child's appointment in a number of locations but all had quiet rooms suitable for interviewing available. The parents of the preimplant group were interviewed either at initial/3-week or 6-week tuning. The ethics committee did not feel it was acceptable for us to approach them before this time. Therefore, they are not strictly an ex-ante group in that their children already had the surgical procedure and device fitted at the time of the interview. However, even 6-weeks after the initial tuning is too soon to see any real benefits from the implant. It was perhaps wise not to interview parents prior to implantation since parents may have felt compelled to inflate their bids. As it was all parents were made aware of the first authors independence from the programme and that anonymity was guaranteed.

The CVM component of the questionnaire was nested in the middle of a wider questionnaire looking at other qualitative and quantitative issues to do with families going through the cochlear implantation process. Therefore, by the time parents were asked the WTP and WTA questions they had had time to consider the issues of implantation and build up a rapport with the interviewer. All participants were interviewed by the first author. The questionnaire design and content was discussed at two focus groups with parents during the design phase and the questionnaire also underwent piloting.

Prior to being asked question 10 on WTP (Appendix A) parents were given descriptions and information about PCI. All of them would have heard such information before, since the NPCIP undertake considerable time during the assessment process (which on average lasts 9 months) to counsel parents on the risks associated with the process, what are reasonable expectations regarding outcomes and what the process involves. The outcomes of PCI were presented as uncertain, as indeed parents are counselled in the assessment phase of the programme, such that the WTP/WTA bids "include risk preference" [28]. Obviously for the post-implant group they will have knowledge on the outcomes their own child have achieved to date. They were then asked the WTP question (see appendix A) which uses a bidding process (BP) and an open-ended (OE) question. The money measure used here was equivalence surplus/variation, that is the maximum the user is willing to pay to avoid the removal of the programme from the NHS (i.e. a quantity decrease). The bidding

process followed a predetermined sequence starting off with a bid of £80 per month for 25 years, a level which is approximately half that which it would cost to provide the service for one implanted child over that time. A monthly figure was used such that parents might think in terms of monthly mortgage payments or bills. The open ended question asked for a one off lump sum payment.

After this parents were asked for socio-demographic information, including gender, age, age on leaving full-time education, employment status, household income, ethnicity, and number of children. After this parents were asked the WTA question 12 (see appendix B), which again used a bidding process and a separate open-ended question. The money measurement used was compensating surplus (that is the minimum amount of compensation a user is willing to accept in return for the removal of the programme).

A bidding game and an open-ended, rather than a take-it-or-leave-it, question format was adopted because of the limited sample size available and as a result of consultation with parents at focus groups who preferred the interactive nature of the bidding game.

Later in the questionnaire the parents were asked for reasons for their responses to the WTP/WTA questions to gain some insight into how parents' thought about and reached their level. Parents were also asked how much they thought it cost to implant and maintain a child to see if, independent of parents' reports, this influenced the level of bids obtained. They were also asked if they had previous experience of valuation questions on health care; whether they had used private health care and therefore, had some appreciation of thinking in monetary terms about health; and whether they considered there to be any alternatives to PCI which would have improved their child's quality of life. Parents were also presented with a discrete choice question – if they had £50,000 would they buy the implant or spend it on something else, so not constrained by income, and asked to rate their strength of preference for their choice.

All statistical analysis was carried out using SPSS Version 10 [34].

## **Results**

The results presented here are based on 162 completed interviews, representing nearly 100 hours of interviews (mean interview time 37 minutes). 208 parents were invited to interview of which 162 (78%) participated, 40 (19%) could not participate on the date suggested but have said they will participate in the future, and 6 (3%) responded that they did not want to participate at any point in time. The subject's socio-economic characteristics are presented in appendix C.

The response rate for the WTP question was 95% for the postimplanted group and 85% for the preimplant group. Of the seven post implanted that gave zero bids three were due to personal financial problems (one was self-employed and therefore, had unpredictable

income), two people found the question difficult (one because they already gave voluntary donations to The Ear Foundation, a charity run for children with cochlear implants), and two were protest bids (one because they paid 40% taxes and didn't feel they wanted to pay more to keep the programme running although they'd pay more to keep their own child on a programme and the other because they felt the programme had lowered the threshold criteria too much such that children are now receiving implants that don't derive great benefit from them). The four zero bids elicited for the pre-implant group were a result of one having financial difficulties, one not willing to put a value on it other than to say "sell my soul for it", one because he felt his taxes were already misspent and didn't feel he should pay more, and one refused to answer because he'd answered a similar question before and found the answer "can be thrown both ways".

The response rate to at least one of the WTA questions was just 39% for the postimplanted group and 37% for the preimplanted group. 56% of those not giving a valuation stated that they felt they could not give a value because they did not know how much it would cost to continue the service in the private sector and that was what they would want compensation for. 8% of these 56% also stated they would want compensation for their child's loss of earning potential (see table 5, all tables in Appendix D).

It was suggested in the introduction to this paper that a number of benefits of PCI were not included in economic evaluations to date. To see how important these benefits were to parents of children with CI the parents were asked to state what benefits they had seen, or hoped to see, from cochlear implantation (see table 1 for results). It can be seen from this that safety (III), educational benefits (II), the range of options opened up (IV) and so forth are all mentioned by over 65% of those interviewed. Altruism (VII) scored lowly, just 35% mentioning this. It thus highlights the importance of non-health outcomes for PCI.

