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Amaya Singh
Epilespy Research Program of the Ontario Brain Institute

Rebecca Woelfle
Epilespy Research Program of the Ontario Brain Institute

Rachel Chepesiuk
Ontario Brain Institute

Carla Southward
Ontario Brain Institute

Jordan Antflick
Ontario Brain Institute

See next page for additional authors

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Authors

Amaya Singh, Rebecca Woelfle, Rachel Chepesiuk, Carla Southward, Jordan Antflick, Katherine Cowan, Kathryn Hum, Marcus Ng, Jorge G. Burneo, and Ana Suller Marti

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Canadian epilepsy priority-setting partnership: Toward a new national research agenda

Amaya Singh [a,b,*], Rebecca Woelfle [a,b], Rachel Chepesiuk [c], Carla Southward [c], Jordan Antflick [c], Katherine Cowan [d], Kathryn Hum [a,b], Marcus Ng [e], Jorge G. Burneo [f,g], Ana Suller Marti [f,h,i], on behalf of the Steering Group

[a] EpLink – The Epilepsy Research Program of the Ontario Brain Institute, Toronto, Ontario, Canada
[b] Department of Pharmacology & Toxicology, University of Toronto, Toronto, Ontario, Canada
[c] Ontario Brain Institute, Toronto, Ontario, Canada
[d] The James Lind Alliance, The Wessex Institute, University of Southampton, Southampton, United Kingdom
[e] Section of Neurology, Department of Internal Medicine, University of Manitoba, Winnipeg, Manitoba, Canada
[f] Epilepsy Program, Department of Clinical Neurological Sciences, Schulich School of Medicine and Dentistry, Western University, London, Ontario, Canada
[g] Neuroepidemiology Unit, Schulich School of Medicine and Dentistry, Western University, London, Ontario, Canada
[h] Paediatrics Department, Schulich School of Medicine and Dentistry, Western University, London, Ontario, Canada
[i] Neurosciences Program, Western University, London, Ontario, Canada

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ABSTRACT

Backgr
ound: Health research agendas are ofte
n set by researchers or by industry and may not reflect the needs and priorities of end users. This priority-setting partnership (PSP) for epilepsy was undertaken to identify the most pressing unanswered questions about epilepsy and seizures from the perspective of people with epilepsy (PWE) and their care providers.
Methods: Using the methodology developed by the James Lind Alliance (JLA), evidence uncertainties were gathered via online surveys from stakeholders across Canada. Submissions were formed into summary questions and checked against existing evidence to determine if they were true uncertainties. Verified uncertainties were then ranked by patients, caregivers, and healthcare providers and a final workshop was held to reach a consensus on the top 10 priorities.
Results: The final top 10 list reflects the priority areas of focus for research as identified by the Canadian epilepsy community, including genetic markers for diagnosis and treatment, concerns about living with the long-term effects of epilepsy, and addressing knowledge gaps in etiology and treatment approaches.
Conclusion: This project represents the first systematic evidence of patient- and clinician-centered research priorities for epilepsy. The results of this priority-setting exercise provide an opportunity for researchers and funding agencies to align their agendas with the values and needs of the epilepsy community in order to improve clinical outcomes and quality of life (QOL) for PWE.
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1. Introduction

Epilepsy is one of the most common neurological conditions worldwide. It is a complex, heterogenous disease that affects around 300,000 Canadians, and 20,000 are newly diagnosed each year [1]. Although antiseizure medication can successfully control seizures in approximately 70% of patients, treatment selection remains challenging and epilepsy-related comorbidities represent a substantial burden [2,3]. People with epilepsy (PWE) report high usage of healthcare resources and significantly lower quality of life (QOL) than individuals living with other chronic conditions [4,5]. Health inequalities among PWE and disparities in access to care represent significant barriers to effective treatment [5,6]. Epilepsy also carries an increased risk of premature death [7], and the

Abbreviations: JLA, James Lind Alliance; PSP, priority-setting partnership; REPRISE, reporting guideline for health research priority setting with stakeholders.

* Corresponding author at: Department of Pharmacology and Toxicology, University of Toronto Medical Sciences Building, 1 King's College Circle, Room 4207, Toronto, Ontario M5S 1A8, Canada.
E-mail address: amaya.singh@utoronto.ca (A. Singh).

