



Research Paper

Results of a National Delphi consensus on the outpatient management of pediatric psychogenic nonepileptic seizures in the United States



Afsaneh Talai^{a,*}, Daniel A Freedman^b, Kristen Trott^c, Maija R Steenari^d, Sigit Plioplys^e, Hillary Kimbley^{f,g}, Jennifer Madan Cohen^h, Priya Tatacharⁱ, Dara V.F. Albert^c, On behalf of the Pediatric Epilepsy Research Consortium

^a Division of Neurology, Department of Pediatrics, University of Texas Southwestern Medical Center at Dallas and Children's Medical Center, 5323 Harry Hines Boulevard, Dallas, TX 75390, USA

^b Department of Neurology, The University of Texas at Austin Dell Medical Center, 1500 Red River Street, Austin, TX 78701, USA

^c Division of Neurology, Department of Pediatrics, Nationwide Children's Hospital/The Ohio State University, 700 Children's Drive, Columbus, OH 43205, USA

^d Division of Neurology, Department of Pediatrics, CHOC Children's/University of California Irvine, 455 South Main Street, Orange, CA 92868, USA

^e Pritzker Department of Psychiatry and Behavioral Health, Ann and Robert H. Lurie Children's Hospital of Chicago, Northwestern University, 225 East Chicago Avenue, Chicago, IL 60611, USA

^f Department of Neurology, Children's Health/Children's Medical Center, 1935 Medical District Drive, Dallas, TX 75235, USA

^g Department of Psychiatry, University of Texas Southwestern Medical Center, 5323 Harry Hines Boulevard, Dallas, TX 75390, USA

^h Division of Neurology, Department of Pediatrics, CT Childrens', University of Connecticut School of Medicine, 263 Farmington Avenue, Farmington, CT 06030, USA

ⁱ Department of Pediatrics, Ann and Robert H. Lurie Children's Hospital, 225 East Chicago Avenue, Chicago, IL 60611, USA

ARTICLE INFO

ABSTRACT

Keywords:

PNES

Functional seizures

Multidisciplinary clinic

Functional neurologic disorder

FND

Background and objectives: The purpose of this study was to develop national consensus based on expert opinion on the optimal outpatient care model of pediatric psychogenic nonepileptic seizures (PNES).

Methods: A core working group (CWG) within the PNES special interest group of the Pediatric Epilepsy Research Consortium was established. The CWG developed a rigorous scoring rubric to select experts in pediatric PNES within the United States of America and a three-round Delphi study was conducted to assess consensus on key components of the management of pediatric PNES in the outpatient setting.

Results: Eighteen experts representing neurology, psychology, psychiatry, social work and nursing participated in the study. Strong consensus was reached that the multidisciplinary clinic (MDC) model is the gold standard for the outpatient management of pediatric PNES. Consensus was obtained that a neurologist, psychologist and social worker are essential members of the MDC and in the setting of unlimited resources, psychiatry and nursing are also recommended. Further consensus was established on the roles of specific personnel, structure of the clinic, billing practices, trainee inclusion, patient inclusion and exclusion, and end of visit management. While consensus was reached that a new term should be developed for this diagnosis, consensus was not reached on the ideal term.

Discussion: Expert consensus was established for the multidisciplinary management of pediatric PNES in the outpatient setting. Specific recommendations were provided that can facilitate the development and implementation of MDCs in other institutions. Further prospective studies are warranted to validate this practice model.

Abbreviations: PNES, psychogenic nonepileptic seizures; CWG, core working group; MDC, multidisciplinary clinic.

* Corresponding author.

E-mail addresses: Afsaneh.talai@utsouthwestern.edu (A. Talai), daniel.freedman@austin.utexas.edu (D.A. Freedman), kristen.trott@nationwidechildrens.org (K. Trott), msteenari@choc.org (M.R. Steenari), splioply@luriechildrens.org (S. Plioplys), hillary.kimbley@childrens.com (H. Kimbley), jcohen01@connecticutchildrens.org (J. Madan Cohen), ptatachar@luriechildrens.org (P. Tatachar).

<https://doi.org/10.1016/j.yebeh.2024.110096>

Received 11 September 2024; Received in revised form 7 October 2024; Accepted 8 October 2024

Available online 20 October 2024

1525-5050/© 2024 Elsevier Inc. All rights are reserved, including those for text and data mining, AI training, and similar technologies.