#### *The nature and scope of differences between WTP and WTA bids elicited*

As can be seen from the completion/response rates given above there was a clear preference for answering the WTP question as opposed to the WTA question format. This was perhaps due to the ease with which participants identified with the scenario presented them but also because intrinsically participants either directly related compensation to the amount they'd need to continue private care or were ethically opposed to taking compensation from the NHS (see table 5). Parents preferences for PCI were also observed when asked if they'd spend £50,000 on PCI or something else, 159 (98%) choose the implant, 2 (1%) choose the money and 1 (1%) stated it was for professionals to decide.

There was also a large variability between WTP and WTA as to which elicitation method parents choose to answer (see table 2). It can be seen that for the WTP question parents who only responded to BP or OE had a clear preference for answering the monthly

BP question. Parents were more indifferent between the two question formats for the WTA question, although clearly more amenable to the OE question when not constrained by income. Only 54% answered both BP and OE for the WTP question and 20% for the WTA question.

#### *Willingness to Pay and Willingness to Accept estimation*

Table 3 presents the mean WTP and WTA bid amounts by user group and question format, where sample size refers to the numbers in each group that answered a particular question format. It can be seen that Mean/median WTP estimated using the bidding game question format for the post implant group (£150/£50) is not significantly different from the preimplant group (£135/£55) ( $P=0.84$ ). The same is true for WTA under the same question format, although both groups give higher mean/median WTA values than WTP, the preimplant group have higher WTA values than the post implanted (£485/£375 compared to £254/£167 per month for 25 years,  $P=0.23$ ). This pattern, of higher (lower) mean/median WTA (WTP) bids from the preimplant group compared to the postimplant, is repeated when looking at the open ended lump-sum question format. Where WTP is £25,832/£3000 for the post-implanted compared to £11,610/£4500 for the preimplanted (the difference was not significantly different,  $P=0.71$ ). However, a statistically significant difference was found between the lump-sum WTA amounts elicited from the two groups. The mean/median minimum one-off lump-sum compensation payment the preimplant group would accept was £613,250/£450,000 versus £197,492/£32,500 for the post implant group ( $P=0.014$ ).

It can be seen from Figure 1 (all figures in Appendix E) that at low and high WTP bid levels pre and post implanted values are similar (chiefly because only a few valued it at the extremes) but for values between £200 and £500 a greater number of the preimplanted parents, compared to post implanted parents, were willing to pay these values. That is they valued PCI more highly over this range. The same finding was found for lump sum WTP.

Figure 2 clearly shows parental valuations were higher when the payment strategy involved monthly instalments rather than a one-off payment. This is not surprising given an income constraint.

Figure 3 shows that the pre implanted group value PCI more highly than the post implanted group under a WTA question format. A similar pattern was found for the lump sum WTA bid levels.

#### *Participants reasoning behind WTP and WTA bids*

Table 4 presents the reasons given by participants for reaching their WTP bid level. Clearly the most common factor influencing bid level is ability to pay (61.5%) with the next highest factor being that it reflected the benefits of PCI (18.6%).

In addition to the explicit reasons given above, parents also made other remarks during the course of the interview regarding WTP. For the majority the first response was one of “you want it so much you’d have to think about selling your house for it”, “you’d find ways to pay for it yourself. Would’ve remortgaged the house for him ” or “if we’d been turned down in Nottingham we’d have gone to Australia or the USA”. Other parents stated that they’d “sacrifice his disability living allowance for it. The benefit is worth more than this but I am constrained by what I can afford” or that insurance would be an alternative “We’d pay £200 but could get medical insurance, like in the US, so would not pay that much”. Others were of the opinion that “no-one could afford to pay for the whole implant themselves”. That is to say that for the majority their first thoughts were to their own child. In these situations parents were given clarification that they were asked to give their WTP for the programme rather than their own child and that they should do this from their current position.

Parents often found the open ended lump sum question more difficult to answer. Comments made with regard it can be characterised by the following statements “depends if I can get a loan”, “wouldn’t have spare amount of money like that”, “pull resources from family” and “years change” the amount one can afford.

Parents made comments that showed an awareness of how time, family structure and uncertainty influenced their bids. “Got to think about it selfishly, our daughters implanted. Got to be able to afford other things but we know how much it’s helped us. We’ve got another child, have to think of that”, “Hard to say because at the beginning it was such an unknown quantity but having seen so many benefits I would sacrifice a lot”, and “very early stages for us so want to know support is going on”. One parent stated that WTP was “hard because I don’t have to. If he was preimplant I could” whilst another stated that “before [name] was deaf I wouldn’t have paid anything”. Some mothers stated that “you’d have to ask my husband...my husband deals with the money”.

A number of parents made reference to the sacrifices they’d already made for their child to be on the programme “we moved for it and my husband took a £15,000 cut in salary to be in Chesterfield rather than Scotland...have to put up with enough as it is”, “I came to Nottingham despite Manchester being on my doorstep because I want the best for my son”, “because we are travelling monthly here we bought a new car and make monthly payments for that” and before the NHS funded PCI some parents “paid the whole cost – we’d sacrifice everything for our child. Impossible question to answer, I put a lot into the cause anyway. Rather launch a campaign. I’d pay £100 per month because I can afford to, I wouldn’t contribute to other charities. Don’t think people would make a large payment for other peoples children on a voluntary basis. Particularly the case for those further down the line. Look at all the terrible disasters around the world, in comparison between cochlear implantation and starving children I’d rather save the starving children”.



