Cost-benefit analysis for the evaluation of new genetic tests: a pilot feasibility study

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Overview

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Inherited eye conditions
Cost-benefit analysis

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Contingent valuation method
Qualitative research

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Qualitative findings

Conclusions
How useful is CVM?
CBA’s potential role in economic evaluation
Background

Economic evaluation

“the comparative analysis of alternative courses of action in terms of both their costs and consequences”

Maximisation of health with scarce resources

Cost-effectiveness analysis (CEA)

CEA

Quality Adjusted Life Years (QALYs)

EQ-5D-3L: (mobility, self-care, usual activities, pain/discomfort, anxiety/depression)

Incremental cost-effectiveness ratio (ICER)

\[
\text{ICER} = \frac{(\text{Cost } a - \text{Cost } b)}{(\text{QALY } a - \text{QALY } b)}
\]

\[\text{ICER} = \text{incremental cost per additional QALY}\]

NICE threshold of £20 to 30K per QALY
Retinal dystrophy & genetic testing

Genetic testing
Meets MRC criteria for being a complex intervention
Often delivered in context of clinical genetics service
Outcomes can ‘go beyond’ health
Externalities

Inherited eye conditions
Retinal dystrophy / retinitis pigmentosa (RP)
Group of inherited eye conditions
No effective treatment or cure
1 in 3500 prevalence

Genetic tests for RP
Can provide definitive diagnosis of RP type
More detailed, reliable prognostic information
Reveals likely inheritance patterns
Cascade testing

Next Generation Sequencing
Quicker and cheaper: many genes can be examined simultaneously
Whole genome testing
Cost-benefit analysis
An alternative evaluative framework

CBA approaches

Based on individual preferences
Assumption: individuals maximise utility
Others preferences do not count
Unconcerned with equitable distribution of resources
Concerned with establishing the net benefits of projects

Typical to convert all costs and benefits to monetary values
Problematic to value resources without market prices
Individuals’ willingness-to-pay has instead been used

Contingent Valuation Method has been used to this effect
It is controversial – especially in healthcare
Aim

To identify if, and how, cost-benefit analysis could be used in the evaluation of new genetic tests to diagnose inherited eye conditions
Methods:
Contingent valuation method

- Sample
  - Two groups recruited through purposive sampling
  - Group A with prior knowledge of RP
  - Group B without prior knowledge of RP

- Hypothetical scenarios developed and piloted
  - Emailed or given to participants ahead of interview
  - Asked to imagine a suspected new diagnosis of RP
  - In a situation where healthcare was purchased
  - First asked if they would seek genetic counselling (GC)
  - Then asked if they would want a genetic test in addition to counselling (GC&T)

- Bidding game
  - Asked how much they would pay for GC and GC&T
  - Reminded to keep in mind their actual budget constraints
  - Iterative bidding game based on one of four randomly selected starting points
  - Open-ended question on maximum WTP on completion of bidding game
Methods:
Qualitative research

Objectives
To understand the reasons behind the WTP values
To provide evidence on validity of WTP values

Data collection
Semi-structured telephone/face-to-face interviews
Perceived (dis)benefits of genetics services
Reasons behind the WTP values
Demographics & financial information

Analysis
Framework approach*
Data coded - framework developed - re-analysis

Results:
Sample characteristics

Female 65% Group A=60% Group B=70%
64% aged under 45 years: Group A=48% Group B=80%

n=52

Group A n=25
Group B n=27

Income bands (£ per year):
- 5k to 12.49k
- 12.5k to 24.9k
- 25k to 39.9k
- 40k to 54.9k
- 55k to 74.9k
- 75k and over

Number of participants: 0, 2, 3, 5, 6, 7, 8
**Results:**

**Stated demand**

- **Counselling:**
  - 92% (48/52) of participants would want counselling
  - 96% (24/25) of Group A participants would want counselling
  - 92% (23/25) of Group A participants would want genetic testing
  - 81% (22/27) of Group B participants would want genetic testing

![Bar chart showing % want counselling and % want testing by group and total](chart.png)
## Results:

### WTP values

<table>
<thead>
<tr>
<th>Group</th>
<th>WTP value for genetic counselling</th>
<th>WTP value for genetic counselling &amp; testing</th>
</tr>
</thead>
<tbody>
<tr>
<td>All respondents</td>
<td>£275</td>
<td>£899.50</td>
</tr>
<tr>
<td>All except zero WTP responses</td>
<td>£275</td>
<td>£899.50</td>
</tr>
<tr>
<td>n=52</td>
<td>n=52</td>
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</tbody>
</table>

### Median

- **Group A**
  - Median: £274.50
  - IQR: £149.50 to £899.50
  - Mean: £612.02
  - 95%CI: £237.56 to £986.48
- **Group B**
  - Median: £149.50
  - IQR: £37 to £299.50
  - Mean: £202.35
  - 95%CI: £114.40 to £290.30

### Total

- Median: £224.50
- IQR: £99.50 to £524.50
- Mean: £399.31
- 95%CI: £211.47 to £587.14

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**Results**

Overall median WTP for counselling = £224.50

Overall median WTP for counselling plus testing was £524.50

£899.50 (Group A) Vs £275 (Group B)
# Results:
## Qualitative findings

### Reasons for wanting genetic tests

<table>
<thead>
<tr>
<th>Themes</th>
<th>Illustrative Quotes</th>
<th>Respondent</th>
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<tbody>
<tr>
<td><strong>Value of information</strong></td>
<td>...how it might affect my family in the future, how it might affect members of my family now, but also just possibly in terms of prognosis. If you've got all the facts then you can plan for your future, you can build coping strategies, you can organise your life down the line based on real facts rather than pure supposition.</td>
<td>Group A, Female, aged 25-34</td>
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<td></td>
<td></td>
<td>Group A, Male, aged 55-64</td>
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<tr>
<td><strong>Acknowledgement of value to others</strong></td>
<td>But I suppose if I don't want children I'm not worried about the inheritance and I'm not going to be passing it on to anyone. I can understand how, if you did have children, it would be different, but I think for me, knowing that it was a genetic disorder or not wouldn't change that I had that disorder or didn't have it.</td>
<td>Group B, Female, aged 18-24</td>
</tr>
<tr>
<td></td>
<td>I think the testing is important, not just for myself but just for researching into the condition. The more that's learnt about it, the more people can be helped.</td>
<td>Group A, Female, aged 35-44</td>
</tr>
<tr>
<td><strong>Influence on well-being</strong></td>
<td>...it's still something you want to investigate to try and put your mind at rest. So I think that's why it's valuable to you...</td>
<td>Group A, Female, aged 35-44</td>
</tr>
<tr>
<td></td>
<td>...you'd sort of feel it's certain if you did get a genetic test result, whereas somebody telling you 'we understand from your family history and things and we know from other people affected by this same condition that's it most likely to be this' but they can't say 100% and that can leave you with that level of uncertainty which is maybe a little bit difficult to live with long term.</td>
<td>Group B, Female, aged 25-34</td>
</tr>
</tbody>
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Discussion

Usefulness of CVM

WTP values were elicited from all participants
All participants stated that they understood scenarios

CVM design issues

Bigger, more representative sample required
Payment vehicle & non-use values

Next steps

Further in-depth analysis of qualitative data
Regression analysis to understand drivers of WTP values

Conclusion

Potential for CBA to inform decision-making
CVM looks to be useful in eliciting £ values for genetic tests
Fundamental shift in decision-making processes would be required
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