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Patient and caregiver preferences for the potential benefits and risks of a seizure forecasting device: A best–worst scaling☆

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ABSTRACT

Background: Epilepsy is the 4th most common neurological disorder and is characterized by recurrent, unpredictable seizures. The ability to forecast seizures is a significant unmet need and would have a transformative effect on the lives of people living with epilepsy. In an effort to address this need, the Epilepsy Foundation has committed effort and resources to promote the development of seizure forecasting devices (SFD).
Objective: To promote user-centered design of future SFD, we sought to quantify patient and caregiver preferences for the potential benefits and risks of SFD.
Methods: A community-centered approach was used to develop a survey incorporating a novel best–worst scaling (BWS) to assess preferences for SFD. A main-effect orthogonal array was used to design and generate 18 "prototypes" that systematically varied across six attributes: seizure forecasting probability, seizure forecasting range, inaccuracy of forecasting, amount of time required to use the device, how the device is worn, and cost. The dependent variable was the attributes that respondents selected the best and worst in each profile, and a choice model was estimated using conditional logistic regression, which was also stratified and compared across patients and caregivers. Respondents also indicated that they would accept each of the prototype SFDs if it were real. These acceptance data and net monetary benefits (relative to the least preferred SFD) were explored.
Results: There were 633 eligible respondents; 493 (78%) completed at least one task. Responses indicated that 346 (68%) had epilepsy, and 147 (29%) were primary caregivers or family members of someone with epilepsy. The data show that short forecasting range is the most favored among experimental attributes, followed by mid forecasting range and notification of high chance of seizure. Having the device implanted is the least favorable attribute. Stated preferences differed between patients and caregivers ($p < 0.001$) for range of forecasting and inaccuracy of device. Caregivers preferred any range of forecasting, regardless of length, more than patients. Patients cared less about inaccuracy of the device compared to caregivers. The groups also differ in impact of fear of having seizures (versus actually having seizures) ($p = 0.034$) and on device acceptance. The acceptance of devices ranged from 42.3% to 95%, with caregivers being more likely to use a device ($p < 0.05$) for the majority of device profiles. Acceptance of devices varied with net monetary benefit of the best device being $717.44 more per month relative to the least preferred device.
Conclusion: Our finding extends previous calls for seizure forecasting devices by demonstrating the value that they might provide to patients and caregivers affected by epilepsy and the feature that might be most and least desirable. In addition to guiding device development, the data can help inform regulatory decisions makers.

1. Introduction

Epilepsy is the 4th most common neurological condition in the United States [1] and impacts more than 50 million people living worldwide [2]. People affected by epilepsy have unprovoked episodes of abnormal brain activity called seizures. Seizures can manifest in a number of ways (e.g., motor, sensory, cognitive) that involve the whole body or be limited to one part [3]. Moreover, there is heterogeneity in the severity and frequency of the seizure event across the population. Adding to this complexity are the many potential underlying

Abbreviations: SFD, Seizure Forecasting Device; PWE, People with epilepsy; BWS, Best–worst scaling; NMB, Net Monetary Benefit.

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causes for epilepsy from genetics, brain structure abnormalities, metabolism changes, immune system abnormalities, infectious disease, and unknown causes [4]. A third of people living with epilepsy do not have seizure control [5], and those whose seizures are controlled still remain at risk of breakthrough seizures. The Epilepsy Foundation Community Survey found that the unpredictability of seizures puts an immense psychological burden on the individual and greatly impacts quality of life.

The epilepsy community desires a seizure prediction device that can accurately gauge the likelihood of impending seizure [6]. Technological progress has made that more attainable, and led to the first human trial for an implantable seizure warning system in Australia. Its goal was to demonstrate the viability of seizure forecasting in long-term recordings for people with uncontrolled seizures [7]. Seizure forecasting shifts away from categorical seizure prediction assessments of whether a seizure will or will not occur and instead focuses on identifying the brain state wherein there is a high probability of a seizure occurrence. Although the trial demonstrated that seizure forecasting algorithms were possible [8], it also raised concerns over whether the potential benefits justify the risks of the invasive device. The Food and Drug Administration (FDA) has not approved a clinical trial solely for seizure forecasting in the United States. An implantable system recently confirmed the feasibility of developing seizure forecasting algorithms with intracranial electroencephalogram (EEG) [9] in conjunction with therapeutic stimulation. With advances in neural engineering allowing for less invasive EEG systems [10] and improvements in the robustness of forecasting algorithms [9,11], minimally invasive or noninvasive seizure forecasting technology may become an available tool in the foreseeable future.