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prevalence of social stigma and discrimination toward PWE continues to be a source of fear and distress [8–10]. Thus, the current scope of epilepsy research is necessarily broad, and knowledge gaps exist across the continuum of care. With limited funding [11] and resources, it is critical to focus on addressing evidence gaps that will have the largest impact on health-related QOL for PWE. While shared decision-making has become an increasingly important aspect of clinical care, the adoption of meaningful patient engagement in research has yet to be normalized [12]. Health research agendas are often biased by vested researcher or commercial interests, leading to a mismatch between the priorities of researchers and those whom research is intending to serve [13,14]. As such, there is growing acknowledgment of the need to engage patients as partners in research and to direct funding to those areas deemed most essential by end users. The James Lind Alliance (JLA), a UK-based non-profit initiative supported by the National Institute for Health Research, has developed a rigorous methodology for increasing public engagement in research by consulting patients, caregivers, and healthcare providers regarding their most pressing unanswered questions or treatment uncertainties [15]. Such priority-setting partnerships (PSPs) provide an opportunity to increase the clinical relevance of research and improve patient outcomes. Importantly, the JLA methodology gives an equal voice to patients, caregivers, and clinicians to help ensure that their needs are represented in health research agendas. In collaboration with the JLA, the Ontario Brain Institute (OBI) and its epilepsy research program (EpLink) undertook a Canada-wide PSP to identify and prioritize unanswered questions relating to epilepsy and seizures in pediatric and adult populations. To maintain transparency and accountability, we have reported on the domains as set out in the reporting guideline for health research priority setting with stakeholders (REPRISE) [16].

2. Material and methods

Ethics approval was obtained from the Community Research Ethics Office at the Centre for Community Based research in Waterloo, Ontario. This project was completed over 23 months (May 2019 to April 2021) and was overseen by an independent JLA advisor (KC). An overview of the JLA process is provided in Fig. 1.

2.1. Establishment of steering group

Based on their professional experience, the project team identified and reached out to established leaders in the Canadian epilepsy community, who in turn reached out to their contacts in the patient and professional communities. In accordance with JLA principles, steering group (SG) members brought with them knowledge of epilepsy, an understanding of the epilepsy community, and access to networks of patients, caregivers, and clinicians. The initial SG comprised five clinicians and seven community members drawn from across Canada (see Fig. 2 for details). Community members were either PWE, caregivers, or members of non-profit agencies. Steering group members were asked to declare any potential conflicts of interest at the outset. The SG met monthly and was responsible for overseeing and guiding the priority-setting process. Members played an active role in setting the scope of the project, determining how questions were categorized, and ensuring that the perspectives brought forward from the community were accurately represented. The project team comprised the SG, the JLA advisor, and knowledge translation specialists from the OBI and EpLink. In addition, an Information Specialist (IS) team led the dataset management and evidence-checking process with the assistance of the SG.

