Scoping Review and Expert-Based Consensus Recommendations for Assessment and Management of Psychogenic Non-Epileptic (Functional) Seizures (PNES) in Children: A Report from the Psychiatric Pediatric Issues Task Force of the International League Against Epilepsy

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Summary

Objective: Limited guidance exists regarding assessment and management of psychogenic non-epileptic seizures (PNES) in children. Our aim was to develop consensus-based recommendations to fill this gap. **Methods**: ILAE Task Force on Pediatric Psychiatric Issues members conducted a scoping review adhering to the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews standards. This was supplemented with a Delphi process sent to pediatric PNES experts. Consensus was defined as ≥80% agreement. **Results:** The systematic search identified 77 studies, the majority (55%) of which were retrospective (only one randomized clinical trial). The primary means of PNES identification was video EEG (vEEG) in 84% of studies. Better outcome was associated with access to counselling/psychological intervention. Children with PNES have more frequent psychiatric disorders than controls. The Delphi resulted in 23 recommendations: Assessment - There was consensus on the importance of (1) taking a comprehensive developmental history; (2) obtaining a description of the events; (3) asking about potential stressors; (4) the need to use vEEG if available whilst parent, self and school reports and video recordings can contribute to a 'probable' diagnosis; (5) that invasive provocation techniques or deceit should not be employed. Management - There was consensus about the (1) need for a professional with expertise in epilepsy to remain involved for a period after PNES diagnosis; (2) provision of appropriate educational materials to the child and caregivers; (3) that the decision on treatment modality for PNES in children should consider the child's age, cognitive ability and family factors. Comorbidities – There was consensus that (1) all children with PNES should be screened for mental health and neurodevelopmental difficulties. **Significance**: Recommendations to facilitate the assessment and management of PNES in children were developed. Future directions to fill knowledge gaps were proposed.

Key Points

- There is limited guidance regarding the assessment and management of PNES in the pediatric population.
- In the scoping review, better outcome was associated with access to counselling or psychological intervention in children with PNES.
- The Delphi process emphasised the need to use video EEG in the assessment of PNES where available.
- There was consensus about the need for effective diagnosis, communication, management plan development and psychological support.
- Children with PNES have a high risk of psychiatric disorders and should be screened and treated for such comorbidities.

Introduction

Psychogenic non-epileptic seizures **(**PNES) are events which can resemble epileptic seizures but without accompanying EEG correlates[1]. They are one of the main differential diagnoses of epilepsy with increasing incidence with advancing age in childhood[2,3,4]. PNES are considered to be a subtype of conversion disorder or functional neurological symptom disorder in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5)[5]. In the International Classification of Diseases, Tenth Revision (ICD-10), PNES falls under the code F44.5, conversion disorder with attacks or seizures[6]. The similarities to epileptic seizures can result in diagnostic delays[7], unnecessary treatment with anti-seizure medication (ASM) and unnecessary investigations[8]. This can expose children to potential iatrogenic side-effects as well as to the stress that accompanies increased hospital visits[9]. PNES can also co-occur with epileptic seizures, contributing to its diagnostic challenge. Over the years, PNES has been referred to by as many as 15 different names[10] including pseudoseizures, nonepileptic attack disorder (NEAD) and functional seizures. There is a range of views regarding what the appropriate name for this condition should be[11,12,13]. For the purpose of the current work, the term PNES is used to acknowledge its previous common use within the International League against Epilepsy (ILAE)[14,15].

Incidence of pediatric PNES in one population-based study from Denmark was 2.4 per 100,000 person/years[3] while its incidence in a Norwegian population-based study was 3.1 per 100,000 person/years for children aged 5-14 years and 9.8 per 100,000 person/years for children aged 15-14 years[4]. However, estimates from registry-based studies are likely underestimated[16].

Misdiagnosis and treatment of PNES as epilepsy[17]or status epilepticus is common[8]. Professional awareness and knowledge about PNES are often deficient and pediatric healthcare professionals report that they want standards for its assessment, diagnosis and treatment in the pediatric population[18,19]. A report by the ILAE indicated that the gold standard for PNES diagnosis is video electroencephalography (vEEG) monitoring, where the event is observed on video, simultaneously co-registered with EEG,[15] although it was acknowledged that vEEG is not always available. This report also proposed levels of diagnostic certainty including 'possible', 'probable', 'clinically established' and 'documented' diagnosis, based on the availability of a history, witnessed event and investigations, including vEEG[15]. The contributory factors, comorbid psychopathology and effective treatments for PNES in children may differ from the adult population[2]. Accurate, early recognition of PNES in children may reduce inappropriate medical investigation and therapy, increase rates of remission, decrease healthcare utilization and improve quality of life for children and their caregivers.

