Patient Preferences in Subglottic Stenosis Treatment: A Discrete Choice Experiment

Matthew R. Naunheim, MD, MBA1,2, Margaret L. Naunheim, MD3, Vinay K. Rathi, MD1,2, Ramon A. Franco, MD1,2, Mark G. Shrime, MD, MPH, PhD1,2,4, and Phillip C. Song, MD1,2

Abstract

Objectives. Subglottic stenosis can be addressed with several different surgical techniques, but patient preferences for these treatment modalities are poorly understood. Economic methods are increasingly being used to understand how patients make decisions. The objective of this pilot study was to assess preferences in subglottic stenosis treatment using patient-centric stated preference techniques.

Study Design. Discrete choice experiment (DCE).

Setting. Academic research facility.

Subjects and Methods. A computer-based DCE was administered in a monitored setting to volunteers from the general population. Signs and symptoms of subglottic stenosis were described, and participants were asked to imagine they had subglottic stenosis. Hypothetical treatments were offered, with 5 systematically varied attributes: need for external incision, length of hospital stay, postoperative voice quality, likelihood of repeat procedures, and risk of complication. A conditional logistic model was used to assess the relative attribute importance.

Results. In total, 162 participants were included. Attributes with the greatest impact on decision making included potential need for repeat procedures (importance 30.2%; \(P < .001\)), amount of operative risk (importance 28.1%; \(P < .001\)), and postoperative voice quality (importance 27.7%; \(P < .001\)), whereas presence of incision (importance 5.0%; \(P = .001\)) was less important, and hospital stay was not (importance 9.0%; \(P = .089\)). Based on aggregate responses for these attributes, the model demonstrated that most participants (80.4%) would prefer endoscopic surgery for subglottic stenosis as opposed to open tracheal resection (19.6%).

Conclusion. In this pilot population, most participants preferred voice-sparing, low-risk procedures as treatment for subglottic stenosis, consistent with an endoscopic approach, even if multiple procedures were required.

Keywords

patient preferences, utility, outcomes research, otolaryngology, contingent valuation, willingness to pay, stated preference methods, conjoint analysis, discrete choice experiment

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Subglottic stenosis (SGS) is a condition characterized by airway narrowing at the level of the cricoid and proximal trachea, and it represents a therapeutic challenge for otolaryngologists. Management options include medical therapies, like topical or injected medications, as well as open and endoscopic surgery. When deciding among surgical options, patients and doctors must weigh the risks and benefits of each therapeutic. Cricotracheal resection with primary anastomosis or laryngotracheal reconstruction with grafting are reported to have high success rates, but complications including anastomotic failure, wound infection, and postoperative voice changes can be significant.1-3 Endoscopic techniques, including laser- and balloon-assisted techniques for widening the subglottic airway, can be successful and cost-effective but are often not definitive and require multiple procedures over the patient’s lifetime.3-6

Although the decision to undergo airway surgery can be daunting for patients, patient preferences for surgical treatment...
have not been well studied in the context of subglottic stenosis. For example, do patients prefer a single surgical procedure that offered a high likelihood of cure if it also meant a higher risk of surgical complications? Would patients strongly prefer to avoid a long hospital stay or neck incision, or are they averse to the need for multiple surgical procedures?

Stated preference techniques such as discrete choice experiments (DCEs) are a widely accepted means of measuring patient-centered assessments of preference and value,\(^7,8\) and they can help answer these questions. This survey-based statistical method has traditionally been used in market research for determining how people value attributes of specific products and services.\(^9\) In marketing, this technique asks consumers to choose between several products, each of which has different attributes, and evaluates the implicit valuations in these choices to model consumer decision making and product design. In medicine, DCEs can be used to evaluate health programs, products, and services.\(^7\) In recent years, these techniques have been used to understand patient preferences within otolaryngology.\(^10-15\)

In this study, we conducted a survey-based DCE to elicit preferences for treatment for the surgical therapies of subglottic stenosis.