1. Introduction

Psychogenic nonepileptic seizures (PNES), also referred to as functional seizures (FS), are a type of functional neurologic symptom disorder in which patients experience paroxysmal events of altered motor activity or consciousness, resembling epileptic seizures (ES). While the semiologies may appear similar, the etiologies are quite different. ES are caused by abnormal electrical activity in the brain that can typically be measured by electroencephalography (EEG), while PNES is thought to be related to biopsychosocial factors without an electrical correlate on EEG [1]. Given this overlap, the diagnosis of PNES is generally established and communicated by neurologists, and mental health clinicians provide treatment [2]. This unique situation poses a challenge for both clinicians and patients. Neurologists must communicate the diagnosis in a way that facilitates understanding for patients and agreement with the need for psychological treatment, then successfully refer patients for this treatment. The latter task is inherently difficult given the shortage of mental health clinicians, especially individuals equipped to treat PNES in children. Patients and families may feel shuffled between providers and may resist following up with psychological treatment recommendation due to a lack of acceptance of the diagnosis or difficulty in accessing and establishing treatment [3]. There is a pressing need for a more effective and comprehensive management model tailored to combine the unique aspects of the neurological and mental health care aspects of this patient population.

One potential strategy to optimize care of pediatric patients with PNES is the integration of neurology and mental health clinicians [4]. There have been several single site studies showing the benefit of the multidisciplinary clinic (MDC) model in the management of pediatric patients with PNES [5–7]. Specifically this group showed that a MDC can lead to consistent follow-up, mitigation of barriers of care, diagnosis acceptance, and improved clinical outcomes with high remission rates [6]. They also showed decreased unnecessary health care utilization, such as emergency room visits and unplanned hospitalizations after patients were seen in the MDC [7]. This work has demonstrated that the MDC can be a successful model for the outpatient management of pediatric patients with PNES.

The aim of this study was to develop national consensus based on expert opinion on the optimal outpatient care model of pediatric PNES. Our hypothesis was that experts would recommend the MDC as the optimal outpatient model for the care of pediatric PNES. We also aimed to develop consensus on the essential and ideal personnel and services provided in an MDC.

2. Methods

2.1. Study overview

The study was conducted using a three-round Delphi technique. To do this, we established a core working group (CWG) within the PNES special interest group of the Pediatric Epilepsy Research Consortium (PERC). This CWG was composed of neurologists, psychiatrists, and psychologists. The three rounds of questions were sent to experts on March 14th, 2023, July 20th, 2023 and January 2nd, 2024, respectively, and experts were given 4 weeks to respond.

2.2. Establishing the expert panel

The CWG established a rigorous scoring rubric to select experts in pediatric PNES (Table 1). Emphasis was placed on the individual's participation in a multidisciplinary PNES clinic, duration of work with patients with PNES, national reputation and scholarly work. Experts were only chosen from clinics in the United States of America (USA) as resources and method of practice are more standardized across institutions as compared to other countries. Experts were identified through review of the literature and nominations from the CWG.

Table 1

The following criteria were used to identify experts in the field of pediatric psychogenic nonepileptic seizures. Experts who scored 3 points or greater were invited to participate in this Delphi study.

Criteria	Points	
Duration Practicing Specialty		
5–10 years	1 point	
>10 years	2 point	
Involved in a MDC for ≥ 2 years	2 points	
Papers published on PNES		
1–3	1 point	
>3	2 points	
National Presentations		
Poster	1 point per poster	
Presentation	2 points per presentation	
National presence in the field	1 point	

Abbreviations: MDC, multidisciplinary clinic; PNES, psychogenic nonepileptic seizures.

Table 2

Diagnostic levels of certainty for psychogenic nonepileptic seizures. Table adapted from Lafrance et al., 2013. Diagnosis requires a history consistent with psychogenic nonepileptic seizures.

Diagnostic Level	Diagnosis established as follows:	
	Type of witnessed event	EEG
Documented	PNES captured with video EEG	Ictal EEG with video normal
Clinically established	Video or in-person observation	Ictal EEG without video normal
Probable	Video or in-person observation	Interictal EEG normal
Possible	Witness description or patient description	Interictal EEG normal

Abbreviations: PNES, psychogenic nonepileptic seizures; EEG, electroencephalogram.

National presence factored in involvement with professional societies and participation in special interest groups about PNES. Experts who scored 3 points or greater on the selection criteria were invited to participate in the study. E-mail invitations were sent to experts and surveys were sent to those who agreed to participate. Unique survey links were utilized and responses remained anonymous. Experts received no compensation for their participation.

2.3. Delphi Questionnaire

The current literature was first summarized by the CWG to determine available data and deficiencies on this topic (Appendix A). Emphasis was placed on the following categories: personnel and structure of the PNES clinic, name of the diagnosis, diagnostic criteria, screening tools, trainee inclusion, patient resources, referrals and treatment. Based on the literature review findings, we created the first round of Delphi questions. A second and third round of questions were developed based on topics in which consensus was not reached during the previous round (Appendix B). Assessment of diagnostic criteria for the inclusion of patients within the MDC was based on the report from the International League Against Epilepsy (ILAE) Nonepileptic Seizures Task Force [8]. Expert responses were collected and managed using REDCap (Research Electronic Data Capture) electronic data capture tools hosted at University of Texas Southwestern (UTSW) Medical Center [9,10]. REDCap is a secure, web-based software platform designed to support data capture for research studies, providing 1) an intuitive interface for validated data capture; 2) audit trails for tracking data manipulation and export procedures; 3) automated export procedures for seamless data downloads to common statistical packages; and 4) procedures for data integration and interoperability with external sources.

