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Eliciting Preferences in Dentistry using Multi-Attribute Stated Preference Methods: A Systematic Review

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Understanding patient, professional and public preferences is fundamental for evidence-based decision-making and treatment delivery. Preference elicitation methods can be used to estimate the value given to health states, service delivery, individual treatments and health outcomes. By describing and appraising the methodology and application of multi-attribute stated preference experiments in dentistry, this review provides an essential first step to wider use of well-designed, high quality preference elicitation methods.

ABSTRACT

Background: Preference experiments are used to understand how patients and stakeholders value aspects of healthcare. These methods are gaining popularity in dentistry but quality and breadth of use has not been evaluated.

Objectives: To describe multi-attribute stated preference experiment use in dentistry through illustration and critique of existing studies.

Data sources: Systematic literature search of PubMed, Econlit and Ovid for Medline, Embase, PsychINFO, PsychARTICLES and All EBM Reviews, and grey literature.

Study eligibility: Multi-attribute stated preference experiments eliciting preferences for dental service delivery, treatments and oral health states from patients, public and dental professional perspective. Outcomes of interest were preference weights and marginal rates of substitution. Study selection was performed by two reviewers independently.

Appraisal: 10-point checklist published by International Society of Pharmacoeconomics and Outcomes Research (2011) was used for quality assessment.

Synthesis: Descriptive analysis.

Results: Searches identified 12 records published between 1999-2015, mostly in non-dental academic journals. Studies were undertaken in high-income countries in Europe and the USA. The studies aimed to elicit preference for service delivery, treatment or oral health states from the perspective of the patients, dentists or the public using Discrete Choice Experiment (DCE) methods. The quality scores for the studies ranged from 53-100%.

Limitations: A detailed description and critique of stated preference methods are provided but it was not possible to provide synthesised preference data.

Conclusions: Multi-attribute stated preference experiments are increasingly popular but understanding the methods and outputs is essential for designing and interpreting preference studies to improve patient care. Patient preferences highlight important considerations for decision-making during treatment planning. Valuation of health states and estimation of willingness-to-pay is important for resource planning and allocation and economic evaluation. Preference estimates and relative value of attributes for interventions and service delivery inform development and selection of treatments and services.

Review proposal registration: PROSPERO 21.3.17 (CRD42017059859).

INTRODUCTION

Preference is described as an individual's liking for one alternative over another and it depends on values built through knowledge, experience and reflection. Patient preferences are a fundamental component of evidence-based healthcare and there is evidence that involving patients in care decisions and delivery results in better choices and more effective, personalised care (Mulley et al. 2012). Preference elicitation methods aim to provide important evidence for decisions about treatment selection and delivery, and service provision, including allocation of resources, clinical and policy decision-making, prediction of demand and planning services and economic evaluations (Centre for Devices and Radiological Health 2016).

Two broad categories of preference elicitation methods exist. Revealed preference methods estimate preferences using data arising from observation of behaviour around actual choices. Stated preference experiments obtain preferences using hypothetical scenarios, enabling existing choice situations to be tested under controlled conditions or exploration of non-existent choices. Multi-attribute stated preference experiments are a subset of stated preference experiments and are based on Lancaster's theory of value (Lancaster 1966). This states that utility (value) is derived from the underlying characteristics (attributes) of a good or service and consumer choice is a result of consideration of more than one attribute. It is assumed that people are rational utility maximisers, which means that when making choices between alternatives, the positive and negative attributes of a good or service are weighed up and traded-off to give a choice that is expected to yield the greatest utility.

Multi-attribute stated preference experiments require characterisation of the intervention, service or health state and identification of the underlying attributes. The most relevant attributes are selected and the dimensions of the attribute (attribute levels) are defined as numerical or categorical variables (Figure 1). Attribute levels are presented in different combinations as hypothetical scenarios. Respondents are asked to choose which scenario they prefer, forcing trading off between attributes. Attribute values are estimated based on analysis of the trade-offs that respondents are willing to make, assuming that less valued attributes are sacrificed for those perceived to have more utility. The requirement for respondents to trade-off multiple attributes within each scenario differentiates these

methodological approaches from simple rating scales where choices are expressed independently without discrimination, and from less complex single-attribute ranking methods. Multi-attribute stated preference experiments provide an estimated value for the utility of the attributes and levels (attribute coefficients) and from this it is possible to calculate part-worth utilities (the additional utility gained from attribute relative to attribute base level) and marginal rates of substitution (attribute values relative to each other) such as money equivalence (willingness to pay), time equivalence (willingness to wait) and risk equivalence (maximum acceptable risk).