**Methods**

This study was approved by the Institutional Review Board at the Massachusetts Eye and Ear Infirmary. We used published guidelines in the field of stated preference research to guide the design of this study.\(^8,16,17\)

**Attribute and Level Determination**

Treatment attributes pertinent to surgery for subglottic stenosis were first compiled by literature review and then subjected to review from experts in the field of otolaryngology and public health.\(^4,5,18-21\) Using a broad list of attributes, qualitative interviews were then conducted with 15 patients presenting for ear, nose, throat, head, or neck complaints. Patients were first asked open-ended questions regarding what factors are important when considering surgery, and they were then asked to discuss the most important factors included on the attribute list. These results were compiled and again reviewed by the team of experts. Five attributes were selected, and levels within these attributes were determined with reference to the literature review. The final list of attributes and levels is in Table 1. We hypothesized that, in general, patients would prefer no incision, shorter hospital stay, better voice outcomes, fewer repeated procedures, and fewer complications.

**Experimental Design and Pilot Testing**

Hypothetical treatment scenarios, varying along the 5 pertinent attributes (need for incision, length of hospital stay after procedure, voice quality after surgery, need for repeat procedures, and risk of complications) were created using Discover Software (Sawtooth Software, Orem, Utah). A partial profile design was used to reduce response burden on participants while balancing the representation of attributes and levels. Fourteen discrete choice sets with 3 profiles each were included in each survey. An example is provided in Figure 1.

Participants were given descriptions of subglottic stenosis, including anatomical diagrams, key clinical outcomes (voice, airway, quality of life, and mortality), and details on the surgical management of this disease. Other variables were collected in separate questions, including sex, age, race, income, highest education level, marriage status, number of children, history of subglottic stenosis, and history of any chronic disease; all of these additional variables were self-reported. Stated comprehension and consequentiaility (the belief by participants that their responses would be considered by researchers) were included as measures of internal validity.\(^22\)

The survey was initially piloted in 10 patients presenting to a tertiary laryngology referral, and responses were elicited to assess understanding through both directed questions and open-ended comments. Nine of 10 demonstrated full understanding of the experimental design, and responses

<table>
<thead>
<tr>
<th>Attribute</th>
<th>Metric</th>
<th>Levels</th>
</tr>
</thead>
<tbody>
<tr>
<td>Incision</td>
<td>Whether an incision (cut) is made in the neck</td>
<td>Incision, No incision</td>
</tr>
<tr>
<td>Hospital stay</td>
<td>Length of inpatient stay</td>
<td>0, 3, 6, or 9 days</td>
</tr>
<tr>
<td>Voice</td>
<td>Descriptive voice quality after surgery</td>
<td>Normal, Soft, Very soft, lower pitched</td>
</tr>
<tr>
<td>Repeat procedures</td>
<td>Number of repeat procedures required to treat breathing</td>
<td>Only 1 procedure ever</td>
</tr>
<tr>
<td></td>
<td>Repeat procedure every 5 years</td>
<td>Repeat procedure every 2 years</td>
</tr>
<tr>
<td></td>
<td>Repeat procedure every 6 months</td>
<td>Repeat procedure every 6 months</td>
</tr>
<tr>
<td>Complications</td>
<td>Risk of having a serious complication after surgery</td>
<td>1%, 5%, 10%, or 20%</td>
</tr>
</tbody>
</table>

**Table 1. Attributes and Levels of the Discrete Choice Experiments.**
were monotonic across attributes (ie, there was a clear best-to-worst ordinal ranking for levels within each attribute).

Subjects and Data Collection
An established participant pool, maintained by the Harvard University Decision Science Laboratory (HDSL), was used to enroll volunteers from the general population. Children (less than 18 years old) and non-English speakers were excluded. The participants performed online surveys in a supervised setting over a period of 5 days. Responses were stored anonymously. Remuneration was $12 per survey. There is no consensus for the ideal sample size in DCEs; sample sizes typically range from 100 to 300 respondents.16 This study aimed for approximately 200 responses.

