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Youth weigh in: Views on advanced neurotechnology for drug resistant epilepsy

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Abstract

Background: Epilepsy affects over 500,000 children in North America of whom 30% have drug resistant epilepsy (DRE). Advancements with neurotechnologies show promising benefits, but the perceptions of these procedures by youth is unknown.

Methods: We conducted semi-structured interviews with 10 youth in British Columbia, Canada who underwent procedures for DRE involving different forms of neurotechnology (subdural grids, vagus nerve stimulation, responsive neurostimulation). Interviews were analyzed using the constant comparative qualitative method.

Results: Four major thematic categories emerged from the interviews. Treatment values, impact of the disorder, personal context, and impact of neurotechnology.

Conclusions: Besides the predictable goal of seizure reduction, a desire for autonomy and the importance of trust in the medical team emerged as dominant values within the four thematic categories that were explicit to the use of new neurotechnologies for the management of DRE.

Background

Epilepsy is the most common childhood neurologic disorder, affecting over 500,000 children in Canada and the USA.^{1,2} For many youth, the burden of epilepsy extends far beyond physical symptoms, with the disorder also adversely impacting mental health and social wellbeing.

Youth with epilepsy experience significantly higher levels of depression, anhedonia, anxiety and obsessive symptoms relative to controlled peers, and diminished health related quality of

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life.^{3,4} Stigma is an ongoing challenge.⁵ They experience poor social competence, high medical and mental health needs, and significant rates of social problems as adults such as decreased employment and difficulty with social relationships.^{6,7} The burden of epilepsy extends to the families and the healthcare system on whom they rely and with whom they interact.⁸

Thirty percent of patients with epilepsy continue to have seizures while on anti-seizure medication, a condition known as drug resistant epilepsy (DRE).⁹ Epilepsy surgery is an option for some youth with DRE, and in carefully selected patients, up to 70% become seizure-free after surgery.¹⁰ Those who undergo successful surgical intervention experience long term improvement in health related quality of life and psychosocial function.^{11,12} However, the invasiveness of epilepsy surgery has motivated the pursuit of alternative, less invasive solutions to diagnosis and treatment.¹³ These include neurotechnologies such as MRI-guided laser interstitial thermal therapy (LITT), robot assisted stereo-EEG (rSEEG), stereotactic radiosurgery (SRS), vagal nerve stimulation (VNS), and responsive neurostimulation (RNS). Although these neurotechnologies allow for focused and less invasive delivery of stimuli, there is still a relative lack of knowledge, data, and standardized guidelines regarding their use.^{14,15} Furthermore, little is known about the perceptions of and receptiveness to these neurotechnologies by youth and families with DRE.

The importance of including youth in medical decision-making has been stressed by international legislation and policy, including the United Nations Convention on the Rights of the Child (UNCRC).¹⁶ A comprehensive understanding of their experiences and perspectives is critical in ensuring goal-concordant decision-making. Furthermore, research has shown that youth want to be included in decision-making regarding their treatment, and that they value the opportunity to voice their preferences and choose how treatments are administered.^{17,18,19} The views held by youth regarding their treatment sometimes differ from that of their caregivers and physicians, and further highlights the importance in understanding their values in treatment.²⁰ Yet, there continue to be barriers to effective pediatric shared decision-making, such as power imbalances and a lack of adequate information.²¹ We sought to close this gap for neurotechnologies that are under development or being tested for DRE.

Methods

English speaking participants between 7–25 years who had undergone surgical procedures for DRE during childhood (defined as < 18 years of age) were recruited using purposive sampling methods. DRE was defined as failure to respond to a minimum of two anti-seizure medications, in accordance with the consensus definition established by the International League Against Epilepsy Task Force.²² This patient population tried many medications, some also on the ketogenic diet. Participants with other significant medical comorbidities were not included in the study.

