Derivation and Initial Validation of the Utility Function for the Hearing Utility Measure (HUM)

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Objective: The Hearing Utility Measure (HUM) is a replacement hearing attribute for the Health Utilities Index, Mark 3 (HUI-3) designed to improve the responsiveness of utility estimates to changes in hearing-related quality of life. The final development step is to derive the instrument's utility scoring function.

Methods: Residents of Ontario, Canada, aged ≥18 years participated in standard gamble and visual analogue scale exercises. Valuations for levels (response options) within each domain, and for each domain relative to the other domains were elicited and used to generate a hearing utility function. The function outputs hearing utility ranging from $0 = '$ unable to hear at all' to $1 =$ 'perfect hearing' for each of the 25,920 hearing states classifiable by the HUM. Performance was assessed relative to the criterion standard: directly elicited standard gamble utility. Distributions of HUM-derived hearing utility were compared with legacy HUI-3 derived estimates.

Results: A total of 126 respondents participated (mean age 39.2, range 18–85 years, 53% female [67/126]). The utility function performed well in the estimation of directly elicited utilities (mean difference 0.03, RMSE 0.06). Using the legacy HUI-3, estimated hearing utility was 1.0 for 118/126 respondents (93.6%) compared with just 66/126 (52.4%) using the HUM.

Conclusion: The new hearing attribute is capable of measuring variations in hearing utility not captured by the legacy HUI-3, especially near the ceiling of hearing function. These findings justify its application and further work to study its measurement properties in hearing loss populations.

Key Words: cochlear implants, health policy, quality of life. Level of Evidence: 3

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INTRODUCTION

Health utility is a single-number summary of healthrelated quality of life anchored at 1 (perfect health) and 0 (dead). Health utility has been used in costeffectiveness calculations for hearing loss interventions and have supported funding and policy decisions in several jurisdictions. $1-3$ However, existing health utility instruments were designed to be generically applicable to all health and disease conditions and this generalizability comes at the cost of specificity. The Health Utilities Index, Mark 3 (HUI-3) includes the most comprehensive classification of hearing status of all available utility instruments but still lacks the content necessary to discriminate many clinically significant hearing impairment and rehabilitation states. For example, there is negligible measured health utility benefit of bilateral compared with unilateral cochlear implantation despite established audiometric and health-related quality of life benefits, in part because the HUI-3 does not contain content about sound directionality, bilateral hearing, tinnitus, and other

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TRM is on the medical advisory board for Envoy Medical. DF reported being an owner of Health Utilities Incorporated which owns the copyright for the Health Utilities Index. SLC reported serving on the speaker's bureau, advisory board and holding a sponsored research agreement for Cochlear Corporation during the conduct of the study; serving on the speaker's bureau for Interacoustics and Cooke Medical, the advisory board for Akouos (Eli Lilly), being a consultant for Decibel Therapeutics (Regeneron), and receiving royalties from Plural Publishing outside the submitted work; and having a patent to 7041–0 issued unrelated to this work. JMC is on the Surgical Advisory Board of MedEl, Austria.

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constructs known to be benefited by bilateral implanta-tion.^{[1,4,5](#page-8-0)} This problem extends to other modern hearing rehabilitation strategies including bone-conduction implants for single-sided deafness. $\frac{6}{6}$ $\frac{6}{6}$ $\frac{6}{6}$ Further, hearing state descriptions by generic measures are not clearly applicable to cochlear implant (CI) users due to references to 'hearing aid' use, which may mediate poor reli-ability in those populations.^{[7](#page-8-0)} Given these limitations, there is a risk of misreporting cost-effectiveness of hearing interventions, a problem that compounds as new technologies and treatment paradigms emerge to address a more nuanced understanding of hearing health.