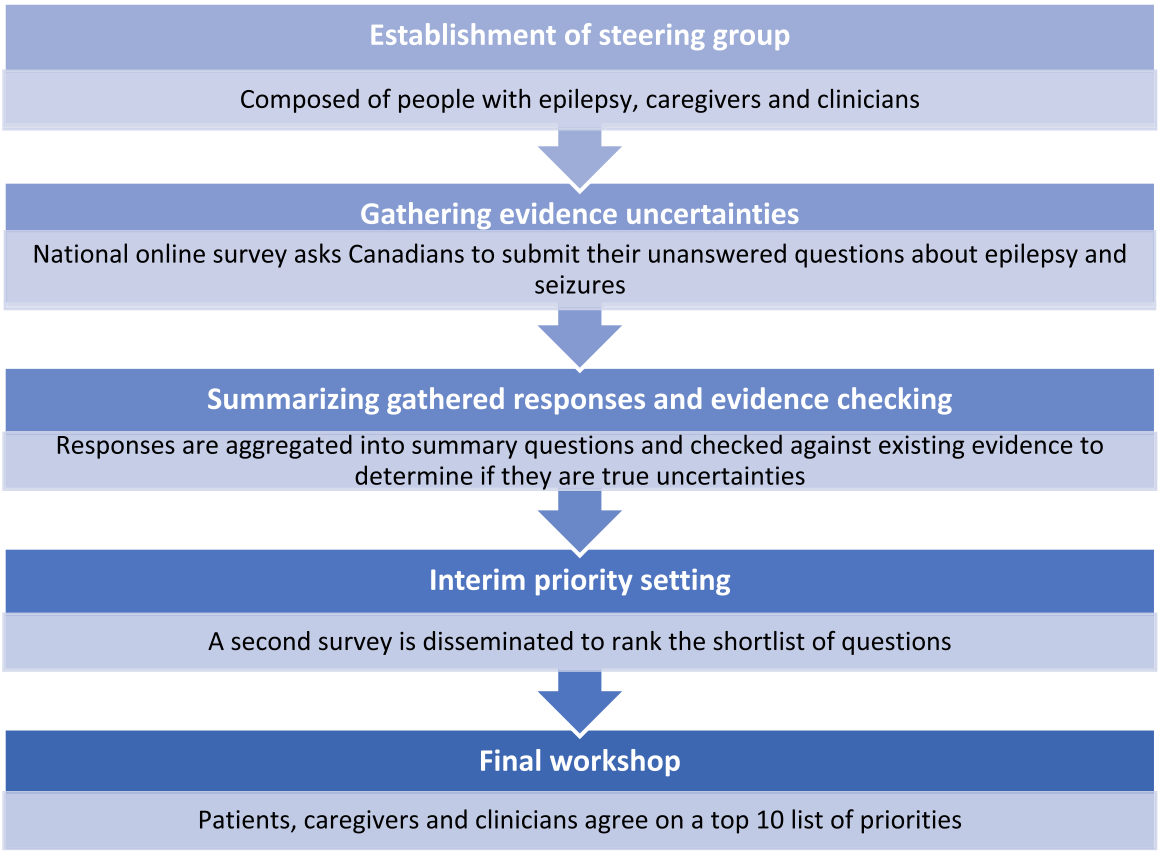

Fig. 1. Overview of JLA process.

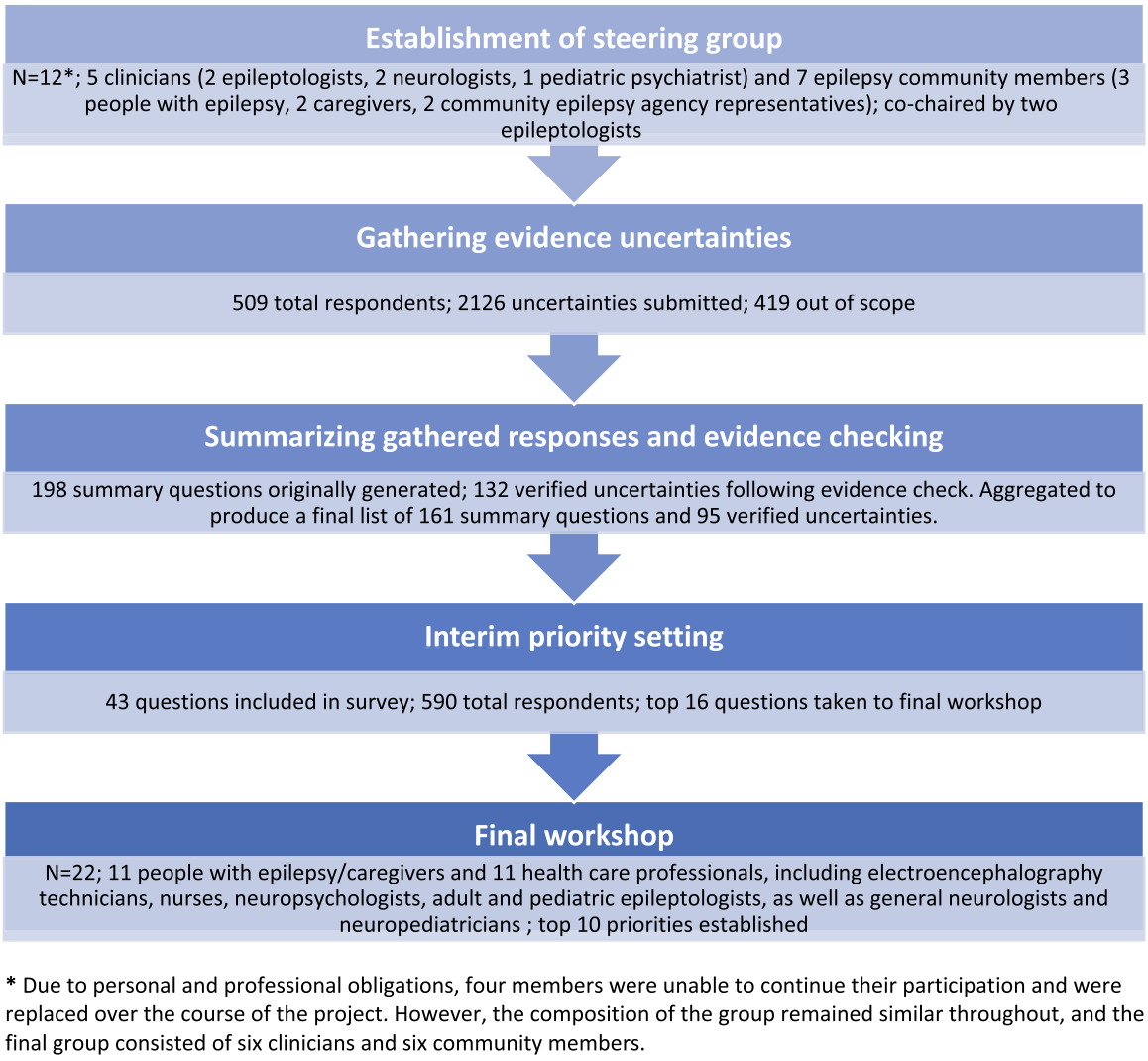

Fig. 2. Results of JLA process. *Due to personal and professional obligations, four members were unable to continue their participation and were replaced over the course of the project. However, the composition of the group remained similar throughout, and the final group consisted of six clinicians and six community members.