The mission of the Epilepsy Foundation's Epilepsy Innovation Institute (Ei^2) is to drive novel improvements in the diagnosis and treatment of epilepsy. The Ei^2 launched the My Seizure Gauge Initiative in 2017. The goal was to create a minimally invasive personalized seizure advisory system to assess the likelihood of seizure on a timescale of hours before possible occurrence. The desire was to leverage biosensors, electrical recordings (EEG), and deep machine learning to improve upon current concepts and create personalized algorithms for people living with epilepsy [12]. As part of the initiative to foster the development of a seizure forecasting device, a community-based approach was used to develop and implement a survey instrument to quantify preferences. It followed the patient preference framework outlined by the FDA and other emerging best practices [13–15]. This study sought to engage the community earlier in the research and development process to assess the stated preferences of people with epilepsy (PWE) and caregivers and to address future FDA concerns regarding the community's acceptance of such a device.

2. Materials and methods

2.1. Experimental design

Best–worst scaling (BWS) case 2, which is called the profile case as it shows one profile at a time, was used to measure preferences and study what factors people impacted by epilepsy value most in a hypothetical seizure forecasting device [16]. The BWS is a survey method for assessing individuals' priorities that identifies the extremes — best and worst items, most and least important factors, and biggest and smallest influences — among sets of three or more items [17]. The BWS has been previously employed to investigate preferences over a wide range of healthcare topics [18], including preferences for medical devices [19]. In the BWS instrument, respondents were presented with 18 different hypothetical device profiles and asked to judge which aspects they thought were the best and the worst based on individual preference (Fig. 1).

The 18-device profiles were generated based on attributes (different aspects of the device) and levels (amount of impact of those attributes) following the current international best practices to identify the values of PWE and stakeholders [20]. The attributes and levels were generated from qualitative interviews with Foundation leadership, healthcare professionals ($n = 3$), and people impacted by epilepsy ($n = 11$), including 4 people living with epilepsy and 7 caregivers. These qualitative interviews focused on potential benefits and risks of having a seizure forecasting device and followed a reiterative process, with insights from the last interview incorporated into the next. This approach, modeled after a community-engagement process described in Hollins et al. [21], resulted in six distinct attributes that generated our device combinations. The six device features include seizure forecasting probability, forecasting range, inaccuracy of forecasting, time burden, location worn, and monthly cost — each defined across three relevant levels (see Table 1).

A 3^6 main-effects orthogonal design, identified from the SAS database of orthogonal arrays, was applied to the available attributes and levels (Table 1) to generate the 18 hypothetical devices for the BWS tasks in the survey [22]. Eighteen device profiles were the minimum necessary to ensure no correlations existed between the attributes. The design is balanced, i.e., all attribute levels appear the same number of times, and orthogonal, i.e., all pairs of attribute levels appear the same number of times, thus achieving optimal efficiency [23]. All respondents were presented all 18 device profiles, but the attribute order was randomized for each respondent. Immediately following each device profile, we also asked respondents if they would use the device were it available. The survey also included questions related to demographics, seizure experience, risks and assessment of risk behavior, and personality characteristics. This study was approved and deemed IRB exempt by the Western International Review Board (WIRB).

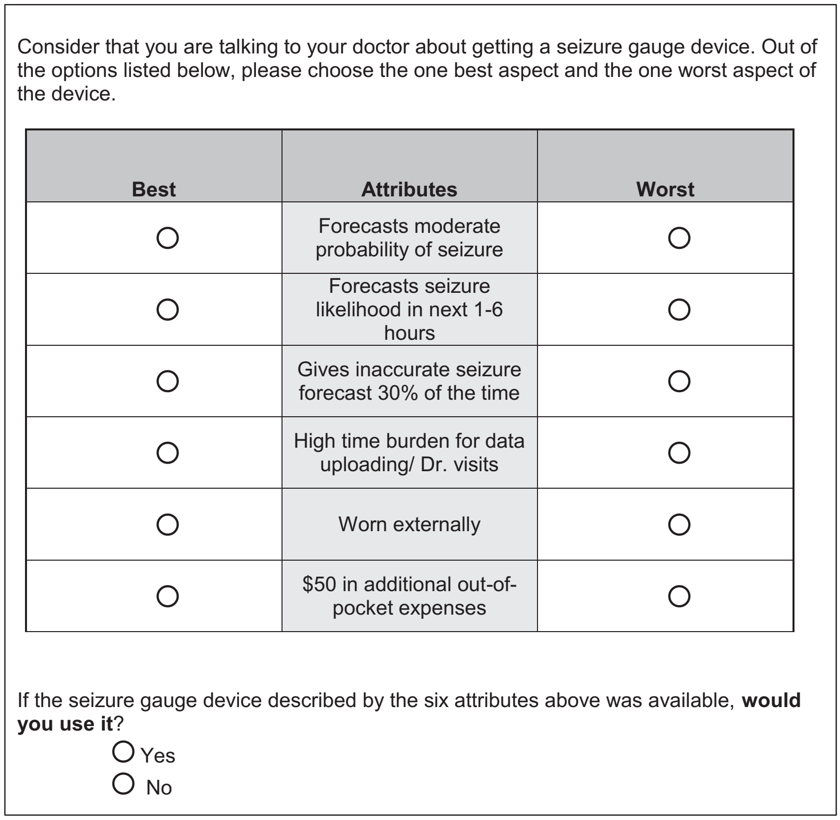
2.2. Survey distribution

The survey was generated using Qualtrics (Provo, Utah) and distributed online. Multiple channels were used to ensure as widespread of a distribution as possible. The survey was conducted between November 2017 and June 2018. Participants were required to be at least 18 years of age and be a person with epilepsy, a primary caregiver of someone with epilepsy, or a family member of someone with epilepsy.