2.4. Standard protocol approvals, registrations, and patient consents

The study was reviewed and deemed exempt by the Institutional Review Board at UTSW. Consent was waived as part of the exempt status.

2.5. Analysis

The following criteria were used to determine strength of consensus, based on methodology used in Wirrell et al [11]. Consensus was defined as follows:

- Strong: more than 80 % of the members agreed or strongly agreed and no more than 25 % disagreed.
- Moderate: 67–80 % agreed or strongly agreed.
- No consensus: if neither of the above criteria were met.

For items that consensus was not reached, a second and third round of questions were sent to the group of experts for further evaluation.

3. Results

3.1. Respondents

Thirty-six potential experts were screened using the scoring rubric, 25 of whom met criteria and were invited to participate. Eighteen pediatric experts completed the first round of the Delphi survey, including 5 neurologists, 5 psychiatrists, 6 psychologists, 1 social worker and 1 nurse (Table 3). Seventeen experts completed the second round of the survey, and 16 completed the third round. The total years in practice ranged from 2 to 40, with an average of 16 years across all respondents. Half worked in academics, 6 % worked in private/community practice, 39 % worked in hospital-based practice and 6 % worked in consulting at the time of the survey. Sixty-seven percent worked in a PNES MDC. Of those, most (58 %) had worked in the PNES MDC for 1–5 years, 17 % had worked 5–10 years and 25 % had worked over 10 years.

3.2. PNES outpatient multidisciplinary clinic model

Table 4 summarizes degree of consensus for major themes surveyed. The multidisciplinary clinic model was reported as the gold standard for outpatient management of pediatric PNES (strong).

Table 3

Demographics of expert panel who participated in the Delphi survey.

Clinical Practice type	Number: N (%)
Academic	9 (50)
Private/community	1 (6)
Hospital based	7 (39)
Clinical Consulting	1 (6)
Involved in PNES MDC	12 (67)
1–5 years	7 (58)
5–10 years	2 (17)
>10 years	3 (25)
Specialty	
Neurology	5
Psychology	6
Psychiatry	5
Nursing	1
Social work	1

Abbreviations: PNES, psychogenic nonepileptic seizures; MDC, multidisciplinary clinic.

Table 4
Summary of results and level of consensus reached.

Variables	Finding Through Delphi	Strength
Clinic model	<ul style="list-style-type: none"> The MDC is the gold standard for outpatient management of pediatric PNES 	<ul style="list-style-type: none"> Strong
Personnel in clinic (resource limited)	<ul style="list-style-type: none"> Neurologist Psychologist Social worker 	<ul style="list-style-type: none"> Strong Moderate Moderate
Personnel in clinic (resource unlimited)	<ul style="list-style-type: none"> Psychiatrist Nurse 	<ul style="list-style-type: none"> Strong Moderate
Mental health clinician	<ul style="list-style-type: none"> A psychologist is preferred over a psychiatrist when only one available 	Moderate
Neurologist role	<ul style="list-style-type: none"> Make the diagnosis Communicate the diagnosis Communicate treatment options 	<ul style="list-style-type: none"> Strong Strong Moderate
Psychologist role	<ul style="list-style-type: none"> Communicate the diagnosis Communicate treatment options Providing treatment Evaluating for comorbid mental health diagnosis Consult with community therapist to aid in care of patient if requested 	<ul style="list-style-type: none"> Strong Moderate Moderate Strong Strong
Psychiatrist role	<ul style="list-style-type: none"> Communicate the diagnosis Communicating treatment options Providing treatment Evaluating for comorbid mental health diagnosis Consult with community therapist to aid in care of patient if requested Consult with community therapist to aid in care of patient if requested 	<ul style="list-style-type: none"> Strong Moderate Moderate Strong Strong
Licensed counselor role	<ul style="list-style-type: none"> Provide treatment Evaluate for comorbid mental health diagnosis Consult with community therapist to aid in care of patient if requested 	<ul style="list-style-type: none"> Strong Strong Strong
Care coordinator	<ul style="list-style-type: none"> Social worker is preferred over case manager 	Strong
Care coordinator role	<ul style="list-style-type: none"> Coordinating with the school Providing resources for treatment in community 	<ul style="list-style-type: none"> Moderate Strong Strong
Nurse role	<ul style="list-style-type: none"> Assess for barriers to care Meeting and rooming the patient Contacting patients after their MDC Answering phone calls 	<ul style="list-style-type: none"> Moderate Moderate Strong Strong
APP presence	<ul style="list-style-type: none"> In the absence of a neurologist, an APP trained in neurology can serve the role In the absence of a mental health clinician, an APP trained in mental health can serve the role 	<ul style="list-style-type: none"> Moderate Moderate
Clinic structure	<ul style="list-style-type: none"> Providers should remain in the exam room during the entirety of the visit Neurologist should see patient first, then mental health clinician The MDC can be administered via telehealth for new and follow-up visits 	<ul style="list-style-type: none"> Moderate Strong Moderate
Billing/Coding	<ul style="list-style-type: none"> Common diagnosis codes used include 'functional neurologic disorders with attacks or seizures' and 'functional neurologic disorders with abnormal movements' Symptom based codes should not be used All providers should bill for their services Time based billing is used more than medical decision making 	<ul style="list-style-type: none"> Moderate Moderate Strong Strong
Trainees	<ul style="list-style-type: none"> Trainees should be included in an MDC Parents/patients should be asked if a trainee can join the visit The recommended number of trainees is 1 to 2 Trainees from neurology, psychology, psychiatry, and general pediatrics can be included Trainees from medical schools, social work and nursing should be included 	<ul style="list-style-type: none"> Strong Strong Strong Strong Moderate