To address the problem poor sensitivity of generic health utility instruments, we developed the Hearing Utility Measure (HUM), the first ever condition-specific health utility instrument designed for hearing impaired populations (Fig. [1](#page-2-0), Panel A). 8 The HUM was designed to function both independently and within the framework of the HUI-3. \degree The HUI-3 has two components: (1) a health status classification system that systematically categorizes a respondents health status according to eight attributes, one of which is Hearing, and (2) a multi-attribute utility function that translates levels of ability or disability within those attributes to health utility estimates. Based on an evidence-based conceptualization of hearing-related quality of life,^{[10](#page-8-0)} the HUM was developed as a replacement for the legacy HUI-3 Hearing attribute. The HUM has an expanded and more comprehensive conceptualization of hearing-related quality of life with seven hearing domains (also called 'sub-attributes' in the context of a utility instrument): Speech understanding, Environmental sounds, Sound localization, Listening effort, Tinnitus, Music, and Reliance on assistive hearing devices[.8](#page-8-0) The HUM discriminates 25,920 hearing states compared with just six hearing states in the legacy HUI-3 Hearing attribute.

The final development step in development is to generate the utility function that translates levels of ability and disability within the sub-attributes to a hearing utility estimate. The output hearing utility has two applications: (1) as a stand-alone measure of society's preference for a hearing state on an 'unable to hear at all' $= 0$ to 'perfect hearing' $= 1$ scale; and (2) to generate a hearing coefficient for the HUI-3 multi-attribute utility function, which in turn can be used to estimate overall health utility on a 'dead' = 0 to 'perfect health' = 1 scale. We hypothesized that estimating health utility using a more comprehensive classification of hearing status will result in improved discrimination of clinically relevant hearing states compared with the legacy instrument.

METHODS

Modeling Theory

A set of hearing states was selected for valuation that is sufficient to generate a function to estimate hearing utility for all 25,920 hearing states. The approach was adapted from that of the HUI-3 multi-attribute utility function.^{[11](#page-8-0)}

Two sets of information are needed to generate the hearing utility function. First, relative preference for each level within a sub-attribute must be estimated. For example, within the

Listening effort sub-attribute: How much worse is "The effort needed to listen somewhat affects my energy level" than "Very little effort is needed to listen?" To answer this and related questions, respondents valued levels of ability and disability within each sub-attribute considered independently. Second, the relative influence (weight) of each sub-attribute is needed to combine the sub-attributes to estimate hearing utility. For example, how much does an individual's speech understanding impact their hearing-related quality of life relative to their ability to localize sounds? These data were obtained by asking respondents to value hearing states in which one sub-attribute was presented at its lowest possible level and all other sub-attributes were at their highest possible level ('corner states').

Setting and Participants

Valuation interviews were conducted with members of the general public at a tertiary care facility in Toronto, Canada, between December 10, 2020 and March 16, 2022. Subjects were recruited through various methods, including online advertisements on local classified websites, printed notices posted on notice boards within an academic health center, and contacting family members of patients attending outpatient clinics for nonhearing-related complaints via telephone and e-mail. Eligible participants were aged 18 years or older and residents of Ontario, Canada. Participants were excluded if they did not have sufficient command of English language or comprehension of the standard gamble exercise (see Data quality assessment below).

Structure of the Modeling Interviews

One of three interviewers trained in econometric valuation exercises met with the study participants in the tertiary care facility. All interviews were audio-recorded and lasted between 60 and 90 minutes. A 10% random sample of the interview recordings were reviewed for quality control to ensure that the interviews followed the prescribed script.