2.3. Analysis

Responses from participants that completed at least one BWS task were included in the analyses. Descriptive statistics between PWE and caregivers were compared using Fisher's exact test or a *t*-test, as appropriate. All tests of significance were two-sided and statistical significance was established at $p < 0.05$.

Two methods were used to analyze the BWS data: best-minus-worst scoring and conditional logistic regression. In best-minus-worst scoring, how many times a level was chosen as worst is subtracted from how many times it was chosen as best to calculate the best-minus-worst (B-W) score. This number is then divided by the total times that the attribute was offered to calculate the relative B-W score. Although this technique has previously been shown to be highly (and often perfectly) correlated with more advanced statistical techniques such as conditional logit regression, analysis was also done via a conditional logit model in order to allow for other more complex analyses, such as estimating preference coefficients [24,25]. A conditional logit regression is similar to a logistic regression model but also incorporates dependency in the data. Here, the conditional logit approach was used to model both aggregated and stratified device preferences for both populations of participants (patients and caregivers/family members). For the conditional logit regression analyses, preferences were estimated through the use of a single anchored dummy coded variable. Here, the middle time burden level was coded as the anchored dummy variable based on the best–worst score. Survey responses for each choice task were coded into two dichotomous variables, one for the best selection and


Consider that you are talking to your doctor about getting a seizure gauge device. Out of the options listed below, please choose the one best aspect and the one worst aspect of the device.

Best	Attributes	Worst
○	Forecasts moderate probability of seizure	○
○	Forecasts seizure likelihood in next 1-6 hours	○
○	Gives inaccurate seizure forecast 30% of the time	○
○	High time burden for data uploading/ Dr. visits	○
○	Worn externally	○
○	$50 in additional out-of-pocket expenses	○

If the seizure gauge device described by the six attributes above was available, **would you use it**?

○ Yes
○ No

Fig. 1. Example task from survey.

one for the worst. All attributes were checked for violations to monotonicity. Preferences for the aggregated and stratified models were estimated from the data using the clogit command in Stata version 15 (StatCorp, College Station, TX, USA) with robust standard errors to account for clustering at the individual level. Preference differences between the respondent types were assessed via a Wald test and the Swait–Louviere test was used to assess if identified differences were due to scale heterogeneity [26,27].

Differences in device acceptance between PWE and caregivers were examined using respondents' answers to the question “If the seizure gauge device described by the six attributes above was available, would you use it?”, which immediately followed each device profile BWS task (Fig. 1). These differences were tested via a two-sample test for equality of proportions.

Willingness to Pay (WTP) modeling can be applied when respondents make trade-offs between attributes and cost [28]. As there was a cost associated with each hypothetical device profile, we could calculate the WTP for individual attribute levels. The issue with simply calculating WTP for each attribute level and then summing over the attributes for each device is that the resulting value is without reference to anything, making the interpretation difficult. Thus, we examined the net monetary benefit (NMB) for each hypothetical device with respect to the least accepted device. The profile of the least accepted device (device *R*, Table 4) was comprised of high seizure chance notification, long forecasting range, 30% inaccuracy, high time burden, and device implanted in the brain. Cost was recoded as a continuous variable, and the other attribute levels were recoded using dummy coding from the reference device *R* in order to calculate the net benefit for any given device. The levels of

Table 1
Attributes and levels.

Attribute	Description	Levels	Levels (Choices)
Seizure chance	Seizure forecasting probability	Low	Forecasts low probability of seizure (i.e., 1% chance)
		Moderate	Forecasts moderate probability of seizure (i.e., 60% chance)
		High	Forecasts high probability of seizure (i.e., 80% chance)
Forecasting range	Seizure forecasting range	Short	Forecasts seizure likelihood in the next 1–10 min
		Mid	Forecasts seizure likelihood in the next 1–6 h
		Long	Forecasts seizure likelihood in the next 12–24 h
Percent inaccuracy	Inaccurate forecasting	10%	Inaccurate seizure forecasting 10% of the time
		20%	Inaccurate seizure forecasting 20% of the time
		30%	Inaccurate seizure forecasting 30% of the time
Time burden	Amount of time and work required to use the device	Low	Up to 5 min weekly uploads and a once a month doctor visit for the first 3 months
		Moderate	Up to 1 h weekly uploads and twice a month doctor visits for the first 3 months
		High	Up to 7 h weekly uploads and twice a month visit for the first 3 months
Location worn	How the device is worn	External	Worn externally on your body (like a watch or jewelry)
		Subcutaneous	Inserted under the skin
		Implant	Implanted in the brain
Monthly cost	Cost of the device	$50	$50 a month in additional out-of-pocket expenses
		$150	$50 a month in additional out-of-pocket expense
		$200	$200 a month in additional out-of-pocket expenses

Table 2
Patient and respondent characteristics.