(continued on next page)

Table 4 (continued)

Variables	Finding Through Delphi	Strength
Trainee role	<ul style="list-style-type: none"> • Observe the visit • Obtain the history • Physically examine patients • Only senior trainees can communicate the diagnosis, discuss treatment options and/or provide treatment 	<ul style="list-style-type: none"> • Strong • Moderate • Moderate • Strong
Patient inclusion/exclusion	<ul style="list-style-type: none"> • Documented level of certainty • Clinically established level of certainty • Probably level of certainty • Possible level of certainty • Comorbid epilepsy and/or intellectual disability • No lower age limit for patient inclusion • Upper age limit for patient inclusion • Referrals from outpatient neurology • Referrals from outpatient non-neurology • Referrals from inpatient services or emergency department with neurology consultation • Referrals from epilepsy monitoring unit • Referrals from inpatient services or emergency department without neurology consultation • Self-referrals 	<ul style="list-style-type: none"> • Strong • Strong • Moderate • No • Strong • Strong • Strong • No • Strong • Moderate • Strong • Strong • No • Strong
End of visit resources	<ul style="list-style-type: none"> • PNES action plan should be provided • Printed reading material, websites, and/or support group information should be provided 	<ul style="list-style-type: none"> • Strong • Strong
Follow-up	<ul style="list-style-type: none"> • Patients should be contacted after the visit to assess for further needs • Patients should be contacted 1 month after their visit • Needs that should be assessed during 1-month contact include diagnosis acceptance, persistence of events, barriers to treatment, barriers to attending school, barriers to activities of daily living • Follow-up in the MDC should be provided as needed per patient • Follow-up with neurology for PNES is recommended, with frequency tailored to each patient 	<ul style="list-style-type: none"> • Strong • Moderate • Strong • Strong • Moderate
Treatment	<ul style="list-style-type: none"> • If resources available, the MDC should provide treatment • All patients should be referred to psychology/counseling • Referral to psychiatry should be given for medical treatment of comorbid psychiatric conditions, or when hallucinations or suicidal ideation is present • Psychoeducation is a recommended treatment option • Psychotherapy is a recommended treatment option • CBT is a recommended treatment option • CBT is preferred over psychotherapy • Hypnosis, physical and occupational therapy, and neurofeedback are not indicated treatments • Group therapy, and medications are not indicated treatments • Referral to higher level of care should be given for inability to attend school • Referral to higher level of care should be given for impairment in activities of daily living or inability to eat/drink 	<ul style="list-style-type: none"> • Strong • Moderate • Moderate • Strong
Terminology	<ul style="list-style-type: none"> • Parents and families are confused by the term PNES • Ideal term for PNES • Ideal prefix or suffix • The term nonepileptic should be included 	<ul style="list-style-type: none"> • Strong • No • No • Moderate

Abbreviations: MDC, multidisciplinary clinic; PNES, psychogenic nonepileptic seizures; APP, advanced practice provider; CBT, cognitive behavioral therapy.

3.2.1. Personnel

In a limited resource setting, a neurologist (strong), psychologist (moderate) and social worker (moderate) are essential members of the clinic. In an unlimited resource setting, in addition to the above-mentioned, psychiatrist (strong) and nurse (moderate) inclusion is also recommended. Given the choice of mental health clinician, experts recommend psychology over psychiatry (moderate).

