Ethical Considerations for Discrete Choice Experiments with Caregiving Populations

Judy Illes[1], Ashley Lawson[2], and Patrick McDonald[2]

[1]University of British Columbia
[2]Affiliation not available

February 08, 2022

Abstract

We discuss challenges experienced with a discrete choice experiment where the lack of sufficient responses to power analysis may have been due to the vulnerability of the population, added caregiving burdens imposed by the pandemic, and an imbalance of benefit-to-load of participation. We also address considerations for the paradigm where the necessarily few testable attributes may limit cultural meaningfulness and generalizability, and bring into question principles of research ethics.

Introduction

Discrete Choice Experiments (DCE) have been used since 1983 to identify preference-sensitive variables where multiple, equally valid, options exist [1]. DCEs force choices between hypothetical conditions, described by key features known as attributes. They have been applied in a wide range of disciplines – marketing, economics, and ecology– and more recently, have gained prominence in health care [2, 3, 4]. From the latter, results have yielded new knowledge about valuing patient experiences, investigating trade-offs in health outcomes and patient experiences, and developing priority-setting frameworks, with a direct impact on treatment [5]. In these circumstances, the best option depends on patients, together with their clinicians, choosing a treatment that aligns best with their preferences. As described by Apakantu et al. (2021), these preferences can be influenced by outcomes (potential benefits and harms), as well as processes (e.g., the way treatment is delivered), and the context (i.e., structures) in which treatment is delivered (e.g. the health care team) [6]. In a study that used a modified DCE to elicit treatment preferences from parents of children with hydrocephalus; surgical risk, minimizing repeat operations, and maximizing cognitive outcomes were the most important attributes driving treatment choice [7].

However, even the best designed research with human participants will fail if not well-suited to its target population. In a study of caregivers of children with drug resistant epilepsy (DRE), our goal was to collect information from both physicians and caregivers on the components they value when it comes to deciding on a neurotechnological intervention. Such treatments may involve invasive implantation in the brain or nerves such as deep brain stimulation or vagus nerve stimulation, or less invasive interventions that still involve the destruction of epileptogenic tissue but do not require open surgery or an incision. We aimed to use the data to determine how specific attributes influence decision making from both the perspectives of physicians and caregivers and, ultimately, to analyze where their values align and diverge. We were successful in obtaining a sufficient number of responses with the physician DCE based on the preparatory qualitative study [8], but unsuccessful in obtaining enough responses in a caregiver DCE, even though the attributes were derived, and DCEs designed using the same methodology. The contributions to this failure, and focus of this report, involve not only a possible methodological mismatch with the target population, but considerations that fall within the realm of research ethics.

Methodological approach

The first step to the caregiver DCE study was to determine the attributes that were most important to the target populations to use. DCEs are underpinned by random utility theory/Lancaster's theory of demand [9, 10]. The theory provides a framework for preference elicitation methodologies by distilling decision-making into its component parts, i.e., decision-making about goods/services can be decomposed into combinations of their attributes and levels, each with different values. Guidelines on the development of DCEs recommend that relevant attributes and levels should be identified by qualitative research [11].

To accomplish this, caregivers with affected children were recruited to semi-structured focus groups or interviews at one of four major epilepsy centers in Eastern and Western Canada and the USA (n=22) [12]. Discussions were transcribed and qualitative analytic methods applied to examine values and priorities (e.g., risks, benefits, adherence, invasiveness, reversibility) of caregivers pertaining to novel technologies to treat DRE. An inductive approach was used to identify major themes: 1) features of the intervention: risks and benefits, with an emphasis on an aversion to perceived invasiveness; 2) decision drivers: trust in the clinical team, treatment costs; and, 3) quality of available information about neurotechnological options. Overall, we found that caregivers' definition of treatment success is more expansive than the single variable of seizure freedom often favored by clinicians.