Variable	Person with epilepsy (N = 346)	Primary caregiver or family member (N = 147)	Total (N = 493)	p-Value
PWE characteristics				
Age – mean (SD)	43.0 (13.7)	20.3 (13.2)	36.3 (17.0)	<0.0001
Do the seizures come in clusters (i.e., three or more in a day)? – (%)				0.3515
No	57.8%	60.0%	58.5%	
Yes	32.7%	34.5%	33.2%	
Not sure	9.5%	5.5%	8.4%	
Reside in the US – (%)	87.9%	84.5%	86.9%	0.4131
Female – (%)	74.8%	45.5%	66.1%	<0.0001
Has had brain surgery – (%)	28.4%	16.0%	24.8%	0.0080
How often do seizures occur? – (%)				0.4318
Every day or at least once a week	28.3%	32.4%	29.5%	
Between 1 and 3 times a month or every other month	31.5%	31.7%	31.6%	
Between 1 and 5 times a year or less than once a year	28.6%	29.0%	28.7%	
Not sure/other	11.6%	6.9%	10.2%	
Currently has a device implanted in the body – (%)	21.1%	26.9%	22.8%	0.2413
Respondent characteristics				
Optimistic – (%)	54.7%	65.8%	57.9%	0.0552
Self-control – (%)	65.7%	64.6%	65.4%	0.9067
Improving health – (%)	79.9%	76.1%	78.8%	0.4157
Risk taker – (%)	33.7%	25.7%	31.4%	0.1491
Good with numbers – (%)	46.8%	49.1%	47.4%	0.7369

device *R* were chosen as the reference categories for each attribute. After estimation via a conditional logit model, the individual WTP values for each attribute level *k* were calculated with respect to the reference level by dividing the coefficient for that attribute level, β_k, by the coefficient for the cost attribute, β_c. This is generally estimated using Eq. (1).

$$WTP_k = \frac{-\beta_k}{\beta_c}\# \tag{1}$$

The individual WTP values for each attribute level represented in a hypothetical device were summed and the monthly out-of-pocket cost associated with that device was subtracted in order to calculate the net benefit for each device, *d*.

$$Net\ benefit_d = \sum WTP_k - Cost_d\# \tag{2}$$

The NMB for a particular device, *d*, was then calculated by taking the difference between the net benefit value and the cost associated with device *R*.

$$NMB_d = Net\ benefit_d - Cost_R\# \tag{3}$$

This value can then be interpreted as the additional monthly cost that respondents were willing to pay for this device over the least preferred device.

3. Results

A total of 637 respondents completed the survey. Four respondents were excluded because they did not meet the inclusion criteria, leaving a total of 633 eligible respondents, of which 493 (78%) completed at least one task. The majority of respondents were people who self-reported having epilepsy (70.2%) and reside in the United States (86.9%). The average age of affected individuals was 36.3 years old (standard deviation [SD] = 17.0), with the majority being female (66.1%). Patient and respondent characteristics can be seen in Table 2.

3.1. Aggregate preferences

Results of the BWS experiment using best-minus-worst scoring and conditional logit analysis are displayed in Table 3. A positive best-minus-worst score indicates that the attribute level was chosen as best more often than it was chosen as worst, and a negative score indicates the opposite. The coefficients in the conditional logit analysis highly correlated with the best-minus-worst maximum difference score with a correlation coefficient of 0.992 ($p < 0.0001$, see Fig. 2). This indicates that the two models are concordant. The attributes that were chosen the most often as best features of a device were short forecasting range (2.06, standard error [SE] = 0.09), followed by mid forecasting range (1.79, SE = 0.08) and notification of high chance of seizure (1.87, SE = 0.08). The features selected as least favorable were device being implanted (−2.32, SE = 0.09), followed by the highest level of cost (−1.99, SE = 0.07) and 30% inaccuracy (−1.69, SE = 0.08).

Table 3
Aggregate preference scores and results.

Attribute	Best minus worst				Conditional logit	
	Best	Worst	B − W	Relative BW score	Coefficient	Robust SE
Chance						
Low	194	170	24	0.8	0.06	0.07
Moderate	320	43	277	9.4	0.62	0.07
High	1091	17	1074	36.3	1.87	0.08
Range						
Short	1482	53	1429	48.3	2.06	0.09
Mid	1330	38	1292	43.7	1.79	0.08
Long	961	116	845	28.6	1.24	0.09
Inaccuracy						
10%	341	347	−6	−0.2	0.14	0.11
20%	63	601	−538	−18.2	−1.00	0.07
30%	38	1041	−1003	−33.9	−1.69	0.08
Time						
Low	472	43	429	14.5	1.00	0.07
Moderate	113	102	11	0.4	0.00	0.00
High	48	478	−430	−14.5	−0.89	0.06
Worn						
Externally	750	81	669	22.6	1.37	0.08
Subcutaneous	205	538	−333	−11.3	−0.68	0.11
Implanted	91	1483	−1392	−47.1	−2.32	0.09
Cost						
$50	232	346	−114	−3.9	−0.05	0.09
$150	15	1018	−1003	−33.9	−1.53	0.07
$200	19	1309	−1290	−43.6	−1.99	0.07

Relative BW Score is calculated as (B − W)/N where N is the total number of times that attribute level was seen across all participants.