Data Analysis
Primary analysis was done with a conditional logistic model. This type of modeling allows for modeling based on response attributes in addition to subject characteristics.23 In this case, the attributes of choice (incision, hospital stay, etc) were analyzed as potentially important determinants of choice, in addition to the personal demographic factors of the participants. Firth bias-adjusted parameter estimates represented part-worth utilities (defined as a unit of desirability associated with each level of each attribute), and attribute importance (defined as each attribute’s part-worth utility range divided by utility range total) was calculated for each participant and averaged.24

A second conditional logistic model was used to analyze the association of attribute utility with demographic, medical, and socioeconomic factors. Income was modeled continuously, and the selected income range midpoint was used as the stated income. Consequentiality, stated comprehension, and response consistency were used as measures of validity. Finally, part-worth utilities from participant responses were used to determine the probability of choosing between several predetermined surgical options to simulate participant choices. The attribute levels used for this simulation were drawn from reviews of both endoscopic and open tracheal surgery published in the literature as well as expert opinion.1-5,18-21,25,26 Significance was considered to be $P < .05$.

Results

Patient Characteristics
Of 163 participants, 162 completed all DCE questions (99.4%). The characteristics of the participants are shown in Table 2. Mean participant age was 35.4 years; 56.2% were female, 48.8% identified as white, average income was $81,381, and 58.7% were college graduates. Approximately 20.4% reported living with a chronic illness, and 1.2% reported a previous diagnosis of subglottic stenosis.

Choice Model
Four of the 5 attributes were significant to patient decision making: voice outcome ($P < .001$), need for repeat procedures ($P < .001$), risk of complications ($P < .001$), and need for an incision ($P = .001$). The length of hospital stay was not significant ($P = .089$). The importance of each attribute is shown in Figure 2. The most important factor for participants was the need for repeat procedures (importance 30.2%). Operative risk (importance 28.1%) and postoperative voice quality (importance 27.7%) were also very important, whereas presence of incision (importance 5.0%; $P = .001$) was less important, and hospital stay (importance 9.0%; $P = .089$) was not important. The utilities for each attribute were as expected: holding other factors constant, participants were shown to prefer no incision to an incision, shorter hospital stay, good voice outcomes, fewer repeat procedures, and decreased complications. This can be seen by the monotonic appearance of the utility graph in Figure 3. The steeper slope of the utility graphs for voice outcome, repeat procedures, and risk of complication reflects the importance of these attributes. The magnitude of the utility range (ie, the difference in calculated utility between the most and least desirable alternatives) was greater for the attributes with higher calculated importance.

Subjects effects were assessed using a separate conditional logistic model. There was a significant interaction between sex and an incision preference; while both men and

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**Figure 1.** An example of a choice set seen by participants. Each column is an option.
women preferred no incision to incision, women’s preference to avoid incision was significantly stronger ($P = .009$). Nonwhite race was correlated with higher tolerance of repeat procedures ($P = .024$). Those without preexisting chronic illnesses and those without children more strongly preferred avoiding risk of complications from surgery ($P = .001$ and $P = .016$). Those with higher reported income more strongly preferred better voice outcomes ($P < .001$). The 2 patients with a history of subglottic stenosis were significantly more likely to accept repeat procedures ($P = .015$). Educational status did not affect attribute preferences. All participants indicated that they understood the format and content of the DCE.

**Scenario Simulation**

Calculated part-worth utilities were entered into a head-to-head scenario simulation designed to mimic the real-world surgical options (endoscopic or open surgery) for subglottic stenosis. The probability of a patient from this population choosing endoscopic surgery (no incision, 0-day hospital stay, normal postoperative voice, repeat procedure every 2 years, 1% risk of complication) vs open surgery (incision, 9-day hospital stay, soft postoperative voice, no repeat procedure, 10% risk of complication) was 80.4%. This was similar to the response when patients were asked with a simple question whether they would prefer an endoscopic scenario or an open scenario (88.3% and 11.7%, respectively).