The role of the neurologist is to make the diagnosis (strong), communicate the diagnosis (strong) and communicate treatment options to patients and families (moderate). In the absence of a physician, an advanced practice provider (APP) trained in neurology can serve the role of the neurologist in the MDC (strong). The role of the APP includes communicating the PNES diagnosis and communicating treatment options only (strong). Psychologists' role in the clinic includes communicating the diagnosis (strong), communicating treatment options (moderate), providing treatment for PNES (strong) and evaluating for comorbid mental health diagnoses (strong). The preferred provider for providing psychoeducation is psychologists (moderate). The psychiatrist's role can include communicating the diagnosis (strong), communicating treatment options (moderate), providing treatment for PNES (moderate) and evaluating for comorbid mental health diagnoses (strong). A licensed counselor's role can include providing treatment for PNES (strong) and evaluating for comorbid mental health diagnoses (strong). In the absence of psychology, psychiatry or a counselor, an APP trained in mental health can be included in the clinic (moderate). Many respondents however emphasized the importance of adequate APP experience and knowledge of PNES, noting: "An APP with training is helpful as a substitute since there is a shortage of behavioral mental health professionals" and that an APP can be included "If that person can provide psychoeducation, response plan and discussion of treatment." The role of any mental health clinician in the MDC should include consulting with the community therapist to help with a patient's treatment of PNES if requested (strong). When present, the role of the care coordinator in the MDC includes coordinating with school (moderate), providing resources for accessing treatment in the community (strong) and for assessing for barriers to care (strong). If given the choice of a social worker or case manager for care coordination, social work is preferred (strong). The role of the nurse should include checking the patient in (moderate), contacting patients after their MDC visit (moderate), and answering patient calls (strong).

It is preferred for all providers in the MDC to remain in the exam room at the same time (moderate). The order in which patients are evaluated is important (moderate), with neurologists seeing patients first, followed by the mental health clinician (strong). A PNES MDC can be successfully administered via telehealth and experts believe new and follow-up visits can be done via telehealth (moderate).

3.2.2. Billing/Coding

Neurology, psychology and psychiatry should each bill for their services in the MDC (strong). Psychiatrists and neurologist report using time-based billing over medical decision making (MDM) (strong). The most common diagnosis code used is 'functional neurological disorders with attacks or seizures' and 'Functional neurologic disorders with abnormal movements' (moderate). Symptom based codes (i.e., abnormal involuntary movements, convulsions, etc) should not be used as diagnosis codes when seeing patients with PNES (moderate).

Billing for psychologists was more varied and consensus was not reached as a result. Of the 6 psychologists, 2 use health and behavior codes due to hospital policy while 2 other psychologists do not use health and behavior codes as they are reportedly not consistently reimbursed. The latter 2 psychologists use Current Procedural Terminology (CPT) code 90791. One psychologist bills similar to physicians since this disorder is listed within the Diagnosis and Statistical Manual of

Mental Disorders (DSM) and consistent with a medical diagnosis. One psychologist does not do their own billing and instead their program bills as a collective whole.

3.2.3. Trainees

Experts agreed that trainees should be included in the PNES MDC (strong). Parents and patients should be asked if a trainee can join the clinic visit (strong). The recommended number of trainees is 1 to 2 residents (strong) with no consensus reached on which number is most ideal. Trainees from neurology, psychology, psychiatry, and general pediatrics can be included (strong); trainees from medical schools, social work and nursing can also be included (moderate). Trainees of any level should be allowed to observe the visit (strong), obtain the history (moderate) and/or complete a physical exam (moderate). In addition, senior level trainees can also communicate the diagnosis, discuss treatment options, and/or provide treatment (strong).

3.2.4. Patient inclusion and exclusion

The following patients were recommended to be included in the clinic for outpatient management using the ILAE diagnostic certainty levels (Table 2): Documented (strong), Clinically established (strong), and Probable (moderate) [8]. Consensus was not reached if patients with a Possible level of diagnostic certainty should be included in the clinic.

Patients with comorbid epilepsy and/or intellectual disability can also be seen in the clinic (strong). Experts believe a lower age limit is not needed for patient inclusion (strong). Consensus on upper age limit was not reached, with experts split between age 18 and age 21.

Outpatient referrals can come from neurology clinics (strong) or non-neurology clinics (moderate), either internal or external to the MDC site. Inpatient referrals can come from inpatient services with neurology consultation and epilepsy monitoring units, either internal or external to the MDC site (strong). Referrals from the emergency department with neurology consultation can also be included (strong). Consensus was not reached if self-referrals, referrals from the emergency departments without neurology consultation or inpatient referrals without neurology consultation should be accepted. A health professional should review all referrals to the PNES clinic prior to scheduling or accepting the patient (strong). A health professional should also contact families prior to their visit to assess needs and/or barriers to attending the visit (strong). Screening tools, such as for anxiety or depression, should be used in the clinic (strong), however consensus was not reached on the best instrument.