For some parents the idea of paying meant they would expect more from the service “I’d expect to see a lot more out of it e.g. more regular check ups” or “happier [to pay] for equipment not operation – give them a sense of responsibility”.

Asking the WTP question also brought out parents views on other issues such as bilateral implants “we asked at the time if we could pay for the other ear and were told it would be £40,000 but didn’t go ahead as we didn’t feel it’d be beneficial” and on the NHS “just know there are quite a few people who bleed the system dry. If I knew people would really benefit I’d willingly pay more but I think that’s the problem with the NHS at the moment, people scam it. I’ve become very cynical about the NHS over time. Money talks”.

Table 5 shows the factors influencing the WTA bids obtained, as illustrated with quotes from the parents.

#### *Relationship between WTP and the gains in quality of life, CAP and SIR*

One way CVM studies can test construct validity is to examine the relationship between WTP bids and variables thought to be predictors of WTP such as income and measures of outcome. One might expect WTP bids to be higher the greater the expected or observed outcomes, the higher a persons incomes and so forth. Since the benefits of implantation take time to observe, WTP/WTA bids might be larger the longer the parent’s child has been implanted. However, competing against this is the knowledge that their child no longer needs the same level of support and this would act to reduce the WTP/WTA amount stated. It is further complicated by the fact that the income of this cohort decreases over time. That is those children first implanted came from higher income households who presumably had greater knowledge about cochlear implants and the resources to battle for it.

To test for this partial correlation’s, controlling for income, and a series of multivariate regression analyses were performed using the data on the post implant group for each of the following outcome measures: change in HRQOL, change in QOL (both measured on the EuroQol VAS scale), CAP score and SIR score.

The partial correlation’s between WTP and HRQOL, QOL, SIR and CAP, controlling for income, showed no significant correlations (see table 6). A series of multivariate regression analyses were undertaken with the natural logarithm of WTP monthly as the dependent variable and income, if the parent stated WTP reflected the benefits of PCI, the number of implanted children a family had, years since implantation, the onset of the child’s deafness, the gender of the child, if the child was in mainstream school or not and outcome (change in HRQOL & QOL and most recent absolute CAP & SIR scores) as the explanatory variables. The results can be seen in tables 7-10. The only significant predictors of WTP were income and whether the parents stated that they formulated their bid on the basis of the benefits of PCI (variable named benefit). Both Income and benefit were positively related to

WTP bid, such that the higher a parents income or if parents reported WTP as a measure of the benefit the higher the bid which is what one might expect.

So why don't the WTP bids reflect the outcomes observed by the post implanted group? There are several possible explanations. Firstly, that parents are not the best judge of their child's outcome. Secondly, parents value PCI regardless of outcome (a view supported by parental comments to the effect that we had "Nothing to lose if it failed be in the same position" and "she couldn't of lost anything, she had no useful hearing"). Of course having put a child through implantation parents are less likely to say it was unwise. Thirdly, parents' valuations were more strongly influenced by other factors, e.g. income. The third of these may be the most relevant given parents were asked their WTP for the UK programme rather than for their own child. In this situation it shows parents understanding of the question and the fact that their valuations should reflect the benefits of the overall programme rather than just those of their child. Lastly, it could also be that the programme is good at selecting a fairly homogenous group in terms of prognosis, therefore little variation in some of these variables, everyone was a success, and there wasn't that big a difference in terms of the success they attained.

*The implications of various biases in reported preferences for the application of contingent valuation at a societal level*

The present results are clearly from a biased sample – a sample that have much experience and well defined preferences for the intervention of interest. Whilst this has advantages, such as the fact that "patients are likely to have greater knowledge about the health consequences of a treatment and hence the cognitive burden of the task may be lower than for non-patients" [28], for the purposes of performing a societal cost-benefit analysis it falls short of being representative. Elicitation of WTP/WTA values from the general populace has only just begun but one might speculate, given the low prevalence rate (1 or 2 per 1000) and probability of being directly affected or knowing someone that is affected, that values will be low. That is the caring externality element is likely to be small [28]. Such that the proportion of individuals WTP for PCI as a function of bid could look something like figure 4.

It might be expected that whilst a very small proportion of the population (likely to be those directly affected or who know someone affected i.e. those captured in this study) are willing to pay quite significant amounts to PCI the vast majority of the population would be willing to pay little or nothing towards the intervention.

This presents a problem – not just relevant to cochlear implantation but to all low prevalence conditions/ rare interventions since it may mean that some very effective interventions will appear to be considered by the general population to hold little value to society. In such a situation it becomes pertinent to ask whose valuations should count? Is it

equitable, just because of low prevalence and the resulting low weighting to attach low priority to an intervention that will offer a good benefit to a minority? Of course, this is a description of a scenario a priori and as such this is a description may be proved wrong but interviews done to date have shown lower WTP bids or WTP bids at the lower end of the distribution presented above.