Participants first valued a set of hearing states on a vertical visual analogue scale (VAS) anchored at 0 ('Least Desirable') and 100 ('Most Desirable'). Cards were presented with written descriptions of hearing states according to the seven subattributes of hearing. Participants first placed the card describing the best possible state ('perfect hearing') at the top of the VAS. Participants were asked to select their least preferred of two options: (1) the hearing state described simply as 'unable to hear at all', or (2) the most disabled hearing state, defined by the lowest levels for each of the seven sub-attributes (Unable to hear what is said in conversation with one other person in a quiet room; Unable to hear a doorbell in a quiet room; Unable to know where the sound of a car horn is coming from without seeing the car, Unable to hear sounds at all on one side; The effort needed to listen is exhausting; Frequent or constant loud tinnitus; Unable to enjoy music because of hearing disability; Routine use of assistive hearing device). Participants placed their least preferred hearing state card at the bottom of the VAS and rated the alternative on the scale between 0 and 100. Next, participants rated cards for three marker hearing states (A, B, C) on the feeling thermometer (Supplement eFigure 1). These states remained in place as scale anchors for the remainder of the exercise.

Participants were assigned to value two of the seven subattributes according to a randomized block design. Three participant blocks for a total of 126 participants (42 per block) were planned in accordance with response variability and precision estimates that guided sample size in the generation of the HUI-3 utility function, and in accordance with observed valuation estimate variability for condition-specific utility function derivations

Fig. 1. Structure of the new multi-subattribute hearing attribute and its function within the HUI3 framework. The new hearing status classification system replaces the existing hearing attribute from the HUI3 and comprises seven subattributes of hearing (A). Levels within the hearing status classification system have corresponding coefficients for the multi-subattribute hearing utility function (bottom of B, Table [II](#page-6-0)). Hearing utility is converted to the hearing coefficient for the HUI3 multi-attribute utility function (b_{hearing}) by applying the HUI3 Hearing scaling constant. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

using similar methods.^{[11,12](#page-8-0)} Corner states (the assigned subattribute at its lowest level and all other sub-attributes at their highest levels) were valued on the VAS. The assigned subattribute was varied along its range of levels, and the participant placed each associated card on the VAS according to their preferences. This process was repeated for the second assigned subattribute.

Participants were then engaged in standard gamble exercises to estimate hearing utility for the three marker states (A, B, C) according to established protocols for developing multiattribute utility functions. $11,12$ In the standard gamble, participants were presented with two options: they could either live in the state presented or opt for a chance of 'perfect hearing' at the expense of a risk of being 'unable to hear at all.' The probability of 'perfect hearing' vs. 'unable to hear at all' was varied until the participant reached decisional equipoise. A dynamic pie chart visual aid illustrating probability called a "chance board" was used throughout this exercise. Standard gamble utilities were then elicited for the 'unable to hear at all' and 'most disabled hearing' states.

At the conclusion of the exercise, participants completed a survey of demographic information, their experience with and understanding of the exercise, and the HUI-3 and HUM.

Data Quality Assessment

Responses were screened for logic demonstrating the respondent understood the exercise. Data were excluded from the primary analysis if responses provided were not congruent with the participant's own stated preferences. For example, after a participant indicated their least preferred hearing state, valuation of any of the other states lower than that of the least preferred state would result in exclusion.

Converting Visual Analog Scale Values to Standard Gamble Utilities

A power function was specified to estimate utility from VAS values using the three marker states (A, B, C) that were valued with both standard gamble and VAS (for details, see eMethods 'Value to Utility Conversion' and eFigure 2 and eFigure 3 in the Supplement). Standard gamble utilities for states valued only with the VAS were estimated using that function.

Deriving the Multi-Subattribute Utility Function

Intervals between levels of each independent sub-attribute were available directly from the data. VAS valuations for states in which only one sub-attribute varied along its range of levels were converted to estimated standard gamble utilities (Supplement, eMethods, 'Single-sub-attribute value functions', and eFigure 4). Scaling constants for each sub-attribute were obtained from estimates for corner state utilities. The product of the set of coefficients for each of the seven subattributes was then transformed by overall scaling constants derived by solving a series of equations (Supplement, eMethods, 'Multi-subattribute utility function').