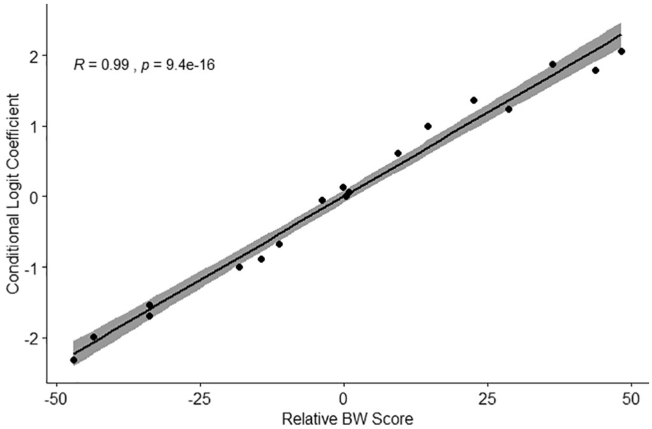


Fig. 2. Comparison of conditional logit and BWS methods.

3.2. Stratified preferences: comparing patients and caregivers

As the conditional logit model allows for more complex analysis, we used this approach to stratify results by relationship to epilepsy to explore if patient and caregiver preferences differ. Caregivers and family members were grouped together because of the small number of family members (n = 15). We hypothesized that people living with epilepsy were impacted by the fear of having seizures more than caregivers, which could change their benefit–harm assessment for a device. In our survey, 44.2% of patients selected fear of having seizures as impacting them more than actually having seizures, while only 33.6% of caregivers and family members were more impacted by fear of the patient having a seizure. This difference was statistically significant ($p = 0.034$).

Characteristics of the respondent stratified by relationship to epilepsy can be seen in Table 2. There were two striking differences between populations. Caregivers were responding for people impacted by epilepsy at younger ages (average age 20.3 years) compared to those who self-reported as persons with epilepsy (average age 43 years) ($p < 0.001$). The second difference was in the gender of the person impacted by epilepsy as reported by patients (74.8% female) compared to caregivers (45.5% female) ($p < 0.0001$). This could potentially account for the differences in responding to whether the fear of having seizures or actually having the seizures would impact them more.

When modeling preference results stratified by relationship to epilepsy (patient or caregiver) the direction remained similar to that of the aggregated conditional logit model. However, the patient model was statistically different from the caregiver model ($p < 0.001$, Wald test) [26]. Differences in the two models were not attributable to scale ($p < 0.001$), as determined by the Swait–Louviere test [27]. Patient and caregiver preferences were statistically different on the following device attributes: notification of high chance of seizure ($p = 0.0062$), all three levels of forecasting range: short ($p = 0.0008$), moderate ($p = 0.0019$), long ($p = 0.0308$), and both 20% inaccuracy ($p = 0.0047$) and 30% inaccuracy ($p = 0.0034$). Preferences did not differ across notification of low or moderate chance of seizure, 10% inaccuracy, time burden, how the device is worn, or cost. Results of the conditional logit model stratification are presented in Fig. 3.

As respondents also had to select whether they would use the device if it were available, we could also examine the difference in acceptance rates of the devices between PWE and caregivers (Fig. 4 and Table 4). Caregivers had higher acceptance of devices for every treatment profile, with statistically different rates for 10 of the 18 treatment profiles. The largest difference in acceptance occurred for the hypothetical device with the following attributes: moderate seizure chance notification, long forecasting range, 20% inaccuracy, low time burden, worn externally, and costing $200 a month, with about 67% of caregivers accepting the device and only about 48% of PWE accepting the device. The device with highest acceptance rate between both PWE and caregivers had the following profile: low chance of seizures notification, short forecasting range, 10% inaccuracy, low time burden, worn externally, and costs $50 a month, with acceptance rates of 83% for PWE and about 95% for caregivers.