There are currently no accepted recommendations about the assessment and management of PNES in children. The aim of the current paper was to conduct a scoping review of the literature regarding the management and assessment of PNES in children and to develop consensus-based recommendations for its diagnosis and management employing a Delphi process.

Method

Scoping Review

A scoping review was performed rather than a systematic review given the relatively limited research informing the assessment and management of PNES in children. A broad approach was preferred rather than more specific questions which would yield very few studies[20]. The Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-SR)[21] reporting standards were followed (See Supplement 1).

Systematic Search and Screening

The search strategy was developed by a librarian with expertise in scoping and systematic review in collaboration with study investigators with relevant expertise (scoping reviews, pediatric neurology, epilepsy and PNES) (see supplement 2). Searches were ran in Cochrane Central Register of Controlled Trials (CENTRAL), Ovid Medline, Ovid PsycINFO, Ovid Embase, CINAHL (EBSCO), and Web of Science Core Collection and ClinicalTrials.gov on April 14 2020 and again on September 13 2021 to capture new articles. No limits were placed on language. Studies published before 1990 were excluded because it was felt that studies completed prior to 1990 were unlikely to have included vEEG as a method of investigation. Eligibility criteria are listed in Supplement 3. The review was registered at Open Science Framework https://osf.io/nsthf/ on June 2, 2022 (https://doi.org/10.17605/OSF.IO/Z6DW7).

All abstracts were uploaded into Covidence, an online tool that helps streamline the scoping review screening process. 4261 total records were imported into the Covidence software, and after deduplication 2339 original records were screened for inclusion. After duplicates were removed, remaining abstracts were screened by two independent reviewers (CR and EJ). The articles selected for full text review were also reviewed by two independent reviewers (CR and SK). Any conflict was resolved by further discussion and consensus of screeners. A data extraction form was developed for data charting, pilot tested and then revised (see Supplement 4). All data were extracted by one study author and confirmed by a second author. The following data were extracted: Authors, year of publication, study type/design, study location, ascertainment source, study focus, sample size, sample age range, sample sex, number and proportion of participants with co-occurring epilepsy, controls (sample size, age, sex), risk factors/stressors/precipitating factors, definition of PNES used, PNES terminology used, assessment method for PNES, definition of epilepsy, comparison between use of vEEG and any other methods to identify PNES, description of PNES semiology, provocation methods used, use of other methods to discriminate between PNES and epileptic seizures, PNES outcomes, approach to PNES management (e.g. cognitive behavioral therapy versus other measures), assessment of psychopathology (criteria used and prevalence), instruments used to measure psychopathology and results of assessment of cognitive functioning.

Due to the scoping nature of the review, a narrative synthesis of all included studies was undertaken[22]. A narrative synthesis is particularly useful when there is a high degree of variation in the available study data[23]. Data from the scoping review are presented in tandem with the results of the Delphi process under three main headings: Assessment of PNES, management of PNES, and assessment and management of psychopathology in children with PNES.

Delphi Process

Due to the limited evidence regarding the assessment and management of PNES in children, a Delphi method[24,25] was employed to seek consensus on recommendations for clinical practice. Delphi process participants were selected based on their expertise and credibility in the field[26] (e.g., PNES, pediatric epilepsy). We also made sure there was representation from all ILAE regions. In addition to selecting participants based on their expertise in the field (from prior publications), nominations for participants in the Delphi process were sought from the chair of each ILAE regions and from members of the ILAE Task Force on Pediatric Psychiatric Issues.