The DCE based on these results was designed by an interdisciplinary team of health economists, ethicists, clinicians and health service researchers. In developing the full set of attributes for the experiment, the core question was: *"Does this drive decision-making ?"* Selecting attributes is a balance between including the most important features, without providing too many attributes which increases the complexity of the tasks for the respondents, and can result in attribute non-attendance, inconsistent responses across choice tasks, and increased dropout rates [13]. The experts and researchers screened all generated attributes and levels using multiple criteria such as decision context and plausibility. As a decision to choose the option of no intervention was plausible, a no intervention opt-out was carefully defined. Multiple versions of the DCE were vetted and refined by the research team as well as select stakeholders who participated in the focus groups and consented to recontact. In testing, time to completion was an average of 25 minutes. The final version was translated from English into both French and Spanish for maximum inclusivity of prospective respondents residing in both Canada and the USA. Minimum N to power the analysis was 70 from a possible pool of 150,000.

Dissemination of the DCE was conducted via recruitment posters posted in epilepsy clinics at the 4 sites involved with the caregiver focus groups featuring scannable QR codes, as well as via a dissemination partner at the Brain Recovery Project who shared the survey links to online patient support networks and social media platforms. An invitation to complete the DCE was posted on the Brain Recovery blog and Facebook page and reposted a month later as a follow-up, shared with pediatric epilepsy surgery support groups, and the rare epilepsies network representing 70 patient advocacy groups.

Outcomes

Nine responders completed the DCE choice sets, including the demographics questions; 10 completed only the DCE choice sets; 15 went past the introduction and consent pages but failed to proceed into the choice sets; 5 began the choice sets and but terminated before completion.

Observations and Discussion

While the simplicity of the DCE exercise and the familiarity of making choices in real-life situations has been cited as important strengths of the approach [14], the response numbers were insufficient to power any analysis. We received feedback from collaborators engaged to assist with dissemination of the DCE that the real-life burden of caring for children with DRE, and the out-of-context nature of the experiment were major impediments to participation.

While we took into account strategies and recommendations of past studies on the methodology of DCE that have addressed ways to reduce cognitive load and low response rates [15, 16], the choice task still in-

volves a considerable cognitive challenge as respondents are required to process large amounts of information contained in the scenarios and consider trade-offs between all of the attributes. Decreasing the number of attributes from 6-8 attributes to 2-4 attributes might have increased response rate, but the meaningfulness of the experiment, and the generalizability of the results, would decrease proportionally [17]. Further, within the healthcare setting, choices may be less familiar to respondents than, for example, marketing choices, making each choice more burdensome or complex such that individuals more quickly reach a threshold beyond which they are unwilling to participate, and thus fail to complete the DCE [18]. Compounding the issue, cognitive load has already been heightened by the ongoing COVID-19 pandemic and the implications for caregivers of young children navigating care and safety of their school-aged children. We did not offer any incentive to participate. The direct effect or interaction of these choices with the current public health crisis is unknown.

We note independently that the requirement for only 6-8 attributes, all ultimately related to biomedical aspects of candidate neurotechnologies, excluded variables involving diverse cultural views about interventions on the brain. For participants for whom wholly biomedical explanations of disease is not sufficient, or for those living in rural and remote geographic regions with challenging access to advances to new technologies [19], the relevance of the DCE was low. Indeed, should the study actually have recruited sufficient responses to power the analysis, adoption of results would have excluded their views at best; at worst, they would only serve to further stigmatize and marginalize them, and increase a gap in already well documented health disparities.

The suitability of DCE to certain vulnerable populations, however vulnerability is defined, is a matter not only of economics and science, but of research ethics. Respect for persons is violated if burden – i.e., risk – exceeds, benefit. Context matters. Inclusivity is at stake given the technical limitations and cognitive load elements of the approach. Meeting the goal of justice is a challenge if implementation falls short of meaningfulness. More discussion during study planning and design, as well as institutional review is needed to ensure that the highly validated, quantitative approach of the DCE method also meets the highest and broadest standards of ethics in research.

Acknowledgements

Author Contributions

JI developed the concept for this report. JI, AL and PJM interpreted the previous efforts and wrote the manuscript. All authors discussed the report and reviewed the manuscript at all stages.