Various alterations to the assumptions were then made. When the scenario for the endoscopic option was changed to reflect a repeat procedure rate of every 6 months, 70.5% of participants were projected to choose the endoscopic option. When the endoscopic scenario was changed to show a risk rate of 20%, the open option was favored by 55.1% of participants. When the open scenario was changed to reflect a perfect voice outcome (ie, “normal voice”), the endoscopic and open scenarios were nearly equal, with 53.6% preferring endoscopic and 46.4% preferring open surgery.

**Discussion**

In this pilot study of 162 participants, we aimed to identify the most important patient preferences associated with surgical treatment of subglottic stenosis. Using a DCE, we determined that the most important attributes in the treatment of subglottic stenosis for this group of respondents were the need for repeat procedures, risk of complications, and voice outcomes. The need for an incision was less important, and length of stay was determined not to be a significant predictor of choice. Our model showed a relative but not absolute preference for endoscopic over open treatment of subglottic stenosis, and this preference was replicated when participants were directly asked to choose between the 2 options.

Although preference data are scarce, satisfaction ratings with various surgical options for laryngotracheal stenosis have been reported. A large series of open tracheal resection for idiopathic tracheal stenosis reported very high satisfaction with operative intervention, with 45% of patients maintaining a normal voice.21 Another study surveying 160 patients with acquired or idiopathic tracheal stenosis reported that patient satisfaction was higher with open surgical resection (76%) than with other modalities (39%).

This finding may be misleading, however, as the definition of satisfaction was limited to improvement in breathing; other important factors, such as voice quality, procedural complications, and cosmesis, were not included. It may be that these factors matter less than breathing, but our pilot data indicate that they do matter to some degree.

Investigation into patient preferences is vital to providing value in medicine. In otolaryngology, many conditions are preference sensitive (ie, several options exist for treatment and there are trade-offs between these options), and quality-of-life concerns play a key role in decision making. Subglottic stenosis is no exception—there are clear trade-offs and quality-of-life implications depending on treatment strategy. While each patient is different, an understanding

### Table 2. Characteristics of the Participant Pool.

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Response</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, mean (SD), y</td>
<td>35.4 (16.7)</td>
</tr>
<tr>
<td>Male, % (n)</td>
<td>43.8 (71)</td>
</tr>
<tr>
<td>Race, % (n)</td>
<td></td>
</tr>
<tr>
<td>White</td>
<td>48.8 (79)</td>
</tr>
<tr>
<td>Black/African</td>
<td>17.9 (29)</td>
</tr>
<tr>
<td>Asian</td>
<td>21.0 (34)</td>
</tr>
<tr>
<td>Hispanic</td>
<td>5.6 (9)</td>
</tr>
<tr>
<td>Other</td>
<td>6.2 (10)</td>
</tr>
<tr>
<td>Income, % (n)</td>
<td></td>
</tr>
<tr>
<td>$0-$25,000</td>
<td>25.3 (41)</td>
</tr>
<tr>
<td>$25,000-$50,000</td>
<td>16.0 (26)</td>
</tr>
<tr>
<td>$50,000-$75,000</td>
<td>12.3 (20)</td>
</tr>
<tr>
<td>$75,000-$100,000</td>
<td>5.6 (9)</td>
</tr>
<tr>
<td>$100,000-$150,000</td>
<td>12.3 (20)</td>
</tr>
<tr>
<td>$150,000-$200,000</td>
<td>5.6 (9)</td>
</tr>
<tr>
<td>$200,000-$250,000</td>
<td>4.9 (8)</td>
</tr>
<tr>
<td>$250,000+</td>
<td>6.2 (10)</td>
</tr>
<tr>
<td>Prefer not to say</td>
<td>11.7 (19)</td>
</tr>
<tr>
<td>Education, highest level % (n)</td>
<td></td>
</tr>
<tr>
<td>High school</td>
<td>15.4 (25)</td>
</tr>
<tr>
<td>Attended college</td>
<td>24.7 (40)</td>
</tr>
<tr>
<td>Graduated college</td>
<td>35.2 (57)</td>
</tr>
<tr>
<td>Advanced degree</td>
<td>23.5 (38)</td>
</tr>
<tr>
<td>Prefer not to say</td>
<td>1.2 (2)</td>
</tr>
<tr>
<td>Number of children, % (n)</td>
<td></td>
</tr>
<tr>
<td>0</td>
<td>82.1 (133)</td>
</tr>
<tr>
<td>1-2</td>
<td>11.8 (19)</td>
</tr>
<tr>
<td>3+</td>
<td>6.2 (10)</td>
</tr>
<tr>
<td>Live with chronic illness of any kind, % (n)</td>
<td>20.4 (33)</td>
</tr>
<tr>
<td>Have diagnosis of subglottic stenosis, % (n)</td>
<td>1.2 (2)</td>
</tr>
</tbody>
</table>
of patient preferences and outcomes expectations allows physicians to engage more fully in shared preoperative decision making.\textsuperscript{15,28} In addition, understanding patient preferences before intervention may help lower health care costs and provide more value to patients.\textsuperscript{29} Prospectively, patient preferences may also soon inform regulatory decisions as well. Authors from the Food and Drug Administration (FDA) recently published a study assessing the importance of preference information for weight loss devices, concluding that DCEs “provide information for making patient-centered, evidence-based regulatory decisions.”\textsuperscript{30} Decision-making tools based on eliciting patient preferences are increasingly being encouraged by regulatory bodies in the context of elective procedures.\textsuperscript{28} Finally, preference information can be helpful in the development of new technologies; that is, consumer preference can change the products and services offered. While this may seem a distant goal for airway surgery, there are nevertheless new treatments being developed (serial intraluminal steroid injection protocols, stenting devices,\textsuperscript{31} endoscopic laryngotracheoplasty with skin grafts,\textsuperscript{32} etc), which can be improved by understanding patient preferences.