### **Conclusion and points for discussion**

It has been shown that for PCI parents preferred answering WTP rather than WTA questions and within this preferred answering a bidding process than an open ended question format. The size of parental bids highlights how strongly parents value the PCI programme, even when not paying explicitly for their own child and regardless of the outcome. Parents were most likely to give a protest value when answering the WTA question as opposed to the WTP question. Many parents thought deeply about the questions asked of them shown by the range of relevant factors they raised in discussion. In the multivariate regressions analysis the significant predictors of WTP were income and whether the parents had stated WTP reflected the benefits of PCI.

However, the work has raised a number of issues, which the authors would be grateful to receive assistance with. For example, given the low number placing a value on the WTA question (despite clearly having a strongly positive value – the value of private care) should one abandon the question or infer the private market cost of PCI and substitute this in? *How should WTA be analysed in the light of this?* Is it correct to treat the pre and post groups as independent samples? How should one apply CVM at a societal level for low volume interventions? Given the high-income status of this group, how should one aggregate the values to reflect this? Why are parents' bids insensitive to outcome? Should the data be trimmed? If so how much and if not is it enough to look at median values? *How should we structure the argument?*

The long-term aim of the study is to perform a cost-benefit analysis of PCI incorporating the other data collected regarding patient costs etc into the analysis. The preimplant sample will also be re-interviewed at 12 months post implantation such that WTP/A bids can be analysed as paired data to measure change over time. It may also be desirable to test the convergent validity by performing a travel cost method study.

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## Appendix A - WTP Question

### Question 10:

The NHS started funding paediatric cochlear implantation in the early 1990's. I would like you to imagine that just recently the NHS had announced it would no longer be funding the paediatric cochlear implantation programme given current resource levels. Therefore, the only way cochlear implants could be provided would be if people were willing to pay money (in addition to the taxes they already pay) towards them.

I am interested in finding out how much you value cochlear implants for children compared to other demands on your household resources. In order to get a comparable value I will be asking you to place a monetary figure on your valuation of cochlear implants. You will not be expected to pay this amount but it should represent the value you would be willing to pay towards funding a paediatric cochlear implantation programme in the UK.

**The NHS does provide cochlear implants and I believe that people should not have to pay directly for health care. This question is simply a method of measuring how strongly you feel about the UK having a paediatric Cochlear implant programme and how much you value this service. May I also remind you that there are no right or wrong answers, I am only interested in your opinion.**

Would you be willing to pay £80 per month for 25 years to fund paediatric cochlear implantation in the UK? *If yes, increase the amount and repeat until you reach an amount they are unwilling to pay. If no, reduce the amount until you find the highest amount they are willing to pay.*

Highest WTP = £ \_\_\_\_\_

Having said that you would be willing to pay £x per month for 25 years which equates to about £x. Would you be willing to pay, instead, a lump sum/one off payment towards a paediatric cochlear implantation programme in the UK? If so how much?

Highest lump sum WTP = £ \_\_\_\_\_

OR

This is quite a demanding question isn't it?, how about if I ask you would you be willing to pay a one off payment towards a paediatric cochlear implantation programme in the UK, is there an amount you would be willing to pay as a one off payment?

Highest WTP £ \_\_\_\_\_

You've said you are willing to pay £x what does this mean to you in terms of what you could buy with an equivalent sum of money? Now you've considered what else the money could have bought you do you want to change the amount you are willing to pay towards a paediatric cochlear implantation programme in the UK?

YES            NO

If yes, to what? £ \_\_\_\_\_

## Appendix B – WTA question

### Question 12:

Earlier I asked you to imagine that the NHS had recently announced that it would no longer pay for a paediatric cochlear implantation programme given current health resources available. I would now like you to consider this scenario again but this time imagine that the NHS does not give people the opportunity to pay extra money towards the NHS to keep the programme running. Therefore, the NHS realises that the loss of this programme will result in a loss of benefit to those children already implanted and to society at large since children that might be born deaf or become deaf in the future will be denied this intervention on the NHS.

I am interested in finding out how much this loss would mean to you. In order to get a comparable value I will be asking you to give a monetary figure of the level of compensation that would just compensate you for the loss of this programme. You will not receive this amount but it should represent the value you would be willing to accept if funding for the paediatric cochlear implantation programme in the UK was withdrawn.

**The NHS does provide cochlear implants. This question is simply a method of measuring how strongly you feel about the UK having a paediatric Cochlear implant programme and how much you value this service. May I also remind you that there are no right or wrong answers, I am only interested in your opinion.**

Would you be willing to accept £80 per month for 25 years as compensation if the NHS decided not to fund a paediatric cochlear implant programme? *If yes, decrease the amount and repeat until you reach the lowest amount they are unwilling to accept. If no, increase the amount until you find the lowest amount they are willing to accept.*

Lowest amount they are WTA £ \_\_\_\_\_;

Having said that you would be willing to accept £x per month for 25 years which equates to about £x. Would you be willing to accept, instead, a lump sum/one off payment as compensation if the NHS decided not to fund a paediatric cochlear implant programme? If so how much?

Highest lump sum WTA = £ \_\_\_\_\_

OR

This is quite a demanding question isn't it?, how about if I ask you would you be willing to accept a one off payment as compensation if the NHS decided not to fund a paediatric cochlear implant programme in the UK, is there an amount you would be willing to accept as a one off payment?