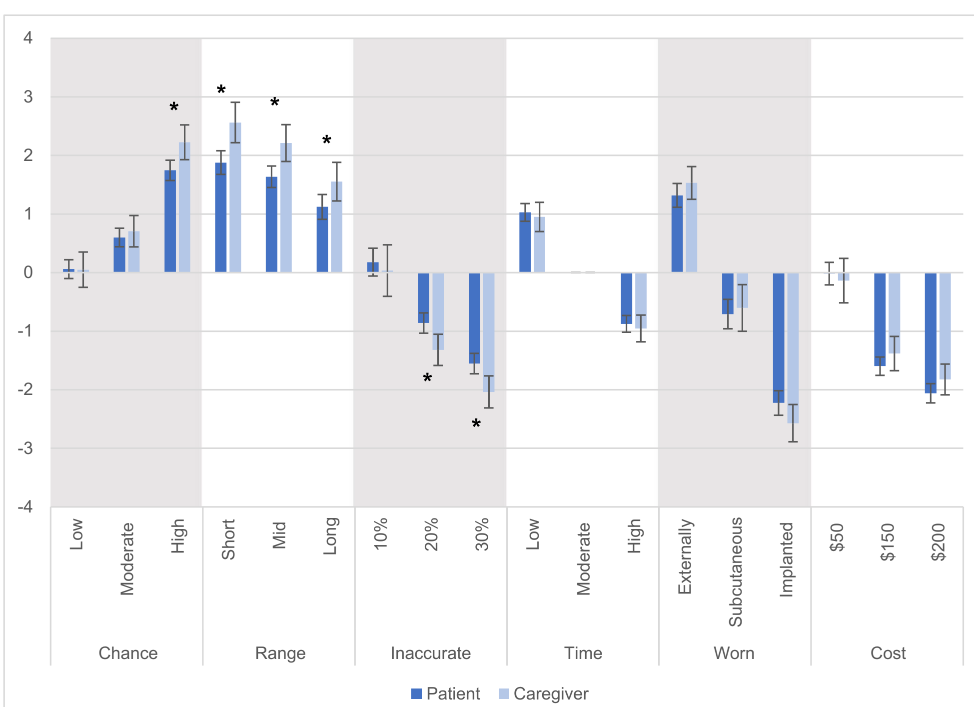


Fig. 3. Patient and caregiver preference estimates from the conditional logit model. * indicates significant at the 0.05 level.

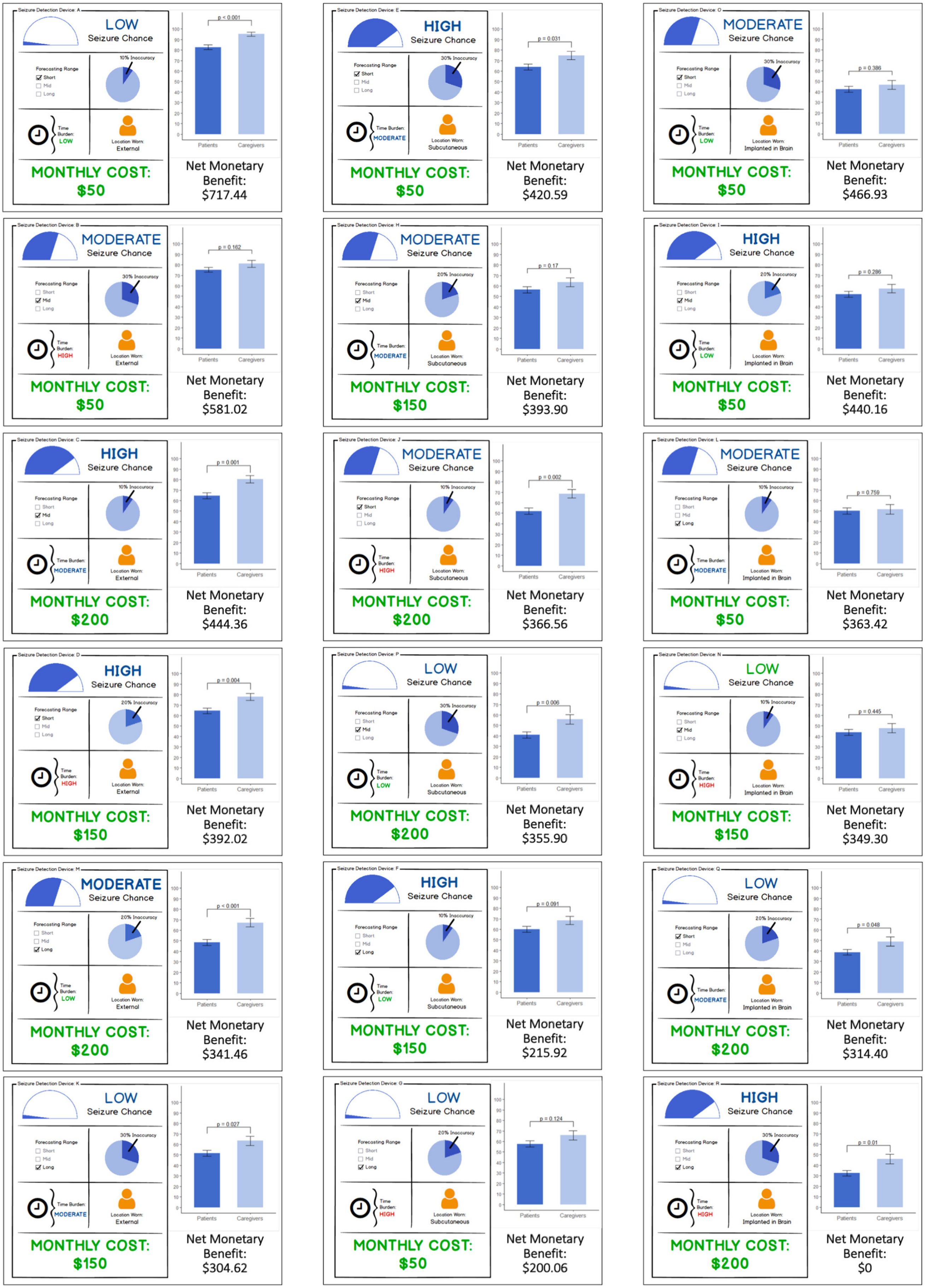


Fig. 4. Device acceptance among patients and caregivers and net monetary benefit (NMB).