Multi-attribute stated preference experiments can be divided into two groups; those quantifying the value of attributes to explore trade-offs in multi-attribute goods or service and those eliciting direct monetary values for an intervention to estimate demand for a single product (Bridges et al. 2011). This review focuses only on the former, also known as conjoint analysis. Multi-attribute stated preference experiments are designed in discrete, sequential stages (Figure 2) These are described in detail in best practice guidance published by the International Society of Pharmacoeconomics and Outcomes Research (ISPOR) (Bridges et al. 2011; Johnson et al. 2013; Hauber et al. 2016) and in methodology papers published by experts in the field (Ryan and Gerrard 2003; Lancsar and Louviere 2008; Vinney et al. 2002; Mangham et al. 2009).

In healthcare multi-attribute stated preference experiments have been used with three key groups of stakeholders 1) Patients - for valuing experience, health outcomes, and trade-offs between experience and outcome 2) Health professionals - preferences for treatment and screening 3) Public - preferences to inform priority setting and development of policy (Clark et al. 2014). With the growing awareness of the potential for preference elicitation methods in dentistry, a review of the quality and breadth of use in this area is timely and pertinent to guide good practice from the outset. This study aims to describe and evaluate how multi-attribute stated preference experiments have been used in dentistry to elicit preferences for dental procedures, treatment outcomes, oral health states and service delivery from a patient, public, professional or stakeholder perspective.

MATERIALS AND METHODS

A scoping review confirmed no reviews were planned or published on this topic. The protocol was published on PROSPERO in March 2017 (CRD42017059859).

Eight electronic databases were searched (Ovid: Medline, Embase, PsychINFO, PsychARTICLES, All EBM Reviews; Pubmed; Econlit). A grey literature search involved hand searching reference lists of relevant publications and reviews and targeted searching of online resources: NICE and SIGN; www.clinicaltrials.gov; Google Scholar; Conference abstracts. Search terms were constructed around two search concepts, stated preference experiments and dentistry (Appendix Table 1) based on a previous review (Clark et al. 2014). No language or date restrictions were applied.

Study eligibility criteria was applied:

Population: any person participating in a multi-attribute stated preference experiment related to dentistry - people receiving dental treatment (patients), public, dental professionals and other key stakeholders e.g. health service providers, service managers. No restrictions for age, sex, ethnicity or country of origin or setting.

Intervention: Studies eliciting preferences for dental service delivery, treatment(s) for dental/oral conditions involving dental professionals and oral health states. Exclusion: Studies eliciting preferences for multiple healthcare services with dentistry as only one subgroup.

Outcomes: Preference weights for services, interventions or oral health states; Marginal rates of substitution. Exclusion: Ranking or rating; Economic outcomes e.g. contingent values, willingness to pay (WTP) were excluded.

Study design: Multi-attributed stated choice experiments with service/interventions described in more than one dimension and preferences elicited between multiple options. Exclusion: Choice experiments that do not provide a relative value; Revealed preference experiments; Protocol papers; Abstracts where full text unavailable.

Language: Full text available in English or translation of report into English.

Titles were collected and imported into EndNote™ X4 (2010). Study selection and data extraction were performed by title, abstract and full text by two reviewers independently and in duplicate. Records were only excluded by title and abstract if obviously irrelevant based on study design, method or topic. Any disagreements

were resolved through discussion and the level of agreement rate was calculated. Studies excluded after full text review were indexed in Microsoft Excel v14.7.4 with reason for exclusion. A standardised data extraction form was used to extract the key information from each study. The following data items were extracted: author, date, country, topic, aim, design, participants, attributes, experimental design, and outcomes. The 10-point checklist published by ISPOR (Bridges et al. 2011) for Conjoint Analysis was used to assess specific quality markers of experimental design. Each of the 10 criteria in the checklist is composed of three questions, which were scored 0=incomplete and 1=complete, resulting in a maximum score of 30. Data extraction and quality assessment were performed by one reviewer (SB) then discussed and revised with co-authors until agreement was reached. Heterogeneity between studies precluded statistical synthesis of preference values and instead a descriptive analysis was undertaken.