Generating a Hearing Attribute Coefficient for Use with the HUI3 Multi-Attribute Utility Function

For the hearing utility generated by the multi-subattribute utility function described in this work to be used in place of the hearing coefficient for the existing HUI-3 Hearing attribute, it must be scaled by the HUI-3 scaling constant for hearing. This conversion step is detailed in the Supplement (eMethods).

Comparing the HUM with the Legacy Hearing **Attribute**

The distributions of estimated hearing and health utilities for individuals participating in the valuation interviews was compared according to whether the legacy Hearing attribute or the HUM was used to estimate hearing utility. Distributions were compared qualitatively and with paired Wilcoxon rank order tests.

RESULTS

Participants

Valuation interviews were conducted with 126 participants (Table [I\)](#page-4-0). The sample ranged in age from 18 to 85 years (mean 39.2, SD 18 years), and 53.2% (67/126) were female. The majority were single (71/126, 56.3%) or married/common law (39/126, 31.0%). Self-reported overall health ranged from fair (4/126, 3.2%) to excellent 14 (11.1%) with 'very good' being the most common response (63/126, 50.0%). Self-reported hearing status was most commonly 'normal hearing' in both ears (93/126, 74%), with 26/126 (21%) reporting some hearing disability but no assistive device use, 5/126 (4.0%) hearing aid users, and 2/126 (1.6%) CI users. Incongruent stated preferences were collected from 2/126 (1.6%) respondents, and 3/126 (2.4%) refused to gamble, always selecting the offered guaranteed hearing state regardless of the presented probability of perfect hearing. These five participants were excluded from the analysis. An additional 7/126 (5.6%) had missing VAS or standard gamble estimates for scale anchor states or marker states and were excluded from generation of the value to utility function but included in the multi-subattribute utility function calculations. Excluded participants were more likely to be male (4/5, 80%) than included participants (48/121, 42%, $p = 0.001$) and did not significantly differ on other measured characteristics.

Value to Utility Conversion

The best-performing value to utility function was derived from aggregate sample mean value and utility estimates without outlier trimming $(U = V^{\theta}$ where $\theta = 0.3272$ $\theta = 0.3272$ $\theta = 0.3272$, Fig. 2). The sum squared error for aggregate value and utility means was 0.00135, and model fit to individual values and utilities was excellent $(R^2 = 0.8)$.

Multi-Subattribute Utility Function for Hearing

Utilities for each subattribute considered in isolation on a best (level 1) to most impaired (level 3, 4, 5, or 6 depending on the sub-attribute) scale are plotted in Figure [3.](#page-5-0) The scaling constants, c_i , for each subattribute represent the influence each subattribute had on respondent preferences. The most influential subattribute was Tinnitus $(c_i = 0.37)$ followed by Speech Recognition $(c_i = 0.31)$ and then Music $(c_i = 0.26)$. The least influential subattribute was Assistive Devices $(c_i = 0.12)$. These scaling constants were applied to convert single subattribute utilities into coefficients for the multisubattribute hearing utility function (Table [II\)](#page-6-0). The multi-subattribute hearing utility function (Fig. [1](#page-2-0), Panel B) was able to predict elicited utilities with a mean difference margin of error of \pm 0.03 (Table [III\)](#page-6-0).

Estimated Utilities in the Respondent Sample

Distributions of estimated hearing and health utilities were compared according to whether they were calculated using the legacy HUI3 Hearing attribute or the HUM (Fig. [4\)](#page-7-0) in the respondent sample as an initial validation step. Single-attribute hearing utility was estimated to be 1.0 for 118/126 respondents (93.6%) when the legacy HUI-3 Hearing attribute was used compared with 66/126 (52.4%) with the new Hearing attribute, indicating that the HUM had greater capacity to differentiate hearing states near the ceiling of hearing utility in this nonhearing-impaired sample. Median overall health utility was higher (0.905, IQR 0.196) when the legacy HUI3 Hearing attribute was used compared when the HUM was used $(0.879, \text{ IQR } 0.185; p < 0.001, \text{ paired Wilcoxon})$ rank sum).