There are limitations to this study. Our subjects represent a sample that shows a higher proportion of women than men, has a generally high level of education, is somewhat younger than the national average age, is wealthier than the average American, and is less likely to be white than the national average. This limits generalizability to the general population. Although the ability of nonaffected individuals to establish patient preferences for specific conditions may have inherent limitations, general population sampling is common practice as a way of establishing utilities that can also be used for societal and policy decisions.\textsuperscript{33,34} In addition, stated preference research is limited by the qualitative nature of

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**Figure 2.** Relative attribute importance.

**Figure 3.** Utilities of attributes.

*Note: Positive utilities are preferred to negative utilities.*
attribute and level determination. As a result, the way in which researchers frame the clinical question can affect the outcome of the experiment.8,9 We minimized this potential source of bias by appropriate interviewing, pilot testing, literature review, and review by experts. Nevertheless, rigorous and standardized assessments of preference and decision aids must be developed in otolaryngology for specific conditions to gather data with the highest fidelity, as has been done in other fields.28

This study begins to shed light on some of the complex treatment decisions made by patients with airway and voice conditions. Elucidation of trade-offs in surgical therapy has broad implications within otolaryngology; similar research has already been used to understand preferences, decision-making techniques, and willingness to pay for medical and surgical treatment within our field.11-15,35

Conclusion

In this study, the most important factors when considering surgery for subglottic stenosis were need for repeated procedure, risk of complication, and voice outcome. Our model predicted that most participants would select an intervention that involved fewer procedures, low risk, and voice preservation.

Acknowledgments

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Author Contributions

Matthew R. Naunheim, contributed to design of project, preparation of survey, execution of analysis, interpretation of data, writing manuscript, coordinating editing process among authors; Margaret L. Naunheim, contributed to design of project, preparation of survey, interpretation of data, writing manuscript; Vinay K. Rathee, contributed to interpretation of data, writing manuscript, coordinating editing process among authors; Ramon A. Franco, contributed to design of project, interpretation of data, writing manuscript; Mark G. Shrime, contributed to design of project, preparation of survey, execution of cost analysis, interpretation of data, writing manuscript, coordinating editing process among authors; Phillip C. Song, contributed to design of project, preparation of survey, execution of analysis, interpretation of data, writing manuscript.

Disclosures

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References