3.2.5. End of visit/Treatment

A PNES action plan, a guide for responding and caring for patients when actively experiencing an event, should be provided to patients (strong). Printed reading material, websites on PNES diagnosis and treatment, and support group information should be provided to patients (strong). Consensus was achieved that it is best practice to contact patients (via phone call or patient messaging) after the initial clinic visit to identify any issues and assess for further needs (strong); the first follow-up contact after the initial visit should be done after 1 month (moderate). The following needs should be assessed during this contact: diagnosis acceptance, persistence of events, barriers to establishing treatment, any barriers to attending school and barriers to participating in activities of daily living (strong).

If the resources are available, a PNES MDC should provide treatment to patients (strong). All patients should be referred to psychology/counseling for treatment (moderate). Only certain patients should be referred to psychiatry (moderate). Patients should be referred to psychiatry when medical treatment is desired for comorbid psychiatric conditions or when hallucinations or suicidal ideation is present (moderate). PNES treatment options include psychoeducation (strong), psychotherapy (moderate), cognitive behavioral therapy (CBT) (strong). Between individual psychotherapy and CBT, the latter was reported to be the best treatment (moderate). Treatments not felt to be helpful for

PNES itself were hypnosis (strong), physical and occupational therapy (strong), neurofeedback (strong), group therapy (moderate), and medications (moderate). Specifically, experts cited lack of data to support the use of group therapy in the pediatric population with PNES. Patients should be referred to a higher level of mental health care (i.e., intensive outpatient program) if they are not able to attend school (moderate), have impairment of activities of daily living (strong), or are not eating/drinking (strong) due to their PNES. Prolonged PNES is not an indication for higher level of care (moderate).

For individual patients, a follow-up in the MDC should be provided as needed (strong). Follow-up with neurology for PNES is recommended (moderate); many experts agreed with this recommendation if events are continuing or diagnosis acceptance has not been reached. Frequency of follow-up with neurology should be tailored to each patient (moderate). Antiseizure medications should be weaned by neurology at the initial PNES visit if there is no comorbid epilepsy present (strong).

3.3. Name of diagnosis

Experts agree that patients and families are often confused by the term PNES (strong), and they report a new term should be developed (moderate). However, consensus was not reached on the ideal name of the diagnosis. Experts believe that the word nonepileptic should be included in the name (moderate). However, consensus could not be reached on whether 'functional' versus 'psychogenic' was most ideal for the first part of the name, with 65 % of experts reporting 'functional' as the most ideal term. Similarly, for the second part of the name, consensus was not reached on whether 'seizures' versus 'events' versus 'spells' was most ideal, with 65 % of experts reporting 'events' as most ideal. Experts provided many comments on this topic in the optional open-ended responses. Three comments pertained to avoiding the term 'seizure' in the name to minimize confusion for schools and caregivers.

4. Discussion

This is the first national Delphi Study to investigate consensus on the integrated outpatient care of pediatric PNES. Similar to previous single-site studies, experts agreed that multidisciplinary team care is the most ideal outpatient care model for helping children and adolescents with PNES [5–7]. This study further complements the recently published recommendations on the management of pediatric PNES from the Pediatric Psychiatric Issues Task Force of the ILAE [12].

Families of children with PNES face barriers to care, including a myriad of social determinants of health factors [13–15]. Outcomes from a pediatric PNES MDC have shown that screening charts prior to the clinic visit to identify potential barriers to care followed by support and mitigation of those barriers with the help of a social worker may lead to reduced barriers to care, especially for non-white patients. A multidisciplinary team helps address the unique biopsychosocial issues that pediatric patients with PNES face, including barriers to care, poor quality of life, discrimination in school, social stigma, and high rates of comorbid psychological symptoms [13].

In order to further understand how best to design and implement an MDC, we surveyed our experts on several key topics. Specifically, there is consensus that at least a neurologist, psychologist and social worker should be involved in the multidisciplinary team, with additional benefits of having psychiatry and nursing present when resources are available. This is reflective of the recommended evaluation and treatment of pediatric PNES from previous reports [1,2]. There was agreement that the MDC could be successful via a telehealth platform and may further improve access to specialty care. This is supported by the successful implementation of an MDC via telemedicine during the COVID-19 pandemic [16].

We investigated current billing practices and found that neurologists and psychiatrists predominantly use time-based billing. This is likely due to the effort spent evaluating a patient with PNES is best captured

with time-based billing as compared to MDM-based billing given the nature of the diagnosis. This observation is imperative for the sustainability of a PNES MDC. Further, the MDC may ultimately lead to cost savings with reductions in unnecessary healthcare utilization, such as emergency department visits, unplanned hospitalizations and neurodiagnostic studies [7]. Consensus was not reached on psychology billing, largely due to differences in hospital practices and rules.

There was also strong agreement that trainees can and should be included in the clinic. Inclusion of trainees can help ensure the next generation of providers gain the knowledge and skills to meet the needs of these patients. Specifically, child neurology residents would benefit from exposure to an MDC approach to PNES care given that patients with functional neurologic symptoms comprise a substantial portion of pediatric neurology patients, with a PNES prevalence of 59.5 cases per 100,000 patients aged 16–19 years [17]. Talai et al (2023) recently surveyed child neurology residents and residency program directors who both expressed a desire for increased and improved education on this topic, further emphasizing the importance of educating trainees [18].

