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"Patients' with obstructive sleep apnoea syndrome (OSAS) preferences and demand for treatment: a discrete choice experiment"

Nicolas Krucien, Amiram Gafni, Bernard Fleury and Nathalie Pelletier-Fleury

Supplementary file

The development of the discrete choice questionnaire was carefully performed following a step-by-step approach.

1st step: Selection of the attributes and their levels

The attributes and levels of each attribute used to describe the options were selected on the basis of a literature review. At this step the objective was to select the most relevant attributes used by patients and clinicians in the medical decision making. There were 4 attributes with two levels: rate of effectiveness (40%, 100%), severity of side effects (severe, not severe), time before improvement in health condition (4 weeks, immediately), negative impact on daily life (high, low). The out-of-pocket expense attribute had four levels (0€, 100€, 200€, and 300€).

2nd step: Design of the experience

With 4 attributes with 2 levels each and 1 attribute with 4 levels, 64 combinations of outcomes could be defined. For a paired generic comparison format (i.e. treatment "A" versus treatment "B"), a full factorial design of choice tasks led to 2 016 possibilities. To reduce the number of possibilities we used an orthogonal main effects plan with fold-over procedure allowing a final solution of 16 tasks. Because choice tasks are cognitively demanding for respondents, the 16 tasks were randomly allocated into 2 blocks of 8 tasks each. In addition 2 other tasks were introduced into each block to verify the hypotheses of monotonicity (i.e. more is preferred to less) and stability underlying the choice behavior of respondents. Respondents were deemed "inconsistent" when they failed at least one of the two tests. If this was the case, they were excluded from the statistical modeling.

3rd step: Sampling and administration of survey

Anticipating a 90% rate of success in the "consistency" tests and following Aspinall et al (in: Invest Ophthalmol Vis Sci, 2008;49(5):1907-15) method to calculate the sample size, a minimum of 140 patients had to be recruited. All participants were informed that their responses were anonymous and will not influence their forthcoming care. Written information was provided that describes clearly, systematically and in an easy way to understand the choice tasks.

4th step: Discrete choice modeling

To model the preferences we used a nested logit model, by separating the options in two different nests, namely a "no treatment" nest composed of the "no treatment" option, and a "treatment" nest composed of options "A" and "B". The model was estimated by logistic regression with the choice made as dependent variable. The respondent (n=1,...,N) at the task (t=1,...,T) will choose the option (A) over the option (B) if the utility of (A) is greater than that of (B).

$$Y_{nt(A)} = \begin{cases} 1, \text{ if } U_{nt(A)} > U_{nt(B)} \\ 0, \text{ if } U_{nt(A)} \leq U_{nt(B)} \end{cases}$$

Where the utility of (A) consist in a systematic observable component (V) and a random unobservable component (ε):

$$U_{nt(A)} = V_{nt(A)} + \varepsilon_{nt(A)}$$

Attributes' levels are used to explain the systematic part of the utility.

$$U_{nj} = \beta_1 NO + \beta_2 A + \beta_3 EFF + \beta_4 SIDE + \beta_5 TIME + \beta_6 IMP + \beta_7 EXP + \varepsilon_{nj}$$

Where "NO" is a constant to estimate overall tendency of patients to choose the "no treatment" nest over the "treatment" nest. "A" is a constant to estimate a potential right/left bias, namely an overall tendency of patients to choose the option A rather than B disregarding its content (i.e. attributes' levels). EFF is the "rate of effectiveness" attribute and its associated parameter β_3 is the marginal utility of moving from a treatment with 40% effectiveness to a treatment with 100% effectiveness. SIDE, TIME, and IMP are respectively the "side effects", "time before improvement", and "negative impact on daily life" attributes. Their associated parameters β_4, β_5 and β_6 are the marginal utility of moving from the worst attribute's level to the best. EXP is the "out-of-pocket expense" attribute. Its associated parameter β_7 is the marginal utility for a one euro change in out-of-pocket expense. The (ε_{nj}) term is a random component identically independently distributed as type 1 extreme value. Under this specification, the choice probabilities can be predicted with a logit model.

$$?(?_{?} = ?) = \frac{?\left(?_{?(?)}\right)}{?\left(?_{?(?)}\right) + ?\left(?_{?(?)}\right)}$$

Following Lancsar et al (in: *Soc Sci Med*, 2007;64:1738-53) we assessed the relative impact of each attribute on the choice of treatment by analyzing changes of the log-likelihood of the model. The variables were "effect" coded (-1; +1) to allow a meaningful estimate of the "model constant". The out-of-pocket expense variable was entered into the model as a continuous variable.

In addition, from the estimated preferences it was possible to predict the probability of choosing a specific treatment with given levels of attributes. Given that a CPAP treatment was considered 100% effective, with non-severe side effects, no time to wait before treatment to be effective, a high negative impact on daily life, and in the French case a 378€ out-of-pocket expense per year, the utility of CPAP treatment was computing as below:

$$U(CPAP) = (2.13 \times 1) + (1.27 \times 1) + (0.82 \times 1) + (3.17 \times 0) + (-0.0038 \times 378) \cong 2.78$$

In the same way, the utility of OA treatment is 2.28. The "No treatment" option is the only one for which its subjective value cannot be recovered, and then we need to fix its value. Conventionally its value is assumed to be null. These utility values were used to predict the probabilities of choice.

$$P(CPAP) = \frac{exp(CPAP)}{exp(CPAP) + exp(OA) + exp(NO)} = 60.2\%$$

$$P(OA) = \frac{exp(OA)}{exp(CPAP) + exp(OA) + exp(NO)} = 36.2\%$$

$$P(NO) = \frac{exp(NO)}{exp(CPAP) + exp(OA) + exp(NO)} = 3.6\%$$

To account for possibility of different out-of-pocket expenses according healthcare systems, a sensitivity analysis was performed using different amounts of out-of-pocket expense (from 0 to 1 000 Euros) for both CPAP and OAs. It appeared that patient demand for CPAP was very sensitive to the variation in the amount of out-of-pocket expense for both CPAP and OAs (see figure). The higher the out-of-pocket expense for CPAP was, the less the likelihood of choice of CPAP was (horizontal reading of the graph). The cheaper the OAs treatment was, the less the likelihood of choice of CPAP was (vertical reading of the graph).

Figure. Probability of CPAP choice according to out-of-pocket expense for CPAP and OAs (in Euros per year)