DISCUSSION

In this valuation study, standardized econometric exercises were used to quantify preferences for different hearing states in a sample of residents of Ontario, Canada. These data were used to generate a utility function for the HUM, a novel hearing attribute designed for use with the HUI-3. The function performed well in estimating empirically elicited utilities for a set of hearing states in an internal validation assessment. Estimated personal health utilities derived from this general population sample with generally good self-reported hearing were slightly lower when the HUM was used as compared with the legacy HUI-3, suggesting a capacity for the HUM to detect hearing-related disabilities that were not detectable with the HUI-3.

The purpose of the HUM is to address the problem of poor specificity of available generic health utility instruments for discriminating clinically important hearing-related health states. Our approach is novel and addresses many of the limitations of alternatives that have been described. 'Mapping' or 'Cross-walking' item responses from a condition-specific non-preference-based PROM onto an available generic health utility instrument

* Reported in Canadian dollars.

has been suggested but does not address the problem of content limitation.^{13,14} The performance of such a function is tied to the correlation between instruments and is therefore still limited by the discriminative ability of the generic health utility instrument. Adding an additional new attribute to an existing generic utility instrument, a 'bolt-on' approach, requires that the entire instrument's scoring function be revised to contextualize the added

content.^{15,16} This introduces the risk of labeling and focusing effects, whereby the respondent may over- or under-value the new and emphasized content.[16](#page-8-0) As a result, it is difficult to compare resulting health utility estimates to those generated by other generic health utility instruments. A similar problem is introduced when an entirely new condition-specific health utility instrument is generated. These risks of bias challenge cross-program

Fig. 2. Value to utility random effects model. The model with utility parameters (black) had lower sum of squared errors (SSE) than the model with disutility (1-utility) parameters (red). [Color figure can be viewed in the online issue, which is available at [www.](http://www.laryngoscope.com) [laryngoscope.com](http://www.laryngoscope.com).]

comparability and have led to expressed preferences by many health technology appraisal agencies for health utility estimates from generic instruments.[17](#page-8-0) However, in the approach described in this work, the overall health utility function and relative importance of each established attribute is retained. Hearing is no more or less influential to overall health utility than it was in the legacy instrument. Health utility estimates can therefore be readily compared with those generated by the generic instrument in any other population sample, a critically important feature for health technology assessments in which the goal is the appropriate distribution of limited resources across health programs addressing disparate disease conditions.

What has changed is the ability to discriminate hearing states within the prescribed range that hearing can influence health utility. 8 Conditions or interventions that affect changes in any of the seven HUM domains can be captured by the new instrument. With this added discriminant ability, the HUM can measure clinically important differences in overall health utility that could not be detected by the HUI-3. In the HUI-3, hearing can influence overall health utility over a range of 0.53 (health

Fig. 3. Single-attribute utility functions on a 'no hearing' = o to 'perfect hearing' = 1 scale. Plotted numbers represent the levels within each subattribute. Scaling constants (c_i) are presented in the boxes at the top of each subattribute. These scaling factors represent the relative
weight respondents placed on each subattribute in making standard gamble decis weight respondents placed on each subattribute in making standard gamble decisions. [Color figure can be viewed in the online issue, which is available at [www.laryngoscope.com.](http://www.laryngoscope.com)]

TABLE II.