There was also agreement that patients can be seen with most levels of diagnostic certainty based on the ILAE diagnostic certainty levels [8]. There was strong consensus that a PNES action plan should be provided to patients. There are no foundation-sponsored action plans for PNES as there are for ES, however schools often request and require guidance on the management of a child with PNES [19]. School nurses struggle with a lack of communication from healthcare providers and want early and clear communication about the diagnosis [20]. Therefore, it is imperative to provide guidance on how best to care for this patient population in the school setting. Further studies are needed to explore this topic and optimize this communication.

Consensus was reached on treatment and referral indications for patients seen in the PNES MDC. All patients should be referred to psychology/counseling and CBT is preferred over individual psychotherapy. This finding is supported by the only prospective study analyzing treatment in pediatric PNES, which showed that employing CBT strategies can lead to a high remission rate [21].

Experts could not agree on the terminology for what to call PNES. This discussion over terminology is important as more patients, advocacy organizations, and professional societies drive the change from PNES to FS. This conflicts with data from the experts surveyed here, who preferred to avoid the term seizure to avoid confusion with ES. Previous surveys done with neurologists, patients and/or parents have also produced conflicting results on the preferred term [12,22–24]. Further data regarding relevant patient stakeholders and expert opinion is needed, with preliminary work already underway [25].

4.1. Limitations of the study

Our main study limitation is that this study employed the Delphi technique and was not based on prospective patient data. In the absence of robust data in the pediatric literature, understanding expert opinion on this topic can serve as the steppingstone for more rigorous studies. Many rare epilepsies have also employed a similar Delphi technique to explore their respective diseases further as well [11,26,27]. Another limitation is that our expert panel consisted of a low number of experts that met our criteria and was largely comprised of academic or hospital-based providers with low representation from community or private practice, including some experts from the same institution. Unfortunately, there is a national dearth of experts in the field of pediatric PNES. Further exploration into best models of practice for PNES management in the latter practice setting is needed, though many of the topics covered in this study are still applicable. A component of our scoring rubric selected for individuals involved in an MDC which may have contributed to a preference for the MDC model, however this represented only 1 of 5 criteria evaluated in the rubric. In addition, only experts from the U.S. were included in order to better understand

practices within the American Healthcare system, and there are no doubt experts in this field outside the US system that could add valuable insights into the care of these patients. Lastly, a limitation of this study is that we did not include patient families in the survey; they are important stakeholders in this arena, and a future iteration of this project will include their input.

5. Conclusion

This national Delphi study provides strong recommendations for the multidisciplinary management of pediatric PNES in the outpatient setting and gives specific recommendations that can facilitate the development and implementation of MDCs in other institutions. Further prospective studies are warranted to validate this practice model.

6. Funding sources

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CRediT authorship contribution statement

Afsaneh Talai: Writing – original draft, Methodology, Formal analysis, Data curation. **Daniel A Freedman:** Writing – original draft, Methodology. **Kristen Trott:** Writing – review & editing, Methodology. **Maija R Steenari:** Writing – review & editing, Methodology. **Sigita Plioplys:** Writing – review & editing, Methodology. **Hillary Kimbley:** Writing – review & editing, Methodology. **Jennifer Madan Cohen:** Writing – review & editing, Methodology. **Priya Tatachar:** Writing – review & editing, Methodology. **Dara V.F. Albert:** Writing – original draft, Methodology.

Declaration of competing 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.

Acknowledgements

This research was done on behalf of the Pediatric Epilepsy Research Consortium.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.yebeh.2024.110096>.

References

- [1] Doss JL, Plioplys S. Pediatric psychogenic nonepileptic seizures: a concise review. *Child Adolesc Psychiatr Clin N Am* 2018;27:53–61.
- [2] Dworetzky BA. What Are We Communicating When We Present the Diagnosis of PNES? *Epilepsy Curr* 2015;15:353–7.
- [3] Howlett S, Grünewald RA, Khan A, Reuber M. Engagement in psychological treatment for functional neurological symptoms—Barriers and solutions. *Psychotherapy (Chic)* 2007;44:354–60.
- [4] Sawchuk TAJ, Terry D. *Psychogenic Non-Epileptic Seizures: Toward the Integration of Care*. New York: Oxford University Press; 2017.
- [5] Fredwall M, Terry D, Enciso L, Burch MM, Trott K, Albert DVF. Outcomes of children and adolescents 1 year after being seen in a multidisciplinary psychogenic nonepileptic seizures clinic. *Epilepsia* 2021;62:2528–38.
- [6] Terry D, Enciso L, Trott K, Burch MM, Albert D-A-O. Outcomes in children and adolescents with psychogenic nonepileptic events using a multidisciplinary clinic approach. *J Child Neurol* 2020;35:918–23.
- [7] Rao S-A-O, Burch M-A-O, Fredwall M-A-O, et al. Multidisciplinary care for pediatric patients with psychogenic nonepileptic seizures may be associated with decreased health care utilization. *Neurol Clin Pract* 2023;13:1–6.
- [8] LaFrance Jr WC, Baker GA, Duncan R, Goldstein LH, Reuber M. Minimum requirements for the diagnosis of psychogenic nonepileptic seizures: a staged

approach: a report from the International League Against Epilepsy Nonepileptic Seizures Task Force. *Epilepsia* 2013;54:2005–18.