Results

Electronic searches were completed on 22nd March 2017 (Figure 3). The database searches identified 773 records, from which eight met the inclusion criteria (Bech et al. 2011; Cunningham et al. 1999; Espelid et al. 2006; Gaeth et al. 1999; Kiiskinen et al. 2010; Krucien et al. 2015; Krucien et al. 2013; Ryan and Farrar 2000). A further four records were included from the grey literature search (Arora 2006; Douglas 2001; Lord et al. 2015; Zhang 2013) resulting in a total of 12 records for synthesis (Figure 2). A high number of records were case reports, laboratory studies and non-dental resulting in a high proportion of exclusions by title and abstract. The number of irrelevant records is likely to reflect the search terms used and the decision not to use filters in the search strategy. The agreement rate between reviewers at each stage was good.

The key data from studies included in the review is given in Appendix Table 2. Studies were published between 1999 and 2015 with the majority published in non-dental academic journals. Two were published by Universities as postgraduate theses (Douglas 2001; Zhang 2013) and one study formed part of a report undertaken by a University and Health Partner Consortium on behalf of NICE (Lord et al. 2015). The studies were all undertaken in high-income countries in Europe and the USA.

Use of multi-attribute stated preference methods

The studies aimed to elicit preferences for dental service delivery, treatment or oral health states from the perspective of the patients, dentists or the public (Table 1). Three studies were methodological investigations using dental service delivery as a model (Bech et al. 2011, Kiiskinen et al. 2010, Krucien et al. 2014). All studies used a common stated preference method, Discrete Choice Experiment (DCE), citing a desire to establish the relative importance of multiple attributes and to reflect real-life decision-making by requiring respondents to choose by making trade-offs.

Synthesis and critique of methods

Attributes varied from two to seven with little justification for the number selected. The attributes were most commonly related to cost (92%), time domains such as waiting time or time for treatment effect (66%), and different measures of effectiveness (50%). Effectiveness attributes included accuracy of diagnostic test, longevity of restoration, effectiveness of treatment, appearance and function. Participants from the target population were directly involved in identification of attributes in three studies (Douglas 2001; Espelid et al. 2006; Zhang 2013), two of which showed evidence of patient-centred attribute identification through interviews (Douglas 2001; Zhang 2013). The third identified attributes from a large general population survey and a dentist survey and verified the selection of attributes using a pilot study, however, no details of the exact methods were given. The remaining studies relied on existing literature and policies without use of patient or public involvement. The methods used to assign attribute-levels were reported in less detail.

There was little uniformity in experimental design, construction of tasks and survey design across the studies. The methods used for construction of tasks were most comprehensively reported. Full profiles were used in all studies, that is, each profile contained all attributes at varying levels. The number of profiles presented in each task varied from two to four alternatives and an opt-out or the option to retain the status quo was given in all but three studies. Generic labelling was used in all studies except one (Kiiskinen et al. 2010), which provided an explicit justification for use of labelling. In contrast the reporting of experimental design was less comprehensive. All studies used a fractional factorial design and three studies reported use of design software to optimise the experimental design (SPEED, SAS and nGene v1.1.1).

Reporting of design properties was highly variable and only two studies provided a statistical evaluation of the design (Lord et al. 2015; Ryan and Farrar 2000).

The total number of tasks ranged from twelve to 32, but in those with more choice tasks blocking was used to limit the number of tasks required of an individual respondent to a maximum of eight. One methodological study (Bech et al. 2011) focussed primarily on whether the number of tasks affected results by varying the number of tasks from 4 to 8 to 16. Elicitation of preference confidence (Bech et al. 2011) and strength of preference (Arora 2006; Ryan and Farrar 2000) were undertaken using rating scales, or by requesting respondents chose their most and least preferred option from a choice of three with an indication of how difficult the response was (Zhang 2013).