Listening Effort (b_{h4})

Localization (b_{h3})

 $MD =$ mean of the differences between predicted and elicited utilities. MAD = mean of the absolute RMSE = square root of the mean of the squared differences between predicted and elicited utilities.

utility is 0.47 with hearing at level 6 and all other attributes at level 1). This utility range contains more than 17 gradations of this instrument's minimally clinically important difference (0.03) .^{[18](#page-8-0)} With only six levels, there is therefore information lost in the ordinal categorization by the legacy Hearing attribute. The HUM's 25,920 hearing states will not all be significantly different from one another with respect to overall health utility, but all significant incremental changes in overall health utility can be measured. Another benefit is the HUM's generalized description of assistive devices, expanding applicability beyond 'hearing aids' to a number of current and future available devices.[7](#page-8-0) Perhaps the most significant contribution of this work is the novel method of estimating condition-specific health utility, whereby an optional comprehensive classification system is nested within a generic health utility instrument; this approach can serve as a framework for future expansion of other attributes of the HUI-3 or other utility instruments to generate an adaptive set of health utility instruments that improve discrimination in specific populations.

Speech Recognition (b_{h1})

Level 1

Level \mathfrak{p}

Level 3

Level 4

Level 5

Level 6

Environmental Sounds (b_{h2})

There exists considerable debate in utility measurement regarding whose preferences should be considered when establishing evidence of the cost-effectiveness of health care interventions and determining health resource allocation. The HUI suite of instruments is designed to (1) characterize the health or disease state of the respondent in objective terms, and then (2) estimate how 'society' values that health state. The arguments in favor of reliance on societal preferences are (1) patient experience-based estimates differ from those of the general public, a phenomenon that may be related to gradual

acclimatization paying for the ance premium with respect to money. Contra society have no they cannot, the preference for 'experience-ba ter ecological validity. Valid arguments on both sides of this debate exist. In the current study, we designed an instrument designed for use with the existing HUI suite and therefore followed similar valuation methods used in the design of the original instrument. Valuations with both societal and patient preferences have been performed in many different jurisdictions for other popular utility instruments including the $Eq5D²²$ $Eq5D²²$ $Eq5D²²$ Future study regarding how societal preferences are different from those with lived experiences of hearing loss would provide valuable insight regarding the estimation of health utility in patients with hearing-related disability.

Our work and approach have limitations. The improved hearing state discrimination comes at the cost of response burden associated with the addition of nine questions to the 15 question HUI-3 questionnaire. It also requires additional steps and calculations to estimate Hearing utility using a multi-subattribute function rather than the comparatively simple single-attribute utility function associated with the legacy HUI-3. These steps may be a source of user error, and a clear set of instructions and procedures need to be provided and followed for the instrument to generate accurate estimates. y.com/terms-and-conditions) on Wiley Online Library for rules of use; OA articles are governed by the applicable Creative Commons Licens

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Fig. 4. Distributions of health utility and single-attribute hearing utilities. Single-attribute hearing utility (top row) and overall health utility (bottom row) were estimated in the respondent sample with both the legacy HUI3 hearing attribute (left column) and the new hearing attribute (right column). [Color figure can be viewed in the online issue, which is available at [www.laryngoscope.com.](http://www.laryngoscope.com)]

Preference estimates were derived from a sample of respondents from Ontario, Canada. Participants were volunteers with access to the internet or with a connection to an academic health center as a patient or a family member of a patient. Utilizing multiple recruitment methods may have mitigated the risk of selection bias, but bias favoring those with higher technological and health literacy may exist in this sample. This sample was chosen to approximate the sample of Ontario residents used in the generation of the HUI-3, which had a similar distribution of age (mean 42.7, SD 17.8 years) and selfreported overall health, which are variables that may influence health state valuations.^{[23](#page-8-0)} Although this sample may justify application of the utility function in developed, English-speaking jurisdictions, whether the

valuations obtained in the sample are more broadly applicable globally requires further study.

CONCLUSIONS

The multi-subattribute utility function for the HUM performs well in the estimation of elicited hearing utilities. The HUM can be used with the HUI-3 as a replacement for its 6-level Hearing attribute to estimate overall health utility and is capable of measuring variations in hearing utility not captured by the legacy instrument. These findings justify application of the HUM in the estimation of hearing and health utility, and ongoing work to study its measurement properties in populations with hearing-related disability.

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