A review of the literature on the cost-utility of bilateral cochlear implantation

Francisco Javier Díez
UNED - Madrid, Spain
www.ia.uned.es/~fjdiez
www.cisiad.uned.es
Effectiveness vs. utility (1/2)

- **Effectiveness (in the strict sense)** = clinical outcomes
  - Objective: can be measured in a clinical setting or in a laboratory

- **Utility** = quality-adjusted life duration
  - Life duration is objective, but quality of life (QoL) if subjective
  - QoL is measured by asking a set of subjects (patients or general population)
  - Utility is measured in quality-adjusted life years (QALYs)

- **Effectiveness (in the broad sense)** includes both.
Effectiveness vs. utility (2/2)

The purpose of cost-utility analysis is to compare interventions for different purposes:

- drugs, vaccines, prostheses, diagnostic tests, educational programmes...

in order to decide which ones should be covered (reimbursed).

Nowadays the vast majority of cost-effectiveness analyses are cost-utility analyses.

But remember: studies aimed at informing public health policies are based on QoL estimates, not (directly) on clinical effectiveness.
How to measure the quality of life

- Visual analog scale (VAS)
  - does not measure quantitative preferences
  - cannot be used directly for cost-utility analysis

- Standard gamble
  [it has not been used for bilateral cochlear implantation]

- Time trade-off
  - “Do you prefer to live in state $s$ for 50 years or do you prefer to live with perfect health for $x$ years?” ($x < 50$)
Quality of life indexes (indices)

- Every index considers a reduced number of attributes (dimensions)
  - HUI-3 has 8 attributes: sight, hearing, ability to converse, ability to walk, manual dexterity, emotional status, cognitive ability and pain

- Every attribute has a limited number of states

- Every individual is characterized by a tuple of states
  - In HUI-3, it is an 8-tuple. Example: (4,5,5,6,4,3,4,5).

- A mathematical function maps each configuration onto a number
  - \( f(4,5,5,6,4,3,4,5) = 0.742 \).

- The function \( f \), specific for each index, is calibrated using a preference-elicitation method (standard gamble, time trade-off).
# Techniques used to measure quality of life associated to cochlear implantation

<table>
<thead>
<tr>
<th>Measures of preference</th>
<th>Other measures</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Direct measurement</strong></td>
<td>Visual analogue scale (VAS)</td>
</tr>
<tr>
<td>Time trade-off (TTO)</td>
<td></td>
</tr>
<tr>
<td>Standard gamble</td>
<td></td>
</tr>
<tr>
<td><strong>Generic questionnaires</strong></td>
<td></td>
</tr>
<tr>
<td>Health Utility Index (HUI-2)</td>
<td>Short-Form Health Survey with 36 dimensions (SF-36)</td>
</tr>
<tr>
<td>Health Utility Index (HUI-3)</td>
<td></td>
</tr>
<tr>
<td>EuroQoL (EQ-5D)</td>
<td>Assessment of Quality of Life (AQoL)</td>
</tr>
<tr>
<td>Short-Form Health Survey with 6 dimensions (SF-6D)</td>
<td></td>
</tr>
<tr>
<td><strong>Questionnaires for hearing loss and hearing aids</strong></td>
<td>Abbreviated Profile of Hearing Aid Benefit (APHAB)</td>
</tr>
<tr>
<td></td>
<td>Glasgow Benefit Inventory (GBI)</td>
</tr>
<tr>
<td></td>
<td>Glasgow Health Status Inventory (GHSI)</td>
</tr>
<tr>
<td></td>
<td>Hearing Participation Scale (HPS)</td>
</tr>
<tr>
<td></td>
<td>Profound Hearing Loss Answer Form (PIPHL)</td>
</tr>
<tr>
<td><strong>Questionnaires for cochlear implant users</strong></td>
<td>Chmiel-Sutton-Jenkins questionnaire for cochlear implant users</td>
</tr>
<tr>
<td></td>
<td>Index Relative Questionnaire Form (IRQF)</td>
</tr>
<tr>
<td></td>
<td>Nijmegen Cochlear Implantation Questionnaire (NClQ)</td>
</tr>
<tr>
<td></td>
<td>Patient Quality of Life Form (PQLF)</td>
</tr>
</tbody>
</table>
Increase in quality of life from unilateral to bilateral implantation