Generally, information regarding the sample, setting and recruitment process was inadequate; few studies reported an inclusion and exclusion criteria for participants and five studies provided no information about the recruitment process. Justification for sample size was provided in four studies (Douglas 2001, Krucien et al. 2013, Krucien et al. 2014, Zhang 2013) (Table 2). Douglas (2001) based sample size on minimum sample size requirements for regression analysis combined with 'rule of thumb' for conjoint analysis. The other three studies followed the parametric approach suggested by Louviere et al. (2000), which provides a minimum sample size for measuring choice probability with some degree of accuracy. These do not correspond to current proposed methods for sample size calculation (De Bekker-Grob et al. 2015). Varying methods of survey administration were used including postal (Douglas 2001; Zhang 2013), internet (Bech et al. 2011; Lord et al. 2015) and face-to-face (Espelid et al. 2006; Krucien et al. 2013; Krucien et al. 2015; Lord et al. 2015; Ryan and Farrar 2000). For the remaining studies the administration method was unclear. An explanation for tasks was reported in seven of the twelve studies (58%). Half the studies reported a piloting stage to assess aspects of experimental and survey design, including 'think aloud' cognitive interviews (Lord et al. 2015) and an extensive piloting using a small-scale version of the full survey (Douglas 2001; Zhang 2013).

All studies used advanced modelling methods for data analysis to produce estimates of attribute coefficients and from this relative importance of attributes, part-worth utilities and marginal rates of substitution, most commonly willingness-to-pay. Three studies used attribute values to estimate demand for a treatment (Cunningham et al.

1999; Gaeth et al. 1999; Krucien et al. 2013) and one study used the values to predict actual choice (Krucien et al. 2015). Subgroup analysis was reported in terms of participant demographics (Arora 2006; Espelid et al. 2006; Lord et al. 2015; Zhang 2013), participant behaviour (Kiiskinen et al. 2010) or participant groups based on experimental design (Bech et al. 2011). The internal validity of responses was tested by using a repeat question or including a task with one obviously superior (dominant) profile. The results of this were variably managed; one study performed a sensitivity analysis to determine the effect of inconsistent responders (Zhang 2013), one study reported the percentage of inconsistent responders but did not clarify how this was managed (Douglas 2001) and three studies excluded inconsistent responders (Cunningham et al. 1999; Krucien et al. 2015; Ryan and Farrar 2000).

The quality scores of the studies varied from 16/30 to 30/30 (Table 2). The highest scores were awarded to the University theses (Douglas 2001; Zhang 2013) and the Consortium report (Lord et al. 2015); two of these studies were recent and for all three, word count was not restricted as it is in an academic journal. Two further studies that scored well were those testing aspects of conjoint analysis methodology, published in Health Economics (Bech et al. 2011; Krucien et al. 2014), possibly because the purpose of the studies and the audience of these journals require a more comprehensive description of the methods, although generally papers focussing on methodological testing did not describe attribute selection in detail. Only one study published after the ISPOR guidelines in 2011 had notable absences in the quality assessment; this was published as a letter and although a supplemental file was available with additional information, there were limitations in the description of the survey instrument, data collection and analyses (Krucien et al. 2013).

Clinical implications of findings

The heterogeneity between studies prevented statistical synthesis, however, clinically relevant key findings are summarised in Table 3. This demonstrates the clinical application of preference data and allows some comparison of results across studies.

DISCUSSION

Preference elicitation methods provide an opportunity to understand patient and stakeholder preferences for the structures, processes and outcomes associated with dental care. This evidence can guide improvements to care at an individual and population level. Multi-attribute stated preference experiments have shown increasing popularity in health care, yet the number of studies identified for this review indicates the uptake is considerably slower in dentistry. There are several contributing factors likely driving this slow adoption: a lack of awareness of preference elicitation techniques secondary to their scarcity in dental journals, barriers to funding and publishing methods beyond testing treatment efficacy, the collaboration required to succeed with the complex experimental design and statistical analysis and, most importantly, the need for a paradigm shift towards a patient-centred approach to research and shared decision-making in dental care. Proposals for upcoming studies incorporating DCE methods suggest this is an emerging field with growing interest (de Silva-Sanigorski et al. 2011; Ke et al. 2013; Clarkson et al. 2013; Fleming 2016) and the increasing emphasis on incorporating values and preferences into healthcare decisions is likely to generate more opportunities for use of these methods in the future.