[9] Harris RT PA, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap) – a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform* 2009;42: 377–81.

[10] Harris RT PA, Minor BL, Elliott V, Fernandez M, O’Neal L, McLeod L, Delacqua G, Delacqua F, Kirby J, Duda SN. REDCap Consortium, The REDCap consortium: building an international community of software partners. *J Biomed Inform* 2019.

[11] Wirrell EC, Hood V, Knupp KG, et al. International consensus on diagnosis and management of Dravet syndrome. *Epilepsia* 2022;63:1761–77.

[12] Reilly C-A-O-X, Jette N-A-O, Johnson E-A-O, et al. Scoping review and expert-based consensus recommendations for assessment and management of psychogenic non-epileptic (functional) seizures (PNES) in children: a report from the Pediatric Psychiatric Issues Task Force of the International League Against Epilepsy. *Epilepsia* 2023;64:3160–95.

[13] Burch MM, Rossiter S, McGinty-Kolbe K, et al. Multidisciplinary care may help to reduce disparities associated with race in youth with psychogenic nonepileptic seizures. *Pediatr. Neurol.* 2024;151:29–33.

[14] Gregerson CHY, Bakian AV, Wilkes J, et al. Disparities in pediatric epilepsy remission are associated with race and ethnicity. *J Child Neurol* 2019;34:928–36.

[15] Teagarden DL, Morton ML, Janocko NJ, et al. Socioeconomic disparities in patients with psychogenic nonepileptic seizures and their caregivers. *Epilepsy Behav* 2020; 110:1–5.

[16] Fredwall M, Terry D, Enciso L, Burch MM, Trott K, Albert DVF. Short-term outcomes in pediatric and adolescent patients with psychogenic nonepileptic events seen by telemedicine during the COVID-19 pandemic. *Epilepsy Behav* 2021; 117:1–5.

[17] Villagrán A-A-O, Eldøen G, Duncan R, Aaberg KM, Hofoss D, Lossius MI. Incidence and prevalence of psychogenic nonepileptic seizures in a Norwegian county: a 10-year population-based study. *Epilepsia* 2021;62:1528–35.

[18] Talai AFD, Albert DVF. Educating child neurology residents about psychogenic nonepileptic seizures: a needs assessment. *Neurology Education* 2023;3:1–7.

[19] Seizure Action Plans, Epilepsy Foundation; 2013 (accessed 3/11/2024, at <https://www.epilepsy.com/preparedness-safety/action-plans>).

[20] Terry D, Trott K. A qualitative analysis of school nurses’ experience caring for students with psychogenic nonepileptic events. *J Sch Nurs* 2021;37:441–8.

[21] Fobian AD, Long DM, Szafarski JP. Retraining and control therapy for pediatric psychogenic non-epileptic seizures. *Ann Clin Transl Neurol* 2020;7:1410–9.

[22] LaFrance Jr WC, Rusch MD, Machan JT. What is “treatment as usual” for nonepileptic seizures? *Epilepsy Behav* 2008;12:388–94.

[23] Stone J, Campbell K, Sharma N, Carson A, Warlow CP, Sharpe M. What should we call pseudoseizures? The Patient’s Perspective *Seizure* 2003;12:568–72.

[24] Morgan LA, Dvorchik I, Williams KL, Jarrar RG, Buchhalter JR. Parental ranking of terms describing nonepileptic events. *Pediatr. Neurol.* 2013;48:378–82.

[25] Wardrope A, Dworetzky BA, Barkley GL, et al. How to do things with words: two seminars on the naming of functional (psychogenic, non-epileptic, dissociative, conversion,) seizures. *Seizure* 2021;93:102–10.

[26] Joshi C, Nickels K, Demarest S, Eltze C, Cross JH, Wirrell E. Results of an international Delphi consensus in epilepsy with myoclonic atonic seizures/ Doose syndrome. *Seizure* 2021;85:12–8.

[27] Wickstrom R, Taraschenko O, Dilena R, et al. International consensus recommendations for management of New Onset Refractory Status Epilepticus (NORSE) incl. Febrile Infection-Related Epilepsy Syndrome (FIRES): statements and Supporting Evidence. *Epilepsia* 2022;63:2840–64.