Table 4
Device acceptance and net monetary benefit.

Task	Seizure chance	Forecasting range	Percent inaccuracy	Time burden	Location worn	Monthly cost ($)	% that would use the device		NMB
							Patients	Caregivers	
A	Low	Short	10%	Low	External	$50	82.6%	95.0%	$ 717.44
B	Moderate	Mid	30%	High	External	$50	75.1%	81.0%	$ 581.02
C	High	Mid	10%	Moderate	External	$200	64.6%	80.5%	$ 444.36
D	High	Short	20%	High	External	$150	64.4%	77.8%	$ 392.02
E	High	Short	30%	Moderate	Subcutaneous	$50	63.9%	74.8%	$ 420.59
F	High	Long	10%	Low	Subcutaneous	$150	60.1%	68.3%	$ 215.92
G	Low	Long	20%	High	Subcutaneous	$50	57.6%	65.8%	$ 200.06
H	Moderate	Mid	20%	Moderate	Subcutaneous	$150	56.3%	63.4%	$ 393.90
I	High	Mid	20%	Low	Implanted	$50	52.0%	57.3%	$ 440.16
J	Moderate	Short	10%	High	Subcutaneous	$200	52.0%	68.5%	$ 366.56
K	Low	Long	30%	Moderate	External	$150	51.6%	63.4%	$ 304.62
L	Moderate	Long	10%	Moderate	Implanted	$50	50.0%	51.7%	$ 363.42
M	Moderate	Long	20%	Low	External	$200	48.3%	67.1%	$ 341.46
N	Low	Mid	10%	High	Implanted	$150	43.8%	47.8%	$ 349.30
O	Moderate	Short	30%	Low	Implanted	$150	42.3%	46.7%	$ 466.93
P	Low	Mid	30%	Low	Subcutaneous	$200	40.9%	55.8%	$ 355.90
Q	Low	Short	20%	Moderate	Implanted	$200	38.7%	48.9%	$ 314.40
R	High	Long	30%	High	Implanted	$200	32.4%	45.8%	–

3.3. Net monetary benefit

As there was a cost associated with each hypothetical device profile, we could calculate the NMB for each hypothetical device with respect to the least accepted device (device *R*, Table 4). The device that respondents are most willing to pay for was the device with the highest acceptance rate described above (low seizure chance notification, short forecasting range, 10% inaccuracy, low time burden, worn externally, and costs $50 a month). The NMB for this device, relative to the reference device (least accepted device), is $717.44 per month. The next device that participants were most willing to pay for constituted a device of moderate seizure chance notification, mid forecasting range, 30% inaccuracy, high time burden, worn externally, and cost $50 a month. Respondents were willing to pay an additional $581.02 more a month for this device than the reference device. The NMB for each of the devices relative to the reference device can be seen along with device acceptance in Fig. 4 and Table 4.

4. Discussion

Recent advances in neural engineering, minimally invasive biosignaling devices, and deep machine learning algorithms suggest that seizure forecasting technology may become an available tool in the foreseeable future. A clinical trial of an implantable seizure advisory system as well as subsequent machine learning competitions demonstrated that seizure forecasting may be possible [7,8,11,29]. Intensive efforts continue toward making seizure forecasting a clinically accessible tool by developing devices capable of computing forecasting algorithms, improving the accuracy of forecasting algorithms, and exploring less invasive monitoring devices for seizure forecasting such as subscalp EEG, wearables and diaries [9,10,30,31]. In 2017, the Epilepsy Foundation selected seizure forecasting as a strategic priority for development [12]. As interest in this area continues to grow, it is imperative that the patient voice is incorporated in the development of seizure forecasting devices and design solutions, as these users will be the most impacted by these breakthroughs.

Our results echo previous studies that have found great interest in a seizure forecasting device to address the unmet need of seizure prediction [12]. We demonstrate what attributes are most important to respondents in a seizure forecasting device and which show favorable acceptance rates for the 18 hypothetical devices presented. The information presented here by the attributes of our devices and corresponding acceptance rates can inform prototype development, so that device manufacturers develop seizure forecasting devices that are likely to be accepted by the epilepsy community.