2.2. Scope of project

This PSP focused on questions relating to the cause, diagnosis, and treatment of epilepsy and seizures, as well as management of day-to-day life and comorbidities. Questions related to healthcare policies, access to services, or treatment of epilepsy outside of Canada were considered out of scope. Given that epilepsy is often used interchangeably with the term "seizure disorder", it was determined that the survey language should refer to both "epilepsy" and "seizures".

2.3. Gathering evidence uncertainties

An anonymous online survey, offered in both French and English, was created using the Research Electronic Data Capture (REDCap) system hosted by the Centre for Advanced Computing (Kingston, Ontario) and posted on the Ontario Brain Institute's public website. In accordance with the JLA aims, eligible respondents included PWE and their caregivers, friends or family members, and healthcare providers. The initial survey consisted of four open-ended questions where respondents were asked to submit their unanswered questions about epilepsy and seizures (Table 1). There was no limit to the length or number of questions a respondent could submit for each of the topic areas. Optional demographic questions were included to help understand the respondent profile, including respondent type, age, gender, ethnic background, and province or territory. Eighty-eight partner organizations, including advocacy groups, professional organizations, community agencies, and health research institutions, were identified by SG members. They were asked to promote the survey through social media, newsletters, mailing lists, websites, blogs, membership communications, or in clinics. Respondents were able to provide their email address through a separate survey process if they wished to receive future information about the study. Paper copies were also mailed to epilepsy monitoring units and non-profit community epilepsy agencies if requested by staff members.

2.4. Formation of summary questions and evidence checking

Each submitted question was assigned a unique identifier and categorized first as in-scope or out-of-scope, and then sub-categorized by topic. Many entries contained multiple questions which were extracted individually where possible. In instances where comments, statements, or personal anecdotes were included, these remained in their original format and were given an identifier if it was clear what the response intended to ask;

Table 1
Initial survey questions.

	Questions
1	What question(s) do you still have about the cause or diagnosis of epilepsy and seizures?
2	What question(s) do you still have about the treatment of epilepsy and seizures?
3	*What question(s) do you still have about managing day-to-day life with epilepsy and seizures? (Examples include life at work or school, memory problems, fear, stigma, or suicidal thoughts)
4	*What question(s) do you still have about managing the co-existing conditions related to epilepsy and seizures? (Examples include depression, anxiety, behavioral issues, or developmental disorders)

* SG members felt that examples of co-existing conditions and day-to-day challenges would be helpful if provided despite the potential risk of influencing responses.

otherwise, these responses were categorized as "question too broad or unclear". Similar questions were aggregated under larger summary questions within each topic. Examples of the categorization process are provided in Table 2. The IS team was responsible for creating summary questions from the initial submissions and cross-referencing these questions with existing research to broadly determine whether they were true uncertainties. Steering group members guided the summary question development by assisting with the interpretation of submissions, reviewing initial summary questions, and determining the appropriateness of combining questions.

In order to inform the evidence-checking strategy, questions were organized by the IS team into three categories: quantitative, qualitative, and mixed methods (qualitative and quantitative). Questions that could be answered primarily through objective measurement and numerical analysis were deemed quantitative, while questions focusing on the knowledge and perceptions of those being studied were categorized as qualitative. Questions that could be answered using both approaches were considered mixed methods.

The JLA defines a question as an uncertainty when no up-to-date, reliable systematic reviews of research evidence addressing the uncertainty exist, or when such reviews are inconclusive. Accordingly, for quantitative questions, sources for evidence checking included the following systematic reviews and existing guidelines: (a) The Cochrane Database of Systematic Reviews; (b) Cochrane Epilepsy; (c) Scottish International Guidelines Network (SIGN), and National Institute for Health and Care Excellence (NICE), as no relevant guidance exists from the Royal College of Physicians and Surgeons of Canada as well as (d) The International League Against Epilepsy and (e) the World Health Organization (WHO). For qualitative questions, key components were identified using the PICO (Population, Interest, Context) framework and relevant search terms were established for each question. Literature searches were carried out using five different databases (Cochrane, Medline, EMBASE, CINAL, and SCOPUS) and any additional sources suggested by the SG, such as the WHO. Study types included qualitative studies, case reports or case series with qualitative study components, and review-level studies when possible. No date limitations were included, but searches were limited to English language publications. An uncertainty was deemed to be any question that could not be answered by the described literature review process.

