RESEARCH LETTER

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

Rationale Despite its high level of effectiveness, initial acceptance of continuous positive airway pressure (CPAP) and regular use in patients with obstructive sleep apnoea syndrome (OSAS) are still an issue. Alternatively, oral appliances (OAs) can be recommended. To improve patient engagement in their treatment, physicians are advised to take into account patient preferences and to share the therapeutic decision. We aimed to determine patients' preferences for OSAS treatment-related attributes, and to predict patients' demand for both CPAP and OAs.

Methods A discrete choice experiment (DCE) was performed in 121 newly diagnosed patients consecutively recruited in a sleep unit.

Results Regression parameters were the highest for impact on daily life and effectiveness ahead of side effects. In the French context, the demanding probabilities for CPAP and OAs were 60.2% and 36.2%, respectively. They were sensitive to the variation in the amount of out-of-pocket expenses for both CPAP and OAs.

Conclusions This first DCE in OSAS emphasises the importance to communicate with patients before the implementation of treatment.

INTRODUCTION

Following the most recent guidelines, continuous positive airway pressure (CPAP) is indicated as a first-line treatment for patients suffering from obstructive sleep apnoea syndrome (OSAS).[1] However, initial acceptance and regular use of CPAP treatment are still an issue.[2] Alternatively, oral appliances (OAs) are recommended in case of initial refusal or failure of CPAP option, and also as a first-line treatment in mild to moderate OSAS.[1] Because of problems of compliance, patients and physicians are faced with difficult decisions regarding which OSAS treatment options to choose. Physicians are encouraged to take into account patients' preferences, and possibly to involve them in the medical decision making.[3] We used a preferences elicitation method, namely the discrete choice experiment (DCE),[4] to determine patients' preferences for OSAS treatment-related attributes, and to predict patients' demand for both CPAP and OAs.

Attribute	No treatment	Treatment "A"	Treatment "B"
Rate of effectiveness (i.e. ability of treatment to eliminate snoring and apneas if used as instructed)	*No improvement of your health state at no cost*	40%	100%
Severity of side effects (a severe side effect was depicted as a side effect that could impose cessation of treatment.		Not severe	Severe
Time before improvement in health condition		4 weeks	Immediately
Negative impact on daily life (i.e. annoying and cumbersome nature of the equipment used in each treatment)		Low	High
Out-of-pocket expense (i.e. expense per year after reimbursement by social and private insurance)		€ 100	€ 300
Which option would you choose?	☐	☐	☐

Figure 1 Illustration of a choice task.

METHODS

Five attributes were used to describe treatment options.[5] The choice tasks were based on a paired comparison format and included an opt-out option (ie, no treatment) (figure 1). We used an experimental design with 16 choice tasks randomly allocated into two versions of eight tasks each. The two versions were randomly administered by a nurse to 121 patients newly diagnosed with OSAS and recruited consecutively in a French hospital sleep unit (67.8% were males, 53.5±12 years old (mean±SD), with a body mass index of 29.3±5.65 and an apnoea–hypopnoea index of 41.5±22.4). Patients' choices led to 2904 observations from which preferences were estimated by logistic regression.

To predict patients' demand for both CPAP and OAs, we assumed CPAP (OA) treatment to be 100% (40%) effective, with non-severe (severe) side effects, no time (4 weeks) to wait before improvement, with a high (low) negative impact on daily life and €378 (€233) out-of-pocket expense per year (in the French context).

RESULTS

All the estimates of the model were significant and of the expected sign. Patients preferred a high rate of effectiveness, non-severe side effects, a short time to wait before treatment to be effective, a low negative impact on daily life and a less expensive treatment. 'Negative impact on daily life' was the most influential attribute on the patients' choices. Its relative impact was twice larger than that of the second most influential attribute, which was the 'effectiveness' attribute (table 1).

Table 1 Nested logit model estimates and impact analysis (n=2904 observations)

Effect	Estimate (SE)	Partial effect*	Relative effect (%)†
Treatment (A)	0.024 (0.186)	–	–
(No) treatment	−0.964 (0.483)‡	–	–
Rate of effectiveness (*ref: 40%*)	1.065 (0.280)‡	−62.7	25.9
Severity of side effects (*ref: severe*)	0.635 (0.202)‡	−21.6	8.9
Time before improvement (*ref: 4 weeks*)	0.412 (0.133)‡	−8.9	3.7
Negative impact on daily life *(ref: high)*	1.586 (0.428)‡	−141.7	58.6
Out-of-pocket expense (*continuous variable*)	−0.004 (0.001)‡	−6.9	2.9

Log likelihood (LL) of 'full' model=−662.3; LL of 'null' model=−420.5.
*Partial effect=LL of the model including only the attribute; LL of the 'null' model.
†Relative effect=100×(partial effect/(LL of 'full' model; LL of 'null' model)).
‡Estimated parameter significantly different from zero for a 5% α-risk.

In the French context, the demanding probabilities for CPAP and OAs were 60.2% and 36.2%, respectively. They were sensitive to the variation in the amount of out-of-pocket expense for both CPAP and OAs.

CONCLUSIONS

To our knowledge, this is the first study that used the DCE method to measure patients' preferences for OSAS treatments. Because it was a single-centre study which took place in one healthcare system in which public insurance covers 65% of treatment cost (ie, in France), we should be cautious with the generalisability of the results. This DCE in OSAS emphasises the importance of communicating with patients before the implementation of treatment, since effectiveness of treatment and impact on daily life constitutes the most important factors of choice ahead of side effects. However, these preferences could be threatened by the high level of out-of-pocket expenses. Further research is needed to investigate more specifically how financial constraint can influence patients' preferences.

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Competing interests BF is consultant for a French company developing and selling oral appliance devices (Orthosom).

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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.

$$\text{[illegible]}\left(\text{[illegible]}_{\text{[illegible]}} = \text{[illegible]}\right) = \frac{\text{[illegible]}\left(\text{[illegible]}_{\text{[illegible]}(\text{[illegible]})}\right)}{\text{[illegible]}\left(\text{[illegible]}_{\text{[illegible]}(\text{[illegible]})}\right) + \text{[illegible]}\left(\text{[illegible]}_{\text{[illegible]}(\text{[illegible]})}\right)}$$

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)