Highest WTA £ \_\_\_\_\_

You've said you are willing to accept £x what does this mean to you in terms of what you could buy with an equivalent sum of money? Now you've considered what can be bought with that money do you want to change the amount you are willing to accept if the NHS decided not to fund a paediatric cochlear implant programme in the UK?

YES            NO

If yes, to what? £ \_\_\_\_\_

<b>Appendix C: Socio-demographic characteristics of the sample</b>					
<b>Participants characteristics</b>	<b>No.s (%) Pre</b>	<b>No.s (%) Post</b>	<b>Participants characteristics</b>	<b>No.s (%) Pre</b>	<b>No.s (%) Post</b>
Sample size	26	136	<b>No. of implanted children:</b> 1 2	25 (96) 1 (4)	129 (95) 7 (5)
<b>Gender:</b> Female only Male only Both together	12 (46) 6 (23) 8 (31)	75 (55) 28 (21) 33 (24)	<b>Do you have private health care?</b> No Yes	16 (62) 10 (38)	82 (60) 54 (40)
<b>Age:</b> <b>Males</b> 25-34 35-44 45-54 55-64 over 65	11 (79) 2 (14) 1 (7) 2 (3) 2 (3)	10 (16) 40 (66) 7 (12) 2 (3) 2 (3)	<b>Asked WTP/A before?</b> No Yes	24 (92) 2 (8)	131 (96) 5 (4)
<b>Females (1 missing)</b> 16-24 25-34 35-44 45-54 55-64	1 (5) 6 (30) 10 (50) 2 (10) 1 (5)	3 (2) 33 (24) 55 (40) 15 (11) 1 (1)	<b>WTP total cost of implant or as much as you possibly can?</b> No Yes	18 (69) 8 (31)	81 (60) 55 (40)
<b>Income:</b> Less than £10,000 £10,000 – 20,000 £20,000 – 30,000 £30,000 – 40,000 £40,000 – 50,000 £50,000 – 60,000 £60,000 – 70,000 Greater than £70,000 Missing	2 (8) 6 (23) 4 (15) 6 (23) 3 (11) 3 (11) 1 (4) 0 (0) 1 (4)	16 (12) 24 (18) 34 (25) 19 (14) 9 (7) 12 (9) 4 (3) 17 (13) 1 (1)	<b>Have you ever considered alternatives to PCI?</b> No Yes	18 (69) 8 (31)	90 (66) 46 (34)
<b>Median age at leaving formal education (male/female) (years)</b>	16/16	16/17	<b>Child characteristics</b> <b>Gender:</b> Male Female	<b>No.s(%)</b> 11 (42) 15 (58)	<b>No.s (%)</b> 73 (54) 63 (46)
<b>Employment status:</b> <b>Males:</b> Missing Employed Unemployed Househusband Retired Other	1 (7) 11 (79) 0 2 (14) 0 0	0 (0) 45 (74) 1 (2) 2 (3) 3 (5) 10 (16)	<b>Onset:</b> Congenital Acquired/progressive	21 (81) 5 (19)	96 (71) 40 (29)
<b>Females</b> Employed Unemployed Housewife Student Retired Other	8 (40) 1 (5) 10 (50) 0 0 1 (5)	49 (45) 6 (6) 40 (37) 1 (1) 3 (3) 9 (8)	<b>School setting:</b> Mainstream Mainstream + unit School for the deaf Preschool Special schools Other e.g. home	10 (39) 6 (23) 2 (8) 8 (31) 0 0	50 (37) 57 (42) 15 (11) 8 (6) 4 (3) 2 (2)
			<b>Years with Implant</b> Preimplant Under 2 years >2 - <5 years 5 - >8 years 8 – 13 years	26 (100)	0 44 (32) 38 (28) 41 (30) 13 (10)
			<b>Mean HRQL/QOL gain</b>		0.38/0.39

## Appendix D

<i>Type of benefit</i>		<i>Percentage that report observing it</i>
I)	Access to environmental sounds and improved hearing	89.6%
II)	Educational benefits	85.7%
III)	Safety	83.1%
IV)	The range of options it potentially opens up	68.2%
V)	The low risk involved	24.7%
VI)	Psychosocial benefits for the child e.g. improved self-confidence and greater social integration	80.5%
VII)	The knowledge you've helped someone	35.1%
VIII)	The rewards families experience e.g. unexpected progress, greater involvement in a child's development etc.	77.3%

		WTP*	WTA*
Elicitation method answered	1) Bidding process (BP) alone	38.3% (41%)	11.1% (28.6%)
	2) Open ended (OE) alone	1.2% (1.3%)	6.8% (17.5%)
	3) BP + OE	53.7% (57.6%)	20.4% (52.4%)
	4) Neither BP or OE	6.8%	61.1%

\* Figures in brackets representing percentages of those who answer 1), 2) or 3).