2.5. Prioritization survey

The uncertainties were refined and reduced to produce a shortlist of questions for the interim prioritization survey, with the SG advising on a number of questions that would be manageable to survey respondents who experience seizures. Similar questions from the same theme were combined, and some questions were removed based on a low frequency of original submissions (<7). Participants were recruited in the same manner as the first survey, and partner organizations again promoted the survey among their networks. Community epilepsy agencies were unable to distribute paper copies due to COVID-19-related limitations on their in-person services. In the first section, respondents were asked to select up to ten questions they most wanted researchers to address. In the second section, respondents had the option to provide their demographic information, to help the SG understand the respondent profile and target promotional activity toward groups who were under-represented in survey responses. To determine the final set of questions to move forward to the final workshop, questions were ranked based on the frequency with which they were chosen, with each response given one point. Rankings were calculated separately for PWE/caregiver and healthcare provider respondent groups to ensure equal influence on the shortlisting. Questions were placed into rank order within each group and rankings were then merged to determine the final order. The top 16 questions were then brought forth to the final workshop.

2.6. Final workshop

A two-day, virtual workshop was held to determine the final prioritization. Community members were invited to send expressions of interest to participate in the workshop via an online form that asked potential attendees about their personal and professional experiences with epilepsy and their reasons for wanting to take part in the workshop. Attendees were selected to ensure a diversity of interests, experience, clinical specialties, and geo-

Table 2
Classification of responses.

Category	Examples of original submissions	Summary question or classification
Aggregation of similar questions	• *"Are multiple medications more effective?"* • *"Is medication the best treatment?"* • *"What is the long-term effectiveness of medication-based treatments?"*	Grouped under summary question: "How effective are antiseizure drugs?"
Comments, statements or personal anecdotes where intended question is clear	*"My question it seems like his epilepsy gets treated and his depression is his to manage through lifestyle changes but his depression is a medical condition as well. There is no across the board treatment for the coexisting condition of depression and epilepsy. It is treat the epilepsy and lifestyle changes for the depression which don't always work."*	Grouped under summary question: "What medical, non-medical and community support methods can assist people who experience seizures in managing depression/anxiety?"
Comments, statements or personal anecdotes where intended question is unclear	• *"What is the impact on youth?"* • *"We should develop better non-invasive treatments."*	Categorized as too broad or unclear

graphic locations. Eleven community members with lived experience and eleven health or care professionals from across Canada attended the workshop (Fig. 2). Participants were provided with the list of questions and asked to rank them independently prior to the meeting. During the workshop, all participants shared their views and had the opportunity to hear the perspectives of others. Using a nominal group technique, four trained JLA facilitators led participants in group-level discussions and two rounds of prioritization, after which a top 10 list was reached by consensus.

3. Results

All submitted questions will be made publicly available on the OBI's central database, Brain-CODE (braincode.ca).

In the initial survey, a total of 509 respondents submitted 2126 unique uncertainties (Fig. 2). Following the development of summary questions, 198 questions underwent the evidence check. Of these, 61 were determined to be answered, 27 partially answered, and 110 unanswered. Partially answered questions were rewritten to include only those aspects which were unanswered and five were subsequently merged with similar unanswered questions. Thus, of the 198 summary questions, 132 were classified as unanswered following the evidence check. This list was further refined by merging similar questions, producing a total number of 161 summary questions and 95 verified uncertainties. Following the removal of low-frequency submissions, a shortlist of 43 questions was included in the interim prioritization.

Five hundred and ninety responses were received to the interim ranking survey. Across the initial and interim surveys, the majority of respondents were PWE (49.4% and 42.5%, respectively), female (75.8% and 79.3%), from Ontario (57.2% and 40.3%) and aged 36–49 (40% and 44.6%) (Table 3). Most respondents denoted their ethnic background as Caucasian (78.7% and 81.4%), in accordance with national demographic data suggesting that 77% of Canadians identify as Caucasian [17].

Table 4 displays the final rankings. As the priorities from the two respondent groups (PWE/caregivers and healthcare providers) were considered and ranked separately following the interim survey, the final shortlist of 16 questions was selected to ensure that the top 10 ranked questions from both groups were brought forth to the prioritization workshop.

