Crowdsourcing Utility Estimation for Spina Bifida in the General Population

Ruiyang Jiang, Brian Inouye, Hsin-Hsiao Wang, J. Todd Purves, John S. Wiener, and Jonathan C. Routh
Disclosures and Disclaimers

• We do not intend to discuss any commercial products or services
• We do not intend to discuss non-FDA approved uses of products or providers of services
• This project is supported by the National Institute of Diabetes and Digestive and Kidney Diseases (NIDDK). The funding sources had no role in the collection, analysis or interpretation of data; in the writing of the manuscript; or in the decision to submit the manuscript for publication. The findings and conclusions in this report are those of the authors.
Introduction

• Spina Bifida (SB)
  • Most common permanently disabling birth defect
  • Range of presentations, management options
  • Diverse management options/patient preferences

• Cost utility analysis (CUA)
  • useful to estimate & compare treatment options
  • Requires reliable “utility” estimates
So what is a “utility”? 

• “Utility” quantifies health state impact on lifestyle, satisfaction
  • 0 = death
  • 1 = perfect health
  • Disease impact and QOL represented as a numerical value between 0 and 1

• Current SB utility scores obtained via traditional interview
  • Range 0.74 – 0.84
  • Traditional approach → time consuming, expensive
  • Newer methods use validated online surveys\textsuperscript{1-3}
  • Unclear if these methods translate to chronic states such as SB

Objectives

• To determine the feasibility of utility estimation for SB
• To estimate utility values for an archetypal child with SB via a validated online tool

Hypothesis → SB utility via online survey will be feasible, and derived values will be similar to SB utility via traditional interview
Methods

- Cross-sectional survey
- Participants recruited via Amazon.com’s Mechanical Turk (AMT)
  - Volunteers register to complete tasks
  - Each worker is assigned an ID
  - Compensated $0.50 per task
    - Typical range for AMT: 10¢ - $1
- All participants anonymous
Methods

• Exclusion criteria
  • Age < 18 years
  • Residence outside U.S
  • Lack English fluency

• Participants were included even if unfamiliar with SB, no children, not married
  • Utility estimate based on community perspective
Methods

• Background provided to respondents
  • Written text described SB
  • 3-minute CDC video about SB

• Survey questionnaire
  • Demographic questions
  • SB time-trade-off assessment
Methods

- Participants were asked to define effect of SB on him/her, his/her child, family
- “Time Trade-Off” method
  - Whether they were willing to trade time from end of combined family dyad (parent + child) to get their child back to perfect health
Imagine that you have a 6-year-old child with spina bifida as just described. Now imagine that a fortune-teller can see the next 10 years of your future. She sees that your child will live with this condition for at least the next 10 years. However, she can make you the following offer: instead of living for 10 years with spina bifida, your child will be in perfect health, but, your and your child’s life will be somewhat shorter. In other words, you and your child will have to give up a certain amount of time so that your child will live in perfect health instead of your child’s current state of imperfect health. Your child’s total lifespan will be unchanged.

So, instead of spending 10 years with spina bifida, the fortune-teller makes you an offer where you and your child have to give up _______ years _______ months, but in exchange your child will spend _______ years _______ months in perfect health. Would you accept this offer?

If “yes”, add time to offer.
If “no”, subtract time from offer.
Methods

- 10-year timeframe used
- Variable time spent in “perfect health” compared to 10 years in disease state
- For responses from 1-9 year TTO, health durations changed in 1-year intervals
  - Could be narrowed to 0-365 days
- Variables include age, gender, marital status, race, prior SB knowledge etc. were collected
Results

• Total participants = 591
• # completed task = 503 (85% response rate)
• ~ 50% male/female respondents
• 46% have children
• Vast majority do not have SB or child with SB
Results – Respondent Demographics

- Mean age 33.6 y
- 51% male
- 77% Caucasian
- 38% married
- 46% parents
- 90% at least some college experience
- 70% household income <$50,000
- 80% had heard of SB
- 15% had a friend with SB, spinal cord injury, or paralysis
- 51% had heard of cost-utility analyses
Results – Utility value

• Mean utility value: 0.85

• Comparable conditions with this value:
  • Pneumonia hospitalization
  • Brain contusion, adequately treated with craniotomy
  • Resistant hypertension
Results – Bivariate analysis

• Prior knowledge of **NTD** \(p=0.007\), **SB** \(p=0.001\) associated with increased utility value

• Having SB, having a child with SB, and having a friend with SB not associated with changes in utility estimates (all \(p>0.1\))
## Results – Multivariate Analysis

<table>
<thead>
<tr>
<th>Variable</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Prior NTD Knowledge</td>
<td>0.32</td>
</tr>
<tr>
<td>Race</td>
<td>0.14</td>
</tr>
<tr>
<td>Marital Status</td>
<td>0.19</td>
</tr>
<tr>
<td>Highest Education</td>
<td>0.1</td>
</tr>
<tr>
<td>Self Condition</td>
<td>0.74</td>
</tr>
<tr>
<td>Kid Condition</td>
<td>0.91</td>
</tr>
<tr>
<td>Friend Condition</td>
<td>0.49</td>
</tr>
</tbody>
</table>
Limitations

• Sample not necessarily representative of U.S. population
  • AMT more representative than other samples
• Estimate based on dyad perspective
• Estimate overall SB utility state
  • Does not distinguish SB with bladder stone vs. SB with augmentation, etc.
• Age limit > 18 years
Conclusions

• SB utility value = 0.85
• AMT is an efficient, valid, and reliable online tool
• AMT may be a reliable tool for future SB