Approval for the study was granted by the University of British Columbia Research Ethics Board (H18–02783-A009). Consent or assent and parent consent were obtained, and all interviews were audio recorded and conducted either in person or over a video calling

platform. Interviews were conducted using a structured guide with open-ended questions that were updated iteratively. The interview guide was divided into three sections: (1) background and impact of epilepsy on participant's life; (2) participant knowledge and attitude about neurotechnology; (3) life post-treatment. The interview culminated in the opportunity for participants to share additional thoughts they had about their experiences with DRE. Sample questions are shown in Table 1. Interviews were conducted by a medical student (author FU) who had no relationship with or role in treatment of the participants.

Interviews were transcribed, made software ready (NVivo QSR 12), and analyzed using conventional thematic qualitative analysis.²³ A coding guide was created based on a literature review and piloting, and revised after several rounds of independent coding by two trained researchers. Discrepancies in coding were discussed to achieve consensus. The guide was then applied to generate dominant themes and their constituent subthemes. Dominant themes were classified as themes that were present in 90% or greater of the interviews.

Results

Ten patients (11–23 years; 6 female, 4 male) participated in the interviews from various locations across British Columbia, Canada (Figure 1). Interviews began in May 2019 and ended after thematic saturation was achieved and defined, for the purposes of this study, as 20% over the number of interviews at which no new themes emerged during the interview process. Five participants underwent intervention with VNS, 4 with surgical resection following subdural grid electrode placements and 1 with RNS. Through the coding process, we identified four major thematic categories of importance to youth based on frequency in the discourse (Figure 2a–d): (1) treatment values, (2) impact of disorder, (3) personal context (4) impact of neurotechnology.

A reduction in seizure frequency was the dominant theme in the treatment value category (30% of major themes), with every participant (10/10) expressing its importance. Trust in the medical team also emerged as a dominant treatment value, present with significance in 9/10 interviews. When emphasizing the importance of trust, one participant responded by saying:

“They’re going to be touching your brain. That’s a sacred part of your body. They could screw your life up. They have all the power there”

(Participant #03, F, grids).

Independence emerged as a secondary theme in this category, for 7/10 participants.

Under impact of disorder (29% of major themes), extracurricular activities emerged as a dominant theme expressed by 9/10 participants. They described how their extracurricular activities in and outside of school were curtailed because of their epilepsy. These activities ranged from sports amongst younger patients, to travelling and driving for older patients. One participant explained:

“I went running one time and I thought, okay let’s try and keep yourself healthy... So I went running and then I had a seizure and I never went running again”.

(Participant #03, F, grids).

Secondary themes in this category related to the impact of epilepsy on patient's academic/work life (7/10) and psychological and mood state (6/10).

The dominant themes for personal context (25%) were emotions/feelings (10/10) and peer and partner support (9/10). Participants expressed a range of emotions with regards to their treatment. Nearly all participants (9/10) were happy overall with the treatment intervention they received. One participant had an equivocal response, claiming to be happy with the fact that the intervention had reduced seizures, but not happy that it had not completely provided a cure. One participant reported that the implanted device resulted in an unnatural feeling "... *like a cyborg*".

Change in seizure frequency dominated the impact of the neurotechnology category (16%), with 10/10 participants reporting a reduction. The degree of the reduction varied amongst participants. One participant, for example, experienced a reduction from 50 seizures per day to just 1–2 at night as a result of placement of subdural grids followed by resection of the epileptic focus. Another reported only a very small benefit while becoming accustomed to using the VNS device during a seizure. Secondary themes in this category pertained to social changes (7/10) and psychological changes (7/10) associated with the intervention.

Participants who experienced positive social impacts as a result of an intervention with neurotechnology were able to spend more time with their families, friends and peers and partake in more activities outside of their own homes. Positive psychological benefit was attributed to having less anxiety about seizure onset due to their intervention.

