A discrete-choice experiment to elicit preferences of patients with epilepsy for self-management programs

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ABSTRACT

Background: There is an increasing number of self-management programs developed for patients with epilepsy, with the goal of supporting treatment management and improving their quality of life. With the aim of increasing medication adherence and effectiveness of self-management programs, it is important to design programs that are engaging to, and align with the preferences of patients with epilepsy. This study aimed to evaluate and compare the preferences of patients with epilepsy for self-management programs in three European countries. This is the first cross-border evaluation of the preferences of patients with epilepsy in Europe for such programs.
Methods: Using a discrete-choice experiment, patients with epilepsy from Germany, France, and the Netherlands were surveyed, and chose repetitively between two hypothetical self-management programs. These differed in the following six characteristics: i) the thematic area which would be the main focus of the program, ii) the method of interaction, iii) the source of information or provider of the program, iv) the amount of time spent on the program per week, v) the cost, and vi) whether the program would start immediately, or if there would be a delay of 3 weeks before its initiation. A Bayesian efficient design was used to construct 15 choice sets, and a mixed panel logit model was used to estimate patients' preferences. Subgroup analyses were conducted according to socioeconomic status, burden of disease, and previous activation in self-management.
Results: A total of 299 people with epilepsy were included in the study, with a mean age of 45.5 years. Only 15% had previously made use of a self-management program, although 44.5% reported having previously heard of them. In all three countries, all attributes barring the content were significant at 10%. The cost attribute – i.e., an out-of-pocket expenditure for a program – was reported as the most important feature in each country and across subgroups (significant at 1%). This was followed by the length of program sessions per week, which ranged from 20 to 90 min per week. Although there was some heterogeneity between countries and subgroups, the patients, overall, had a preference for a face-to-face meeting with a doctor. In the Netherlands, a preference for online programs and physician assistants was observed when compared with the other countries. Other attributes, including the information source – whether a program was led by a physician, another patient with epilepsy, or another combination – was also important to patients, who appear willing to trade preferences in order to gain their favored attribute level. However, 20% of the population chose consistently to not participate in any self-management program.
Conclusion: Given the heterogeneity of the epilepsies, preferences, and dispreferences across subgroups, our study highlights that if full account is not taken of different segmentation strategies when designing a self-management program, a large proportion of the population may not be attracted to it.

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1. Introduction

Epilepsy is a severe brain condition which has a significant impact on patients' quality of life, on their families, and their communities [1,2]. The main feature of the condition is a predisposition to recurrent unprovoked seizures [3], and it is one of the most common noncommunicable neurologic diseases worldwide [4]. While many people with epilepsy (PWE) can reduce the frequency of their seizures with medication, this is often not a simple endeavor, potentially requiring complex

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dosing schedules [5]. Between 30% and 60% of PWE are thought to be nonadherent to their medication [6], and nonadherence is thought to be responsible for around 30% of all epileptic seizures in those diagnosed with epilepsy [7].

Self-management (SM) has become an interesting approach to increase the effectiveness of treatment strategies, and positively affect the patient's quality of life, as well as potentially constrain the long-term cost of care. As it is a spectrum ranging from the provision of information to active education and electronic support, all ultimately with the intention of providing insights into disease and treatment, SM cannot be considered as a discrete set of actions or tools [8]. It has the potential to reduce long-term care costs through both a shrinking of resources used per patient from the standpoint of medical institutions [9,10], and increasing adherence to therapy, reducing seizure occurrence [11]. Following the implementation of SM programs (SMPs), an increase in quality of life for chronic disease patients in general has been observed [12], as well as clinically meaningful results following use of electronically delivered programs, e.g., in prediabetes and diabetes [13,14]. Nevertheless, the Cochrane collaboration has reported limited evidence of efficacy in epilepsy SMPs [15]. One of the reasons is that SMPs can be challenging for some PWE, and adherence to programs is not always optimal, with some PWE not completing them either in person or online because of scheduling or other reasons [16,17].