<table>
<thead>
<tr>
<th>Study</th>
<th>Target</th>
<th>Informants</th>
<th>Method</th>
<th>Increase in quality of life</th>
</tr>
</thead>
<tbody>
<tr>
<td>Summerfield et al. [2002]</td>
<td>adults</td>
<td>experts</td>
<td>TTO</td>
<td>0.031</td>
</tr>
<tr>
<td>Summerfield et al. [2006]</td>
<td>adults</td>
<td>bilaterally implanted patients</td>
<td>HUI-3</td>
<td>0.015 (initial)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>0.030 (adjusted)</td>
</tr>
<tr>
<td>Bichey and Miyamoto [2008]</td>
<td>adults and children</td>
<td>bilaterally implanted patients (or their parents)</td>
<td>HUI-3</td>
<td>0.11</td>
</tr>
<tr>
<td>Lovett [unpublished]</td>
<td>children</td>
<td>parents of bilaterally implanted children</td>
<td>VAS</td>
<td>0.33</td>
</tr>
<tr>
<td>Lovett et al. [2010]</td>
<td>children</td>
<td>parents of unilaterally and bilaterally implanted ch.</td>
<td>VAS</td>
<td>0.04</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>HUI-3</td>
<td>-0.003</td>
</tr>
<tr>
<td>Summerfield et al. [2010]</td>
<td>children</td>
<td>experts, students and parents of children with other disabilities</td>
<td>VAS</td>
<td>0.06</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>TTO</td>
<td>0.05</td>
</tr>
<tr>
<td>Sparreboom et al. [2012]</td>
<td>children</td>
<td>bilaterally implanted children</td>
<td>HUI-3</td>
<td>0.04</td>
</tr>
</tbody>
</table>
### Drawback of the TTO method

- Questions seem to be biased.
  - Psychological studies:

> Many experiments have shown the magnetic attraction of the status quo. In one, a group of people were randomly given one of two gifts of approximately the same value—half received a mug, the other half a Swiss chocolate bar. They were then told that they could easily exchange the gift they received for the other gift. While you might expect that about half would have wanted to make the exchange, only one in ten actually did. The status quo exerted its power even though it had been arbitrarily established only minutes before.

(Hammond, Keeny and Raiffa, 1998)
Framing of the questions

- “Do you prefer A or B?”
  - is neutral.
- “Let’s imagine you have A. Would you give it up for B?”
  - favors A.
- “Let’s imagine you have B. Would you give it up for A?”
  - favors B.
Estimates of the Cost-Effectiveness of Pediatric Bilateral Cochlear Implantation

Arthur Quentin Summerfield, Rosemary E. S. Lovett, Hannah Bellenger, and Georgina Batten

INTRODUCTION

This article reports estimates of the cost-effectiveness of providing bilateral cochlear implants to young deaf children. Although the cost data are specific to the healthcare system in the United Kingdom, the estimates of benefit and the methods for relating costs and benefits to decision criteria are relevant to all healthcare systems.

In many countries, healthcare policy makers have struggled to decide whether deaf children should receive bilateral or unilateral cochlear implants. In England and Wales, for example, the policy-making body, the National Institute for Health and Clinical Excellence (NICE), first published draft guidance that recommended bilateral implantation for newly diagnosed young deaf children but not for children who already possessed one implant (NICE 2007). A second draft guidance excluded bilateral implantation for the majority of deaf children, except in the context of research (NICE 2008a). The final guidance (NICE 2009), which determines policy in the health service in England and Wales, recommends bilateral implantation for newly diagnosed young deaf children and allows a second implant for some children who already have one implant.

NICE requires evidence that health technologies are safe, clinically effective, and cost-effective before they are adopted. Decision making about bilateral implantation for children was hampered by the absence of data on the additional quality of life associated with using two implants compared with one. These data are required to estimate the cost-effectiveness of bilateral implantation; that is, to determine whether the incre-
Estimation of incremental utility (questions)

Question 1 (VAS): “Mark your daughter’s QoL on this scale.”

Question 2 (time trade-off):

Question 2 • Now we would like you to think about your daughter’s quality of life in a different way. Remember, you are 33 years old. Imagine you will live for 50 more years until you are 83 years old. Now, imagine that you could give up some years of your own life in order for your child to have normal hearing. She would have normal hearing for the rest of her life. Imagine that the years you give up would be taken off the end of your life. This question does not measure whether you are a good or bad parent—it is simply a method of obtaining your judgment about how challenging this scenario would be for your daughter.

Please write the number of years that you would give up in this box:

I would give up this number of years:
Drawback of the HUI-3 method

Ø Effect of retrospective comparisons:
   • recall bias: people may overestimate the benefit of implantation
   • confounding factor: child’s maturation

Ø Lack of sensitivity: ceiling effect.
# Hearing states in the HUI-3

<table>
<thead>
<tr>
<th>HEARING</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Able to hear what is said in a group conversation with at least three other people, <strong>without a hearing aid</strong>.</td>
</tr>
<tr>
<td>2</td>
<td>Able to hear what is said in a conversation with one other person in a quiet room <strong>without a hearing aid</strong>, but requires a hearing aid to hear what is said in a group conversation with at least three other people.</td>
</tr>
<tr>
<td>3</td>
<td>Able to hear what is said in a conversation with one other person in a quiet room <strong>with a hearing aid</strong>, and able to hear what is said in a group conversation with at least three other people, with a hearing aid.</td>
</tr>
<tr>
<td>4</td>
<td>Able to hear what is said in a conversation with one other person in a quiet room, <strong>without a hearing aid</strong>, but unable to hear what is said in a group conversation with at least three other people even with a hearing aid.</td>
</tr>
<tr>
<td>5</td>
<td>Able to hear what is said in a conversation with one other person in a quiet room <strong>with a hearing aid</strong>, but unable to hear what is said in a group conversation with at least three other people, even with a hearing aid.</td>
</tr>
<tr>
<td>6</td>
<td>Unable to hear at all.</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>SPEECH</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Able to be understood completely when speaking with strangers or friends.</td>
</tr>
<tr>
<td>2</td>
<td>Able to be understood partially when speaking with strangers but able to be understood completely when speaking with people who know me.</td>
</tr>
</tbody>
</table>
## Increase in quality of life from unilateral to bilateral implantation

