Value of Knowing Risk of Developing Neurodegenerative Disease: A Discrete Choice Experiment

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Background

- Additional value can be gained from genetic tests through the reassurance of a negative result or ability to plan from a positive result[1-3]
- This is referred to as the "value of knowing" in the ISPOR value flower[4]
- This concept has been measured for several genetic and diagnostic tests, but not for genetic tests for neurodegenerative disease[5-9]

Objective

To explore the value of knowing the results of a genetic test for a neurodegenerative disease from the perspective of patients or family members, and how this varies based on attributes of the test, characteristics of the disease, and characteristics of the individual.

Table 1: Discrete Choice Experiment (DCE) Attributes and Hypothetical Scenarios Presented to Respondents

DCE Attribute	Levels
Test Cost (patient "out of pocket")	$50, $250, $1,000
Test Type	Invasive, Non-Invasive
Chance of False Negative Test	0%, 5%, 20%
Chance of False Positive Test	0%, 5%, 20%
Hypothetical Scenario	**Levels**
Severity of Disease	Fatal, High, Moderate, Low
Perspective (Who was tested)	Self, Family Member, Child
Perceived Risk of Developing Disease	High, Low, Uncertain
Treatment Availability and Effectiveness	None, Temporary Relief, Permanent Relief, Improved Life Expectancy, Cure

Scan the QR code to access the full text of the survey

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Methods

Survey

- Participants were recruited from NORC AmeriSpeak, a national probability sample reflective of the general US population[10]
- Demographic and clinical characteristics were collected including prior experience with genetic tests
- Participants were surveyed with a discrete choice experiment (DCE).[11] Respondents were asked to choose between 2 hypothetical genetic tests for neurodegenerative disease, which were described using 4 attributes **(see Table 1).** These were varied on a random basis with each choice task.
- A genetic counselor and neuropsychologist were consulted to evaluate the face validity of the survey (see Acknowledgements).
- Approved by Tufts Medical Center Institutional Review Board (IRB) (STUDY00004150)

Statistical Model

- Responses were evaluated using a generalized multinomial logit (G-MNL) model to predict average willingness to pay (WTP) for attributes of the genetic test.[12-14] This result we refer to as the "base case WTP"
- The interactive models evaluated how alternative hypothetical scenarios, experiences, and demographics influence WTP for test and attributes of test. We report these results versus the base case.

Figure 1: Willingness to Pay (WTP) Changes Relative to Base Case WTP

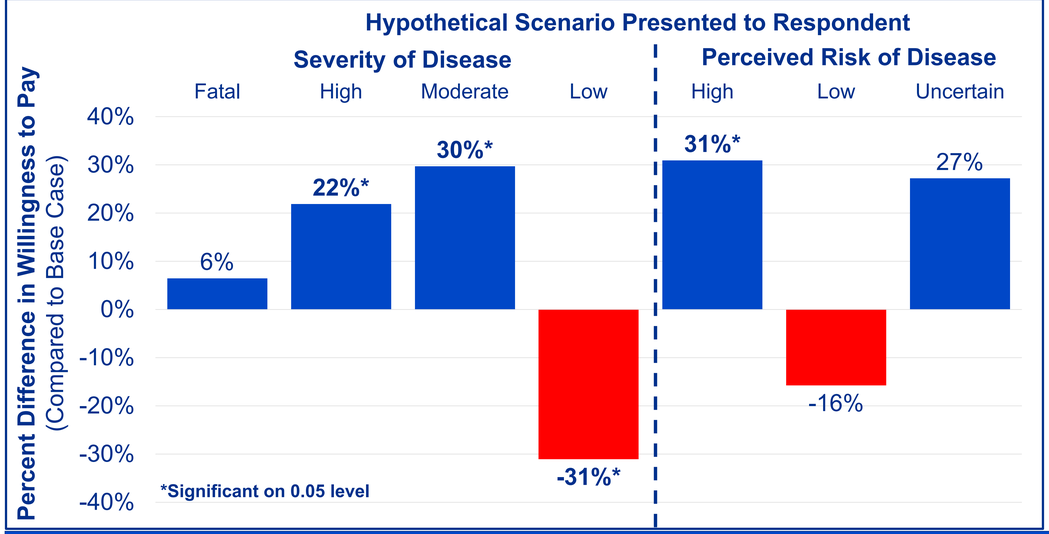

Table 3: Groups with Higher Willingness to Pay (WTP)

WTP more for a test	WTP more for improved accuracy
Scenarios	**Scenarios**
Severity of disease is high or moderate	Severity of disease is fatal
Risk of developing disease is high	Risk of developing disease is high
Treatment provides temporary relief	
Experiences	**Experiences**
Themselves or a family member has had a genetic test	
Had a genetic test to find out risk or learn family history	
Themselves or a family member has a neurodegenerative disease	Relative has a neurodegenerative disease

Results

- 1,034 respondents completed the survey
- Demographics of respondents were representative of the US population
- The probability of a false positive or negative had a strong influence on the WTP, as did invasiveness **(see Table 2).**
- WTP was influenced by differences in disease severity, perceived disease risk, availability of treatment, and prior experience with genetic tests or neurodegenerative disease influence **(see Table 3, Figure 1)**

Limitations

- 0% accuracy level for chance of false negative/positive is not realistic for genetic tests, this level was used to facilitate ease of comprehension and to allow incremental effects of each percentage point change to be computed.
- The value of genetic testing is a complex, individual decision. Genetic counseling with a board certified, neurogenetic counselor prior to genetic testing is strongly recommended.

Key Takeaways

- Participants facing these hypothetical options placed higher values on genetic tests with greater accuracy.
- Participants also placed higher value on a genetic test when the severity of disease was has high or moderate, or they already perceive their risk of developing the disease as high (e.g., have family history)
- People are WTP more for genetic tests for neurodegenerative disease when they have previous experience with genetic tests or neurodegenerative disease

Table 2: G-MNL Results

Attribute	Average WTP
Test*	$2,865**
Invasive (vs non-invasive)	-$91**
0% chance of false negative (vs 5%) (per % point)	$109**
20% chance of false negative (vs 5%) (per % point)	-$50**
0% chance of false positive (vs 5%) (per % point)	$90**
20% chance of false positive (vs 5%) (per % point)	-$41**

*Value determined by levels of cost attribute, serves as a comparison
**Significant on 0.05 level

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