Despite a growing number of SMPs being developed for PWE in both the private and public sectors [18,19], there is a paucity of research on the preferences and needs of this target group, particularly outside of the United States (US) [20]. As the development of SMPs continues, it is important that they both improve in delivering desired outcomes, as well as their attractiveness for their target population. Given that patient subgroups (such as age, gender, disease burden, or socioeconomic status) may have different preferences and treatments for different conditions can have a range of different attributes, it is important to ensure that these preferences are assessed correctly. This knowledge can then be used to develop or customize future programs and interventions. It has been seen that patients' use of healthcare services is significantly impacted by their preferences [21], and therefore using patient preference information in program development may well increase recruitment and retention. Patient preference information is also beginning to be introduced into regulatory submissions, such as with the Food and Drug Administration, suggesting that patients' preferences will increasingly be considered with regards to risk/benefit tolerance [22,23].

However, empirical data are scarce for the relative priority given by PWE to the different possible attributes of SMPs, and whether there are differences between countries within Europe with regards to patients' preferences toward them.

Using a discrete-choice experiment (DCE), this study aimed to assess preferences of PWE in France, Germany, and the Netherlands for the characteristics of SMPs, and to provide a comparison between the three countries and across three subgroup populations. These are burden of disease, socioeconomic background, and level of previous "activation" in self-management.

2. Methods

2.1. Discrete-choice experiment (DCE)

We used a DCE to assess the preferences of PWE for SMPs. A DCE is an attribute-based stated-preference valuation technique [24]. In a DCE, the preferences of participants (in this case, patients) are elicited through questionnaires consisting of repetitive choices of two (or more) options that differ according to a list of attributes. DCEs have increasingly been used in healthcare [24] for medication and drug attributes (i.e., [25,26]), as well as for nondrug medical choices (i.e., [27]) and outcome preferences (such as [28]). This technique is decompositional, as products or interventions are considered merely a product of their own components (attributes) [29]. These attributes

have differing levels. By varying each level within each attribute for each question, it is possible to develop scenarios for each choice. After the completion of the survey, it is possible to analyze the results through the use of a regression model. This analysis is based on the notion that preferences of respondents can be derived based on the choice variation between levels [30].

2.2. Identifying attributes and levels

Identifying appropriate attributes and levels is important for the generation of valid results [31,32]. We conducted a literature review of SMPs for epilepsy, including meta reviews and original research, to identify the most important characteristics for them [15,17,20,24, 33–37]. In addition, we evaluated clinically validated SMPs for other conditions, notably in the field of diabetes [13,14,37,38]. The list of potential attributes and levels was further discussed and validated by clinical experts (n = 3) in epilepsy and SM through semi-structured interviews. Experts were asked to comment on the relevance and applicability of the attributes, and the applicability of these attributes to "real-world" SMPs with which participants may be familiar. The total number of attributes was limited to six, to limit cognitive burden and complexity of evaluation of different features [24] and included the following: i) the thematic area which would be the main focus of the program, ii) the method of interaction, iii) the source of information or leader of the program, iv) the amount of time spent on the program per week, v) the cost, and vi) whether the program would start immediately or 3 weeks after their decision to begin it. The final list of attributes and levels is in Table 1.

The levels in which the "main topic of the program" component could focus were adapted from the US Institute of Medicine's (IOM) definition of SM, which splits activities into the following three groups: medical management, role management, and emotional management [39]. Our experiment also included a feature found in some novel interventions considered to be distinct from these three, which is the personal tracking of a condition. The basis for the other attributes in this experiment was the work of Fraser et al., which assessed these in the US as part of a study associated with the Managing Epilepsy Well network [20].

Table 1
Attributes and levels.

Attributes	Levels
Main program component	Disease management
	Role management
	Emotional management
	Self-monitoring
Method of interaction	Personal meeting (face-to-face)
	Telephone meeting
	Group meeting
	Smartphone based
	Online coach
Information source	Physician
	Physician assistant/epilepsy nurse
	Physician/PWE expert combination
	Other PWE
	Computer-tailored advice
Time per week	20 min
	40 min
	60 min
	90 min
Cost for you	Free
	5€/week
	10€/week
	25€/week
Length of time until access	Immediate
	Delayed (after 3 weeks)

PWE, patients with epilepsy.