Although high seizure forecasting probability was preferred to low or moderate forecasting probability, the results indicate that seizure forecasting is favorable regardless of whether the device gives a short-, mid-, or long-term range warning in advance. This is in contrast to a previous survey suggested that individuals only cared about short-term range predictions [32]. The earlier survey had fewer respondents (n = 89) and administered the question about best length of warning time as a multiple-choice question with 10 possible answers. They found that participants thought the best length of warning time is 3 to 5 min and reasoned from a follow-up question that longer lengths of time would be more stressful. In our conjoint-analysis approach, we found that longer forecasting times still provide a benefit to users.

Perhaps not surprisingly, cost was one of the worst aspects of the device. This is an important consideration for future reimbursement issues if we want these devices to be used at scale by the community. This is in alignment with previous research on user-centered design considerations of seizure detection devices, which suggested that individuals would only be willing to pay between $200 to $300 for a device plus an additional $50 per month [6]. The NMB analysis in our study demonstrates that user's willingness to pay depends on the device attributes. Therefore, the cost for a device that the community would be willing to pay for could be higher than previously reported. This may be due to the fact that the previous research was focused on a seizure detection device while we are considering a seizure forecasting device.

Although the community has predominantly focused on assessing when an individual is at high risk of seizure in their forecasting algorithms, the most favorable device in our survey gave notification of low seizure chance. This suggests that there is also value to the individual in knowing when there is a “safe-zone,” a period of low seizure likelihood. This could be critical to design considerations, as algorithms indicate better specificity and sensitivity to detecting when someone is at a low risk of having a seizure rather than a high risk [11].

Differences in PWE and caregivers' responses were explored to assess whether PWE would take on more risk for a seizure forecasting device. We formulated this hypothesis based on the finding that PWE are more impacted by the fear of having seizures (versus the actual seizure event) than caregivers. Consistent with our hypothesis, caregivers find inaccuracies of the device a worse feature overall than those living with epilepsy. This difference could be attributed to respondent type but could also be due to the personality characteristics or demographic differences across the two categories. For example, caregivers were responding for those at younger ages (Table 2). Interestingly, caregivers had a higher acceptance of devices for every hypothetical description. We did not explore reasons for differences in PWE and caregiver preferences or acceptance rates. These differences could be due to reasons such as clinical differences, socioeconomic differences, or other explanations not evaluated here. Specifically, previous work has discussed that caregivers, who are often parents, are more likely to participate in 'yea-saying' as they prefer some option over no option [33]. Caregivers may also have a better capacity to pay than PWE, a phenomenon that has also been discussed previously and applies to the current research [34]. We will explore this further by tailoring to specific disease types in future studies through the use of inclusion and exclusion criteria.

Another way to survey user preferences is through discrete choice experiments (DCE); DCE are more common and admired by some researchers because of their explicit tradeoffs, yet their experimental designs can be prohibitively complicated and potentially cause bias in preference estimation [35]. We utilize a catalog-based main effects orthogonal array that presents an advantage over DCE.

We demonstrate the value of using BWS as a tool to study preferences for seizure forecasting, but our study has some limitations. The survey included theoretical devices, and so our study is susceptible to hypothetical bias, as a respondent's stated values may differ from their real values [36]. The study is also limited in specificity of the attribute levels. There are currently no seizure gauge devices on the market, and thus there is no prior information on what realistic levels of some of the attributes included in the study should be. Appropriate levels were discussed among both the stakeholders committee comprised of Foundation leadership and healthcare professionals as well as with the people living with epilepsy action committee comprised of individuals impacted by epilepsy to elicit plausible, yet hypothetical levels of attributes. This approach demonstrates a blend between technical and patient-centered methods to study hypothetical devices.

The study may also be susceptible to selection bias as the survey was web-based and certain populations may be under-represented because they have limited access to Internet. Recruitment was based on self-selection and participants were not prescreened by epilepsy type. This resulted in a heterogeneous sample, but as a community-based approach was used to design and implement the survey, it was better to be inclusionary with our sampling. Another limitation is that there was no opt-out included as a holistic model. We are unaware of models that can incorporate this in BWS. Hollin et al. demonstrated that there is consistency between BWS profile case and conjoint analysis where an opt-out is allowed [13].

User preferences are important to consider despite these limitations. Consensus continues to build throughout the healthcare system that patient and stakeholder input is necessary from research and development to delivery [37–42]. This study illustrates patient and caregiver preferences around factors related to potential seizure forecasting devices. Patient-centered approaches are increasingly accepted, and we demonstrate the usefulness of this approach in prototype development. The process we used can be a model for facilitating user-centered device progress through an exploration of the priorities and preferences of PWE and families.

5. Conclusion

As seizure forecasting devices move closer to clinical reality, it is crucial to incorporate user-design considerations. This study incorporates conjoint-analysis modeling to assess community preferences of a seizure forecasting device to better inform device developers early in the research and development process. Results suggest that there may be different preferences and willingness to pay between caregivers and those impacted by epilepsy when considering a seizure forecasting device. Future studies will dive deeper into understanding the different user profiles and design considerations to ensure better adoption of such devices in the future.

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