		Sample size	Mean (£2001/2)	Mean difference	P-Value	Percentiles:		
						25	50 (median)	75
WTP Monthly	Post implant	127	£150	£15	P = 0.84	25	50	150
	Pre implant	22	£135			20	55	263
Lump sum WTP	Post implant	79	£25,832	£14,222	P = 0.71	1000	3000	10,000
	Pre implant	10	£11,610			500	4500	16,250
WTA monthly	Post implant	43	£254	-£231	P = 0.23	50	167	200
	Pre implant	8	£485			125	375	950
Lump sum WTA	Post implant	36	£197,492	-£415,758	P = 0.014	10,000	32,500	175,000
	Pre implant	8	£613,250			66,250	450,000	937,500

<i>Category</i>		<i>Percentage that state the reason</i>
I)	The amount reflects a reasonable/fair amount	14.7%
II)	The amount reflects a token/nominal amount	15.4%
III)	Found the question difficult to understand/answer	9.6%
IV)	The amount reflects the benefits of cochlear implantation	18.6%
V)	This is how much I perceive the service to cost	8.3%
VI)	How much I can afford to pay	61.5%
VII)	Other	3.5%

\* Adapted from Frew E, et al (2001) [31].



<b>Table 5: Factors influencing WTA bids obtained</b>	Percentage that state the reason (some state more than one reason)
<b>Category</b>	
1) to “cover the cost of continuing care privately”, “No idea how much, depends on the cost of finding support privately”	56%
2) Moral reasons e.g. “I’d feel guilty taking money”, “anything would be an insult”, “we’d move to the states”, “it’s a gift, accept whatever”	8%
3) Time e.g. “they’re just starting it – he’s just 2 years old and has the rest of his life”, “My child has already benefited a lot so don’t feel entitled to compensation”, “That’s not for me to decide – it’s up to him when he’s 18”, “fund it ourselves now”,	21%
4) “I’d want compensation for his loss of earnings”, “put him out of the employment market completely”, “how much would it devalue her life?”, “enough to buy a house”, “psychologically could affect the rest of his life”, “it’s an emotional, social thing as well”	8%
5) Protest e.g. “How can you put a price on that”, “there isn’t a sum of money that could compensate”, “difficult to put a price on it”, “wouldn’t want the money, want the service – the moneys not important, money can’t do everything”, “how long a piece of string”, “like taking someone’s life”, “not something you think about”, “how would anyone calculate such an amount”	35%
6) “Whatever I’d be willing to pay”	4%
7) “Fight for the programme to continue”, “raise money”, “Think beyond that to other people”	13%

<b>Table 6: Partial correlation coefficients between WTP/A and outcome, controlling for income</b>					
	HRQL gain	QOL gain	Most recent SIR score	Most recent CAP score	Has it met your expectations? *
WTP monthly	0.710	0.826	0.186	0.835	0.827
WTP lump sum	<b>0.022</b>	0.463	0.215	0.745	0.603
WTA monthly	0.264	0.056	0.215	0.156	0.781
WTA lump sum	0.571	0.080	0.903	0.304	0.576

\* where 1 was exceeded, 2 met and 3 below expectations.

<b>Table 7: Variables</b>	<b>Coefficients (standard error)</b>	<b>P Value</b>	<b>95% confidence interval</b>
Constant	3.849 (0.860)	0.000	(2.11, 5.59)
Income	0.212 (0.063)	<b>0.002</b>	(0.09, 0.34)
Benefits	0.853 (0.393)	<b>0.036</b>	(0.06, 1.65)
No. of implanted children	-0.453 (0.599)	0.455	(-1.67, 0.76)
Years child implanted	-2.097E-02 (0.68)	0.760	(-0.16, 0.12)
Mainstream or not (1 = mainstream, 0 = other)	-8.375E-02 (0.459)	0.856	(-1.01, 0.85)
Onset of child’s deafness (1 = acquired, 0 = congenital)	0.432 (0.394)	0.280	(-0.37, 1.23)
Gender of child implanted	0.135 (0.380)	0.723	(-0.63, 0.91)
HRQOL gain	-0.161 (0.459)	0.280	(-1.27, 0.95)
<b>R-Square</b>			<b>0.360</b>
<b>F –Statistic (significance)</b>			<b>2.603 (0.023)</b>

<b>Table 8: Variables</b>	<b>Coefficients (standard error)</b>	<b>P Value</b>	<b>95% confidence interval</b>
Constant	5.064 (0.777)	0.000	(3.52, 6.61)
Income	0.112 (0.51)	<b>0.030</b>	(0.01, 0.21)
Benefits	0.830 (0.259)	<b>0.002</b>	(0.32, 1.43)
No. of implanted children	-1.126 (0.689)	0.106	(-2.50, 0.24)
Years child implanted	-2.806E-02 (0.037)	0.452	(-0.10, 0.05)
Mainstream or not (1 = mainstream, 0 = other)	-0.209 (0.251)	0.407	(-0.71, 0.29)
Onset of child's deafness (1 = acquired, 0 = congenital)	0.269 (0.22)	0.225	(-0.17, 0.71)
Gender of child implanted	1.635E-02 (0.203)	0.936	(-0.39, 0.42)
QOL gain	-0.498 (0.493)	0.315	(-1.48, 0.48)
<b>R-Square</b>			<b>0.227</b>
<b>F –Statistic (significance)</b>			<b>3.080 (0.004)</b>