In the final top 10 list (Table 4), the consensus highest priority question was how genetic markers can be used in diagnosis and treatment. The second highest-ranked question related to long-term impacts and side effects of antiseizure drugs (ASDs), which was also ranked as the top priority for both respondent groups in the interim survey. One question related to the effectiveness of surgical treatment in both pediatric and adult populations. The etiology, diagnosis, and long-term impact of seizures were also ranked in the top ten. Other questions were focused on the causes of memory impairments and behavioral problems, sudden death in epilepsy, and ASD polytherapy.

4. Discussion

This PSP brought together PWE, caregivers, and healthcare providers across Canada to determine priority areas of focus for research on pediatric and adult epilepsy. The top 10 priorities reflect the continued challenges in diagnosing, treating, and managing epilepsy, which includes both seizures and the behavioral, cognitive and psychosocial aspects of the condition. Among the top 16 questions from the interim survey, reducing the risk of SUDEP, managing behavioral issues, and initiating ASD polytherapy were all ranked more highly by healthcare providers, while non-drug lifestyle treatments, mood changes, and the links between hormones, seizures, and pregnancy were ranked more highly by patients and caregivers. Despite these differences, there was meaningful congruence between the two groups on several questions prior to the workshop (Tables 4 and 5).

Table 3
Demographics of respondents from initial and interim surveys.

Category	Initial survey (%)	Interim survey (%)
Total responses	516	590
Respondent type		
Person with epilepsy or seizures	255 (49.4)	251 (42.5)
Caregiver/former caregiver of someone with epilepsy or seizures	74 (14.3)	83 (14.1)
Friend/family member of someone with epilepsy or seizures	113 (21.9)	147 (24.9)
Healthcare provider	46 (8.9)	97 (16.4)
Representative of a community epilepsy agency	8 (1.6)	7 (1.2)
Other	10 (1.9)	<5 (<0.8)
Not answered	10 (1.9)	<5 (<0.8)
Healthcare providers		
Family doctor	6 (1.2)	5 (0.8)
Specialist	19 (3.7)	46 (7.8)
Nurse or nurse practitioner	7 (1.4)	19 (3.2)
Pharmacist	<5 (<1.0)	<5 (<0.8)
Allied health professional (e.g., physician assistant, social worker, psychologist, occupational or physical therapist, midwife)	7 (1.4)	12 (2.0)
Other	<5 (<1.0)	<5 (<0.8)
Not answered	<5 (<1.0)	11 (1.9)
Gender		
Male	111 (21.5)	109 (18.5)
Female	391 (75.8)	468 (79.3)
Prefer not to say/non-binary	<5 (<1.0)	8 (1.4)
Not answered	10 (1.9)	<5 (<0.8)
Age		
Under 18	6 (1.2)	6 (1.0)
18–35	132 (25.6)	154 (26.1)
36–49	201 (40.0)	263 (44.6)
50–69	146 (28.3)	173 (29.3)
70 +	19 (3.7)	14 (2.4)
Prefer not to say	<5 (<1.0)	5 (0.8)
Not answered	8 (1.6)	<5 (<0.8)
Ethnic or cultural background*		
Indigenous/First Nations/Métis/Inuit	15 (2.9)	19 (3.2)
Arab	6 (1.2)	8 (1.4)
Black	12 (2.9)	15 (2.5)
Caucasian	406 (78.7)	480 (81.4)
Chinese	8 (1.6)	9 (1.5)
Filipino	<5 (<1.0)	<5 (<0.8)
Japanese	<5 (<1.0)	<5 (<0.8)
Korean	<5 (<1.0)	<5 (0.8)
Latin American	9 (1.7)	10 (1.7)
Pacific Islander	<5 (<1.0)	<5 (<0.8)
South Asian	9 (1.7)	9 (1.5)
Southeast Asian	6 (1.2)	5 (0.8)
West Asian	<5 (<1.0)	<5 (<0.8)
Other	34 (6.6)	20 (3.4)
Prefer not to say	9 (1.7)	23 (3.9)
Province		
Alberta	32 (6.2)	51 (8.6)
British Columbia	8 (1.6)	68 (11.5)
Manitoba	17 (3.3)	51 (8.7)
New Brunswick	8 (1.6)	10 (1.7)
Newfoundland	6 (1.2)	18 (3.1)
Nova Scotia	20 (3.9)	24 (4.1)
Nunavut	<5 (<1.0)	<5 (<0.8)
Ontario	295 (57.2)	238 (40.3)
Prince Edward Island	<5 (<1.0)	9 (1.5)
Quebec	52 (10.0)	23 (3.9)
Saskatchewan	15 (2.9)	17 (2.9)
Yukon and Northwest Territories	<5 (<1.0)	<5 (<0.8)
Not answered	59 (11.4)	80 (13.6)

* Selection of more than one category was permitted. Demographic categories with between 1 and 5 respondents are indicated as <5 to ensure privacy.