Multi-attribute stated preference methods ask respondents to consider multiple competing attributes simultaneously, which presents a decision-making process that is much more reflective of real-life choices than simple ranking or rating methods. This does, however, add a complexity to experimental design and analysis processes that may weaken studies if expert knowledge is not sought. The design of a valid and relevant preference experiment is dependent on identification and selection of attributes that are most relevant to the study perspective and most useful for informing translation into practice. The studies in this review largely under-reported how attributes and levels were identified and selected. Research bodies advocate the inclusion of patient and public representatives during the design and conduct of healthcare research and this, combined with rigorous qualitative methods involving the participants from the target population, ensures attributes are not limited to the researcher perspective.

Reporting of design features was variable across studies and even in cases where task construction and survey presentation were described, the rationale underpinning the decisions was often lacking. To enable critique and support better understanding

and advancement in methodology, it would be advantageous for reasons for design choices to be explained. For example, elicitation of responder certainty is one aspect of task construction that may have important implications of this for interpreting preference data (Lundhede et al. 2009; Regier et a. 2014), yet no studies explicitly stated why a measure of certainty was used or not. A further area of notable under-reporting was around the selection and recruitment of the sample. As with other research designs, it is expected that a clear inclusion and exclusion criteria are provided, there is transparency in recruitment and comparison is made between the study and target population. Preferences are influenced by values, experiences and environment, so it is critical that the sample population is fully described to enable interpretation of the results.

Piloting is a discrete stage in multi-attribute stated preference experiment development and due to the complexity of the preference elicitation, careful pre-testing is recommended. This allows the validity of attributes to be tested, ensures task construction and complexity is appropriate and provides data to inform the experimental design. Methods for assessing reliability and validity are ideally incorporated within the design (Janssen et al. 2017) and may also benefit from pre-testing. The ISPOR guidelines provide a comprehensive guide to all aspects of design and conduct and interested readers are referred to these reports (Bridges et al. 2011; Johnson et al. 2013; Hauber et al. 2016).

Cost attributes can be problematic in healthcare systems where treatment is free at the point of delivery, as respondents are not used to placing a monetary value on treatment. The widespread use of co-payment models for dental care in the UK and worldwide makes dentistry an attractive model for testing methodological issues related to willingness to pay (WTP). However, difficulties in anchoring cost attributes when respondents have existing price knowledge may present a different challenge for accurately estimating WTP and this needs to be tested further.

Multi-attribute stated preference experiments are likely to have an important role in the future for economic evaluation in dentistry. Lord et al. (2015) is the first study to use public preferences to value health states. Further work is required to determine a benchmark for valuing dental health for cost-effectiveness evaluation and measuring dental health improvement.

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CONFLICT OF INTEREST

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DATA

Full data set (all data extracted from the included studies) is available from the corresponding author by request.

FIGURE & TABLES LEGENDS

Figure 1: An example of five attributes with two or three attribute-levels for a hypothetical orthodontic assessment

Figure 2: Stages in designing a multi-attribute Stated Preference Experiment

Figure 3: Study selection process

Table 1: Use of multi-attribute stated preference methods in dentistry

Table 2: Quality assessment scores

Table 3: Clinically relevant findings from multi-attribute stated preference studies in dentistry