<table>
<thead>
<tr>
<th>Study</th>
<th>Target</th>
<th>Informants</th>
<th>Method</th>
<th>Increase in quality of life</th>
</tr>
</thead>
<tbody>
<tr>
<td>Summerfield et al. [2002]</td>
<td>adults</td>
<td>experts</td>
<td>TTO</td>
<td>0.031</td>
</tr>
<tr>
<td>Summerfield et al. [2006]</td>
<td>adults</td>
<td>bilaterally implanted patients</td>
<td>HUI-3</td>
<td>0.015 (initial) 0.030 (adjusted)</td>
</tr>
<tr>
<td>Bichey and Miyamoto [2008]</td>
<td>adults and children</td>
<td>bilaterally implanted patients (or their parents)</td>
<td>HUI-3</td>
<td>0.11</td>
</tr>
<tr>
<td>Lovett [unpublished]</td>
<td>children</td>
<td>parents of bilaterally implanted children</td>
<td>VAS</td>
<td>0.33</td>
</tr>
<tr>
<td>Lovett et al. [2010]</td>
<td>children</td>
<td>parents of unilaterally and bilaterally implanted ch.</td>
<td>VAS</td>
<td>0.04</td>
</tr>
<tr>
<td>Summerfield et al. [2010]</td>
<td>children</td>
<td>experts, students and parents of children with other disabilities</td>
<td>VAS</td>
<td>0.06</td>
</tr>
<tr>
<td>Sparreboom et al. [2012]</td>
<td>children</td>
<td>bilaterally implanted children</td>
<td>HUI-3</td>
<td>0.04</td>
</tr>
</tbody>
</table>
### Cost-effectiveness analyses of bilateral CI

<table>
<thead>
<tr>
<th>Study</th>
<th>Country</th>
<th>Source of utilities</th>
<th>Population</th>
<th>ICER</th>
</tr>
</thead>
<tbody>
<tr>
<td>Summerfield et al., 2002</td>
<td>United Kingdom</td>
<td>own data</td>
<td>adults</td>
<td>£61,734 / QALY</td>
</tr>
<tr>
<td>Summerfield et al., 2003</td>
<td>United Kingdom</td>
<td>Summerfield et al., 2002</td>
<td>adults</td>
<td><strong>£100,000 / QALY</strong></td>
</tr>
<tr>
<td>Cochlear Europe Ltd. (submitted to NICE)</td>
<td>United Kingdom</td>
<td>projection of data</td>
<td>adults</td>
<td>£32,909 / QALY</td>
</tr>
<tr>
<td>Bond et al., 2007, 2009</td>
<td>United Kingdom</td>
<td>Summerfield et al., 2006</td>
<td>adults</td>
<td>£49,559 / QALY</td>
</tr>
<tr>
<td>Bond et al., 2007, 2009</td>
<td>United Kingdom</td>
<td>Summerfield et al., 2006</td>
<td>children</td>
<td>£40,400 / QALY</td>
</tr>
<tr>
<td>L-Pedraza Gómez et al., 2007</td>
<td>Spain</td>
<td>Summerfield et al., 2002</td>
<td>adults</td>
<td>€53,018 / QALY</td>
</tr>
<tr>
<td>Bond et al., 2007, 2009</td>
<td>United Kingdom</td>
<td>own data</td>
<td>adults and children</td>
<td>$2,187 / QALY</td>
</tr>
</tbody>
</table>

Huge differences are mainly due to discordant estimations of QoL increase.
Conclusions

- Most studies have underestimated the incremental utility of BCI, i.e., the gain in quality of life from unilateral to bilateral CI.

  ⇒ Overestimation of the cost-utility ratio.

- Those studies have delayed the approval of BCI in several countries.

- Further studies are necessary.
On-going study: CUA of BCI in Spain

- Goal: prove that BCI is cost-effective in Spain and in other countries for children (and also for many adults?)

- Estimation of the quality-of-life gain
  - Almost 500,000 people will be invited to participate, including hundreds of users of unilateral and bilateral CI.
  - Analysis of psychological biases.
  - Purpose: obtain more reliable measurements.
  - The data will be publicly available.

- Markov model
  - Built using new artificial intelligence methods.
  - The model will be publicly available.
If you wish to stay informed about this study, please write to:

fjdiez@dia.uned.es

Your feedback will be highly appreciated.

Thank you very much!