Table 4
Top 10 list of research priorities and interim rankings.

Final ranking	Interim survey ranking		Question
	Patients/caregivers	Healthcare professionals	
1	7	3	Can genetic markers be used to diagnose and treat epilepsy and seizure disorders?
2	1	1	What are the impacts of long-term use of antiseizure drugs, the causes of side effects of these treatments, and how can we prevent the side effects?
3	2	5	What are the long-term impacts of seizures on a person's brain, and overall health and development?
4	13	6	How can the risk of SUDEP (Sudden Unexpected Death in Epilepsy) be reduced in people with epilepsy?
5	16	12	What is the most effective testing protocol for determining causes of seizures and/or a diagnosis of epilepsy or other seizure disorders and to reduce time to diagnosis?
6	9	9	What are the brain changes, on a cellular level, that lead to seizure development?
7	11	11	How effective is surgical treatment for adults and children who experience seizures/epilepsy?
8	3	4	What causes memory problems associated with seizures? Can these memory problems improve over time, and what are the best treatment options for memory loss in people who experience seizures?
9	14	8	Aside from antiseizure drugs and some brain lesions, what causes behavioral changes in people who experience seizures? What is the best way to treat behavioral issues?
10	20	7	What is the efficacy (i.e., the effectiveness of reducing seizures) of adding a second antiseizure medication compared to changing to a different antiseizure medication? How can we determine which combinations of antiseizure drugs are effective?

Table 5
Priority questions 11–16 with interim rankings.

Final ranking	Interim survey ranking		Question
	Patients/caregivers	Healthcare professionals	
11	5	28	How do seizures impact the mood of people who experience them and what are the best methods to manage mood swings?
12	4	2	Are cannabis products (e.g. marijuana, CBD oil) a safe and effective treatment for seizures alone or in combination with standard treatments (e.g. antiseizure drugs)?
13	8	17	Is there a relationship between hormonal changes (e.g. puberty, menopause, pregnancy) and seizure onset and/or frequency, and what are the effects of seizures during pregnancy?
14	12	10	Is it safe to wean a person who has experienced seizures off of antiseizure drugs and if so, when is the right time to wean off antiseizure drugs?
15	10	19	What are the best ways to support people who experience seizures secure and perform in employment (e.g., through accommodations) and what interventions can reduce workplace discrimination?
16	6	13	What non-drug lifestyle treatments (e.g., cardiovascular exercise, yoga) are effective for controlling seizure frequency with or without standard treatments (e.g., antiseizure drugs)?

The consensus priority question was how genetic markers can be used in diagnosis and treatment. Research continues to uncover the genetic traits linked to seizure development, and hundreds of genes are now known to be related to epilepsy [18]. Several inherited and de-novo variants are recognized as pathogenic, particularly in the epileptic encephalopathies, and the therapeutic potential of genetic sequencing is also of great interest. The second highest-ranked question relates to managing the side effects of ASDs. As with other priorities in the top 10 that are focused on broad themes, this is a multi-part question that encompasses both the impact of side effects and their prevention. Interestingly, the only previously published exercise in epilepsy priority setting, a series of focus groups organized in Wales, also found that the top-ranked treatment uncertainty among Welsh people related to side effects of ASDs [19]. Despite being the first-line therapy for epilepsy, ASD selection is an inexact science, as the efficacy of a particular drug is impossible to predict in individual patients. 30% of patients have seizures that fail to respond to ASDs, and despite the introduction of more than a dozen new ASDs in the past decade, this number remains unchanged [20]. Even when seizure control is achieved, the adverse effects of drug treatment can be considerable, and are a leading cause of treatment failure [21]. Indeed, questions focused on alternative therapies, such as medical cannabis and lifestyle treatments were also among the top 16. Most questions in the top ten relate to diagnosis and etiology, seizure control, and QOL, which are necessarily interrelated. Misdiagnosis or delayed diagnosis may have serious implications and can prevent initiation of appropriate and effective treatment [22,23], which directly correlates to QOL [24,25]. Priority #4 addresses SUDEP, or sudden unexpected death in epilepsy, which occurs

when an otherwise healthy person with epilepsy dies suddenly and unexpectedly, and no clear cause of death can be found. Although the search for SUDEP biomarkers is a promising field of research, the biggest known risk factor is uncontrolled generalized tonic-clonic seizures [26], highlighting the importance of seizure control. Finally, questions about memory impairments were understandably ranked highly by both patients and care providers, as they are difficult to manage clinically and are the most frequently reported cognitive complaint among PWE [27,28]. As declines in cognitive function and behavioral changes are linked to both uncontrolled seizures [29,30] and ASD treatment (15,25), links between priorities must again be emphasized. Interestingly, the prevention of acquired epilepsy was not a major theme, possibly because this accounts for a minority of cases, but perhaps also reflecting that the survey captured the concerns of those already living with epilepsy. However, questions about prevention are inherent in understanding the genetic causes of epileptogenesis and the neuronal changes that lead to seizure development.