<b>Table 9: Variables</b>	<b>Coefficients (standard error)</b>	<b>P Value</b>	<b>95% confidence interval</b>
Constant	4.395 (0.609)	0.000	(3.19, 5.60)
Income	0.194 (0.051)	<b>0.000</b>	(0.09, 0.30)
Benefits	0.654 (0.262)	<b>0.014</b>	(0.13, 1.17)
No. of implanted children	-1.009 (0.473)	<b>0.035</b>	(-1.95, -0.07)
Years child implanted	-6.027E-02 (0.044)	0.178	(-0.15, 0.03)
Mainstream or not (1 = mainstream, 0 = other)	-5.731E-02 (0.283)	0.840	-0.62, 0.50)
Onset of child's deafness (1 = acquired, 0 = congenital)	5.163E-02 (0.241)	0.831	(-0.43, 0.53)
Gender of child implanted	-9.695E-03 (0.209)	0.963	(-0.42, 0.41)
Recent CAP score	3.202E-02 (0.092)	0.729	(-0.15, 0.21)
<b>R-Square</b>			<b>0.195</b>
<b>F –Statistic (significance)</b>			<b>3.291 (0.002)</b>

<b>Table 10: Variables</b>	<b>Coefficients (standard error)</b>	<b>P Value</b>	<b>95% confidence interval</b>
Constant	4.640 (0.575)	0.000	(3.50, 5.78)
Income	0.155 (0.054)	<b>0.005</b>	(0.05, 0.26)
Benefits	0.756 (0.285)	<b>0.009</b>	(0.19, 1.32)
No. of implanted children	-0.890 (0.475)	0.064	(-1.83, 0.05)
Years child implanted	-3.861E-02 (0.050)	0.437	(-0.14, 0.06)
Mainstream or not (1 = mainstream, 0 = other)	-9.021E-02 (0.285)	0.753	(-0.66, 0.48)
Onset of child's deafness (1 = acquired, 0 = congenital)	0.261 (0.243)	0.286	(-0.22, 0.74)
Gender of child implanted	0.114 (0.225)	0.614	(-0.33, 0.56)
Recent SIR score	-8.248E-02 (0.126)	0.513	(-0.33, 0.17)
<b>R-Square</b>			<b>0.183</b>
<b>F –Statistic (significance)</b>			<b>2.683 (0.010)</b>

## Appendix E

Figure 1: Proportion of individuals WTP for PCI as a function of bid (Monthly BP amount)

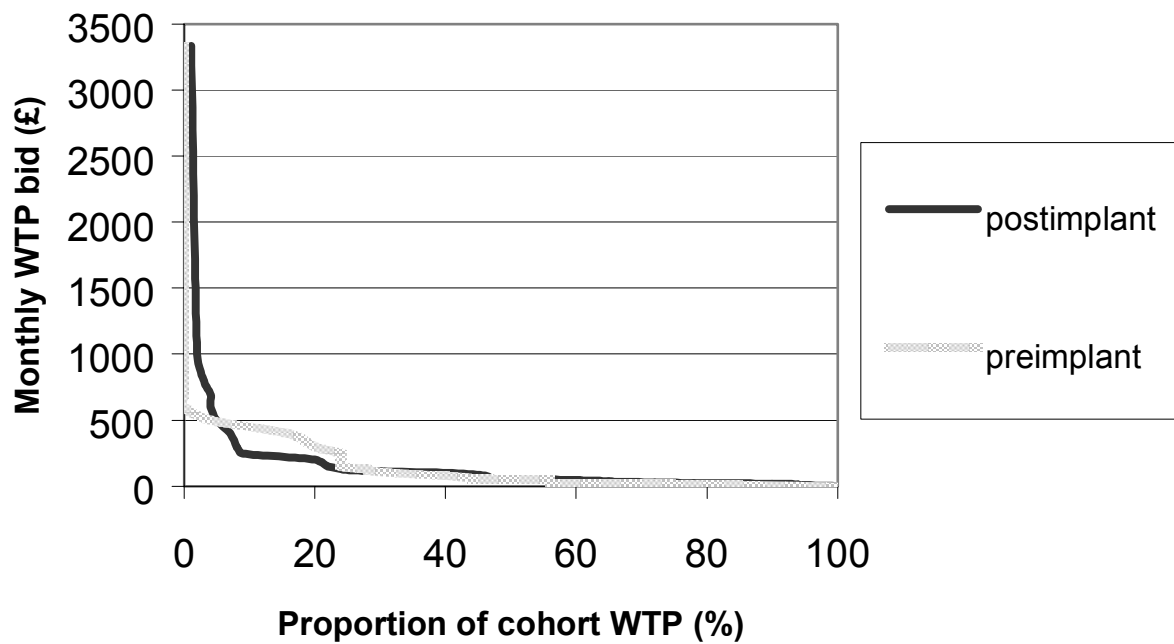


Figure 2: Proportion of individuals WTP for PCI as a function of bid (BP over 25 years compared to OE amount)