Discussion

Neurotechnologies such as VNS, RNS and subdural grids followed by surgical resection have the potential to mitigate DRE. As expected, reduction in seizure frequency was the primary disease-specific interventional goal. Trust in the medical team emerged as a dominant treatment value, and speaks to the importance not only of the effectiveness of the intervention itself, but the importance of the patient-physician relationship. Independence also appeared as an important goal, a strong value residing alongside the direct physiological benefits that treatment can provide. Indirect psychosocial benefits were also observed in the study, consistent with other recent reports.²⁴ Although we predicted from previous work utilizing focus groups with families who care for youth with DRE that cosmesis would be a major contributing factor to decision-making, this theme did not emerge from the interviews with youth themselves as an important attribute *per se*. Rather, when probed, youth discussed cosmesis mostly in a negative context as an unimportant factor.²⁵

Important differences between the specific neurotechnologies had to do with their physical presence, notably for example, that VNS that prevents youth from engaging in some extracurricular activities that could harm the device. However, the number of participants in each group was limited, a function of the small, self-identifying sample to which we had access. This limitation also applies to the question of potential differences based on age and gender which were not detectable in this study.

The present findings are similar to other studies that describe the impact of extracurricular restrictions, social isolation and significant emotional distress caused by DRE.²⁶ While participants in the present study spoke about workarounds to extracurricular and school related difficulties, they also expressed frustration at the limitations imposed on them by others with whom they interact daily. Some articulated that they faced unnecessary restrictions that seemed to be driven out of fear for their best interest. On the other hand, while previous literature indicates resistance to epilepsy surgery, the majority of participants noted their comfort with and confidence in the decision to pursue surgery.²⁷ They admitted to being nervous, on reflection, but many recalled being optimistic about their surgery and intervention. One participant described a certain sense of feeling unnatural, or cyborg-like with the neurotechnological implant; an ethical concern regarding sense of self and authenticity that has also been noted elsewhere.²⁸

The widespread positive impact that the neurotechnologies had on patient seizure frequency is a testament to the effectiveness of interventions when used in appropriately selected patients. Bearing in mind the limited sample size, the single center nature of the study, and possibility of a positive social bias toward the interviewer, this study nonetheless adds to that optimism. It provides contextually rich narrative information about social and psychological values and benefits that epileptologists may consider when evaluating the suitability of a given intervention for a patient. Future opportunities for more generalizable data across obtained both cross-sectionally and longitudinally will enrich these findings.

In conclusion, the findings suggest that youth with DRE tend to be receptive to intervention with neurotechnological devices. Their values highlight direct clinical benefits alongside an expanded desire for independence and the importance of clinical trust.

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	1	2	3	4	5	6	7	8	9	10	
	VNS #1	VNS #2	VNS #3	VNS #4	VNS #5	Grids #1	Grids #2	Grids #3	Grids #4	RNS #5	Total
Treatment Values											
Independence	1	3	0	2	0	2	2	3	0	8	21
Trust in Medical Team	2	2	1	1	3	2	3	1	1	4	20
Seizure Frequency	5	2	2	1	1	2	2	1	1	1	18
Impact of disorder											
Extra-curricular Activities	3	5	4	2	4	4	5	4	3	0	34
Academic/Work	15	2	1	1	1	4	3	2	4	0	33
Psychological/Mood	4	0	1	0	6	0	8	3	0	6	28
Personal Context											
Emotional Disposition Towards Intervention	11	9	5	4	4	5	2	5	2	5	52
Partners and Peers	4	1	2	1	1	2	3	2	0	1	17
Impact of neurotechnology											
Change in Seizure Frequency	2	2	3	2	2	7	1	3	1	2	25
Change in Social Life	0	1	0	0	1	4	2	2	2	3	15
Psychological/Mood Change	3	3	3	1	2	0	1	1	0	0	14

Figure 1.
Heat map of themes important to youth across interviews.

Figure 2a-d.
Major categories of themes of importance to youth (occurrences [#] of respective themes).

Table 1:

Sample interview questions

How has epilepsy impacted your life growing up?
Tell me about what is most important to you in terms of what you want your treatment to achieve?
Do you know anything about neurotechnologies, such as devices or electrodes that are placed in your brain, or devices that stimulate parts of your brain?
What did you like about the technology that was used for you?
How has your treatment impacted your life since?