TABLES

Table 1: Use of multi-attribute stated preference methods in dentistry

Study	Topic	Purpose	Perspective
Preferences for dental intervention			
Douglas 2001	Caries detection device	Estimate attribute utilities and willingness to pay (WTP) for interventions	Patient and dentist
Espelid et al. 2006	Restorative materials		Patient and dentist
Zhang 2013	Dental prosthesis		Patient and dentist
Arora 2006	Tooth-whitening products	Estimate utility of attributes of products and effect of risk on choice	Public
Krucien et al. 2013	Sleep apnoea treatment	To estimate attribute utility and predict demand for treatment	Patients
Preferences for service delivery			
Cunningham et al.1999	Dental plan benefits	Estimate relative utility and importance of attributes and predict demand	Public
Gaeth et al. 1999	Dental plan benefits	Predict demand for difference configurations of plans and estimate willingness to wait for local service	Public
Ryan & Farrer 2000	Orthodontic service	Estimate utility of attributes and willingness to travel or wait.	Patients
Valuation of dental health states			
Lord et al. 2015	Decay, pain and removal of incisors, premolar and molar teeth and gum problems.	Estimate WTP values for oral health and economic modelling to estimate the cost-effectiveness of different health promotion methods	Public
Methodological investigations			
Bech et al. 2011	Dental service delivery	Test impact of survey presentation (number of choice sets)	Public
Kiiskinen et al. 2010	Dental service delivery	Test analysis methods on estimates of choice behaviour	Public
Krucien et al. 2014	Sleep apnoea treatment	Test the external validity of DCE methods	Patients

Table 2: Quality assessment scores

	Arora	Bech et al.	Cunningham et al.	Douglas	Espelid et al.	Gaeth et al.	Kiiskinen et al.	Krucien et al.	Krucien et al.	Lord et al.	Ryan & Farrar	Zhang
Question and choice of design	3	3	3	3	3	3	3	3	3	3	2	3
Choice of attributes and levels	3	0	3	3	3	3	0	1	0	3	1	3
Construction of tasks	2	3	3	2	2	3	3	3	3	2	3	3
Choice of experimental design	1	2	2	1	1	0	2	2	2	3	3	3
Preference elicitation	0	3	2	1	1	1	1	1	2	1	2	3
Data collection instrument	2	1	1	3	2	1	2	1	1	3	0	3
Data collection plan	0	2	1	2	1	1	1	2	3	2	0	3
Statistical analyses & model estimation	1	3	1	2	1	1	1	1	1	2	1	3
Results and conclusions	2	3	2	2	1	1	1	1	2	3	2	3
Study presentation	2	3	2	3	2	2	2	1	2	3	2	3
OVERALL QUALITY SCORE	**16**	**23**	**20**	**22**	**17**	**16**	**16**	**16**	**19**	**25**	**16**	**30**

Table 3: Clinically relevant findings from multi-attribute stated preference studies in dentistry

Attribute importance	
Caries detection device	Patients: discomfort > cost > accuracy > time (Douglas 2001)
Restorative material	Patients: appearance > longevity Dentists: longevity > appearance (Espelid et al. 2006)
Dental prosthesis	Patients: Longevity > appearance > function (Zhang 2013)
Sleep apnoea treatment	'Negative impact on daily life' > effectiveness > severity of side effects (Krucien et al. 2013)
Tooth whitening products	Duration of results > time taken to see effect > reporting of side effects > pricing (Arora 2006)
Dental plan benefits	Maximum annual benefit > coverage of orthodontic & restorative treatment > service delivery > premium (Cunningham et al. 1999; Gaeth et al. 1999)
Marginal rates of substitution	
Willingness to Pay (WTP)	Patients are willing to pay: £6.59 extra per check up to avoid discomfort £0.73 extra per check up for 1% increased accuracy £0.45 extra per check up for every one minute in the dental chair avoided (Douglas 2001)
	Patients are willing to pay: £3357 to change from artificial to natural looking appearance £2657 to change from compromised to good chewing function £31.37 for 1% improvement in treatment 5-year success rate (Zhang 2013)
	Respondents were willing to pay £333 to avoid removal of an anterior tooth Respondents were willing to pay £37 to avoid removal of a posterior tooth (Lord et al. 2015)
Willingness to Wait (WTW)	Patients are willing to give: 14.9 minutes extra in the dental chair to avoid discomfort 1.7 minutes extra for 1% increase in accuracy (Douglas 2001)
	1 month increase in waiting time reduces benefit score by 0.59 (Ryan and Farrer 2000)
Willingness to Travel (WTT)	Patients were willing to wait an extra 1.3 and 1.5 months for a local clinic for their first and second appointments (Ryan and Farrer 2000)
Risk equivalence	Patients require 16.2-17 years additional longevity from restoration to accept small risk of adverse reaction Dentists require 4.2-4.5 years additional longevity from restoration to accept small risk of adverse reaction (Espelid et al. 2006)