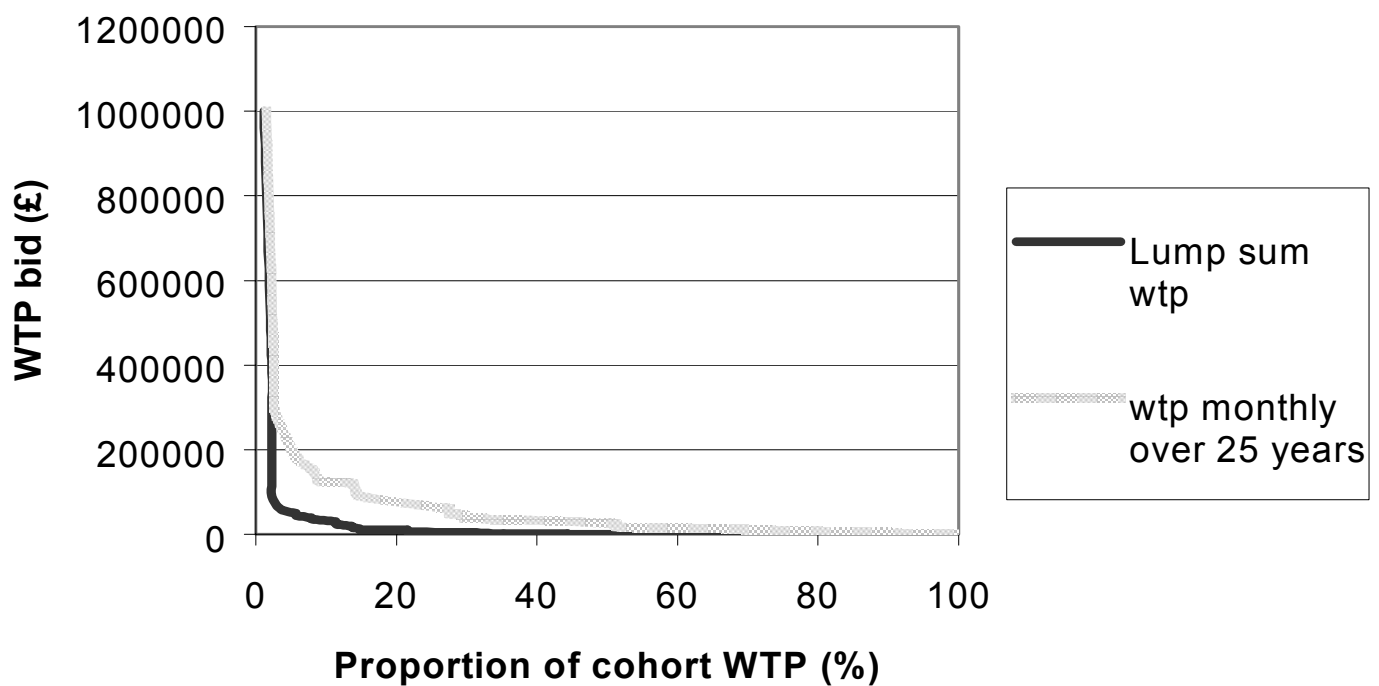


Figure 3: Proportion of individuals WTA for PCI as a function of bid (Monthly BP amount)

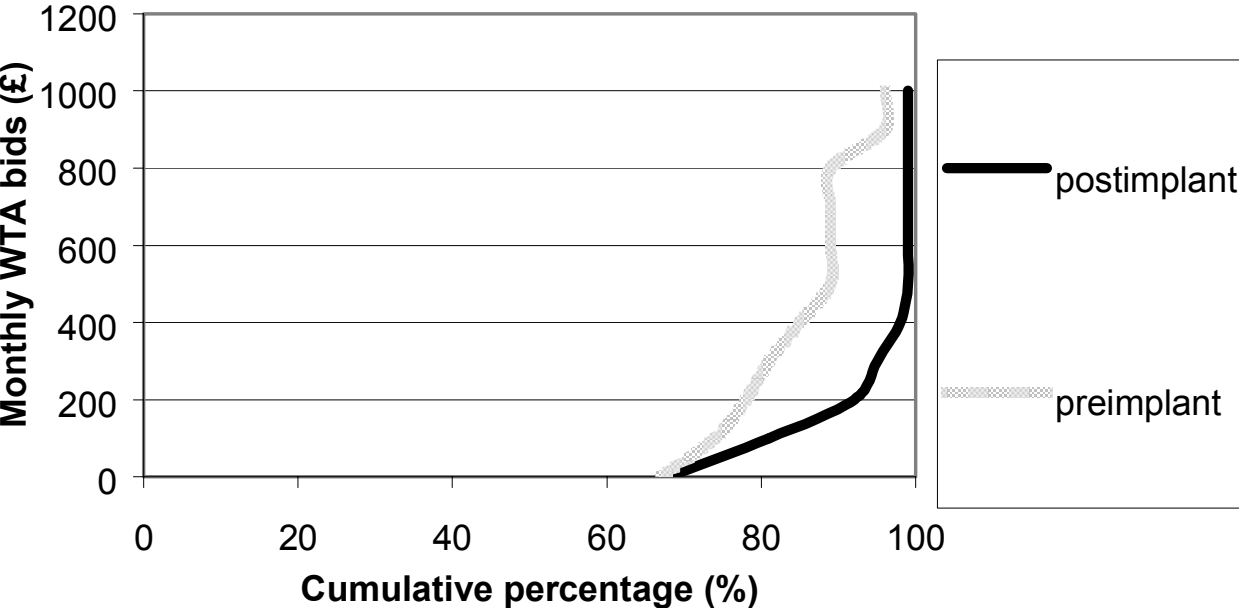


Figure 4: Hypothetical proportion of individuals WTP for PCI as a function of bid (General population)