References

1. Elwyn G., Frosch D., & Rollnick S. Dual equipoise shared decision making: Definitions for decision and behaviour support interventions. *Implement Sci* **4,** 1-8 (2009).
2. Louviere, J.J. & Woodworth, G. Design and Analysis of Simulated Consumer Choice or Allocation Experiments: An Approach Based on Aggregate Data. *Journal of Marketing Research* **20** , 350-367 (1983).
3. OECD, *Cost-Benefit Analysis and the Environment: Further Developments and Policy Use Ch. 5* (OECD Publishing, Paris, 2018).
4. Ryan M. Discrete choice experiments in health care. *BMJ* **328,** 360–361 (2004).
5. Soekhai, V., de Bekker-Grob, E.W., Ellis, A.R., & Vass, C.M. Discrete Choice Experiments in Health Economics: Past, Present and Future. *Pharmacoeconomics* **37** (2)**,** 201-226 (2019).
6. Apakantu, G., Aguilar, M., Kaal, J.J., McDonald, P.J., Connolly, M., *Hrincu, V* ., Illes, J., & Harrison,

M. Understanding Attributes that Influence Physician and Caregiver Decisions About Neurotechnology for Pediatric Drug-Resistant Epilepsy: A Formative Qualitative Study to Support the Development of a Discrete Choice Experiment. *Patient,* 1-14 (2021).

7. Tamber M.S, & Naftel R.P. Patient and parental assessment of factors influencing the choice of treatment in pediatric hydrocephalus.*JNS Pediatrics* **26,** 490-494 (2020).

8. McDonald, P.J., Hrincu, V.M., Connolly, M.B., Harrison, M., Ibrahim, G., Naftel, R., Chiong, W., Udwadia, F., & Illes, J. Novel Neurotechnological Interventions for Pediatric Drug-Resistant Epilepsy: Physician Perspectives. *Journal of Child Neurology***36** , 222-229 (2021).

9. Mcfadden D. *Frontiers in Economics Ch. 4* (Academic Press, New York, 1973).

10. Lancaster K. *Consumer Demand: A New Approach* (Columbia University Press, New York, 1972)

11. Lancsar E. & Louviere J. Conducting Discrete Choice Experiments to Inform Healthcare Decision Making. *PharmacoEconomics***26** , 661–77 (2008).

12. Hrincu, V.M., Harrison, M., Connolly, M.B., McDonald, P.J., Naftel, R., Ibrahim, G., Chiong, W., & Illes, J. Choice and Trade-Offs: Parent Decision-Making for Neurotechnologies for Pediatric Drug Resistant Epilepsy. *Journal of Child Neurology* **36** , 222-229 (2021).

13. Obadha M., Barasa E., Kazungu J., Abiiro G.A., & Chuma J. Attribute development and level selection for a discrete choice experiment to elicit the preferences of health care providers for capitation payment mechanism in Kenya. *Health Econ Rev* **9** (2019).

14. Ryan M. & Farrar S. Using conjoint analysis to elicit preferences for health care. *BMJ* **320,** 1530–1533 (2000).

15. Coast J., Al-Janabi H., Sutton E.J., Horrocks S.A., Vosper A.J., Swancutt D.R., & Flynn, T.N. Using qualitative methods for attribute development for discrete choice experiments: issues and recommendations. *Health Econ* **21** , 730–41 (2012).

16. Bryan, S. & Dolan, P. Discrete choice experiments in health economics. *Eur J Health Econom* **5,** 199–202 (2004).

17. Watson, V., Becker, F., & de Bekker-Grob, E. Discrete Choice Experiment Response Rates: A Meta-Analysis. *Health Econ***26** (6)**,** 810-817 (2017).

18. Vineya, R., Savagea, E., & Louviere, J. Empirical investigation of experimental design properties of discrete choice experiments in health care. *Health Econ* **14** (4)**,** 3489-62 (2005).

19. Harding, L. & Illes, J. Response to: Canadian Assessment of Deep Brain Stimulation Access: The Canada Study. *Can J Neurol Sci***48,** 130-131 (2021).