The categorization of a question as unanswered does not suggest the topic has not been investigated, but rather that the strength of the evidence is not sufficient to reach a consensus. For example, although the effectiveness of surgery has been widely studied, most submissions focused on the reasons for failed surgeries and the prediction of post-surgical outcomes, about which considerably less is known [31]. Similarly, although there is consensus on the optimal timing of ASD withdrawal in children, there is insufficient evidence to determine this in the adult population [32]. Nevertheless, the categorization of 66 of the original 161 summary questions as "answered" suggests that greater efforts are needed to effectively translate existing knowledge to the epilepsy community. As described in the knowledge-to-action framework, identifying a knowledge gap is the first step in a cycle that involves developing contextually relevant tools or interventions for specific audiences, evaluating the impact, and ultimately ending with sustained knowledge use by stakeholders [33]. Current and future knowledge translation tools, including a plain language report on the PSP process and results can be found at braininstitute.ca/epilepsy-psp.

Although the JLA has completed over 100 formal PSPs globally, this is the first to address epilepsy and seizures. While the results reflect the Canadian perspective, given similar health service delivery models, access to care and burden of epilepsy across many high-income countries [34,35], there is likely considerable overlap with other populations. Another epilepsy PSP in the United Kingdom is currently in progress, which may provide an indication of the generalizability across countries. However, as 80% of epilepsy cases occur in low- and middle- income countries [36], where infectious etiologies predominate and the majority lack access to treatment, research priorities may differ substantially in resource-poor settings. Importantly, the societal impact of seizures extends beyond the clinical impact. The authors also note the importance of many other topics raised in the surveys to the health and well-being of PWE, such as addressing societal stigma, employment issues, and coordination of care, and thus researchers are encouraged to look beyond the top 10 priorities for potential areas of focus.

The emergence of the COVID-19 pandemic halfway through the PSP process posed many logistical and personal challenges. Not surprisingly, these impacted on the PSP and the SG, such as the inability to meet in person as planned, impact on clinical workloads, the availability of care and support for family members, and the option to distribute information about the PSP in person. However, it also presented opportunities for innovation and adaptability as new paradigms for virtual engagement emerged, including growing familiarity with videoconferencing facilities. As future PSPs also navigate through the challenges of the pandemic and its aftermath, it is helpful to highlight the flexibility and resilience of such a model of engagement.

4.1. Limitations

Certain limitations should be considered when interpreting these results. Survey respondents were largely limited to English- or French-speaking Canadians with internet access, suggesting a possible nonresponse bias associated with geographic or socioeconomic disparities. In the move to virtual engagement, PSPs should be mindful of who they might not be reaching in their target population(s) and make efforts to close these gaps. Although the composition of the steering group was heterogenous in terms of ethnicity, gender, age, and location in order to represent a range of perspectives, many minority and marginalized groups, as well as those aged under 18 and over 70, were underrepresented in the survey results, and thus the final list of priorities may not fully reflect the views of the diverse Canadian population. Clinical best practices, availability of diagnostic testing, and access to specialists can vary widely across the country, and systematic inequities exist on a regional scale, particularly in the Arctic territories and among Indigenous populations [37].

As with any non-random sample, some degree of self-selection bias is assumed in survey respondents, and this is not meant to be a definitive top 10 list. Rather, the strength of the JLA approach lies in its dialogue-based model of consensus development. The final workshop enables a unique exchange of knowledge and perspective and critically, shared decision-making between those with lived and professional experience. Given the limited engagement of patients and clinicians in setting epilepsy research agendas to date, these results provide valuable insight into consensus priority topics to guide patient-oriented research in epilepsy and seizures.

5. Conclusions

The results of this PSP provide a focus for epilepsy research across the spectrum and lifespan of the disease. The priorities identified by the community serve as a valuable tool to help guide patient-oriented research with the aim of improving clinical care and management of epilepsy. The top 10 list affords an opportunity for researchers and funders to better align their agendas with the needs of PWE and their caregivers.

Declaration of Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Data sharing

Raw and aggregate data will be made freely available on the OBI's data-sharing platform, Brain-CODE (www.braincode.ca).

Ethical publication statement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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