Delphi statements (see Supplement 5) were developed by members of the ILAE Task Force on Pediatric Psychiatric Issues. The Delphi survey contained 28 statements, 26 of which were based on a 5-point Likert response scale (Strongly agree (1), agree, neither agree or

disagree/disagree/strongly disagree (5)) and two based on ranking of preferences with respect to PNES terminology. The initial survey hosted on *SurveyMonkey* was emailed to 66 participants on August 18, 2021. Three email reminders were sent. Thirty-three participants responded to the initial survey (see Supplement 6 for characteristics). Five of the 33 studies provided demographic data but did not proceed to the main PNES questions as they indicated that 'they were not involved in the care of young people with epilepsy'. The level of agreement for consensus was set at 80% (Agree/strongly agree). Participants were encouraged to elaborate on their answer if they 'disagreed' or 'strongly disagreed' with a statement.

The second round of the Delphi survey included six questions (see Supplement 5) where 80% agreement had not been reached in the first round. The second-round questionnaire was sent on November 19, 2021 to those (n=28) who responded in the first round and were involved in the care of young people with seizures/PNES. These six questions were modified based on feedback from round 1. Again, a total of three reminders were sent.

Statistical Analysis and Consensus Formulations

We used descriptive statistics (means, medians, and ranges) to describe the results of the scoping review and responses to the Delphi statements.

Formulating the Recommendations

The survey responses were converted into recommendations if consensus was reached i.e., ≥80% "agree/strongly agree". We adopted the following strategy:

1- Consensus reached i.e., ≥80% "agree/strongly agree" on first round of Delphi: This was included as a recommendation.
2- Consensus not reached i.e., <80% but ≥50% "agree/strongly agree": Such recommendations were revised by members of the ILAE Task Force on Pediatric Psychiatric Issues based on the feedback received in the first round and was subjected to a second consensus round. It was included as a recommendation if consensus was reached i.e., ≥80% "agree/strongly agree" during the second Delphi round.
3- Consensus not reached i.e., <80% "agree/strongly agree" during the first round or after rewording for the second round resulted in recommendation being removed.

Results

Systematic Search

The results of the scoping review search process are shown in Figure 1. The initial search identified 4261 records, 2339 abstracts after duplicates were removed, 808 articles eligible for full text review and 77 studies meeting all eligibility criteria.

Study Characteristics (see Table 1)

Studies were from 21 different countries whilst two were multinational. The countries where most studies were from included the United States (26), India (9) Turkey (7) and the United Kingdom (6). Thirty-five studies were prospective whilst the remaining 42 were retrospective. Fifty-one studies were cohort/cross-sectional without a control group, 15 case-control, one randomized control trial and 10 'other' designs. Seventy-four studies were 'hospital based' whilst the other three were 'population-based' in that they aimed to identify all children with PNES in a defined geographical area. Studies focused on assessment of PNES (26), semiology of PNES (24), assessment and/or management of psychopathology in PNES (20), risk factors for PNES (16), management of PNES (11), cross-cultural comparisons (1) and other (3). Studies could have more than one focus. Sample size ranged from 10 to 399 (Mean 56.03 and median 35, Interquartile range 25-55) participants. The mean age in the 56 studies where it was

reported was 13.18 (SD 1.92). In the 68 studies where sex was reported, 31% (1236/4041) of children were male.

Characteristics of PNES studies in children with respect to age of onset, age of diagnosis, number with epilepsy and definition of PNES (see Table 2)

In 18 studies where mean age of onset was reported, the pooled mean age of onset was 12.75 with a mean range of 8.9 to 14.3 years. The pooled mean delay to diagnosis (n=17 studies) was 1.21 years with a range of 0.25 to 3.5 years (Median 0.95 years). The median percentage of children with epilepsy who had PNES was 26% (n=44 studies).

Terminology and criteria used to diagnose PNES in children

In terms of criteria used to define PNES, eight studies specifically mentioned the Diagnostic and Statistical Manual of Mental Disorders (DSM) criteria[47,54,57,61,64,66,82,94], seven ICD criteria[30,31,33,49,50,56,87,] and in six studies the minimum requirements outlined by La France[15] et al. 2013[29,30,31,32,34,37] were employed. Two studies required both La France et al. and ICD criteria[30,31]. All other studies (n=55) employed study specific criteria or did not provide the criteria used to define PNES cases. All studies which employed La France et al. criteria[15] were published since 2020. The terminology used to describe PNES was: PNES (n=53/69%), pseudoseizures (n=8/10%), non-epileptic attack disorder (NEAD) (n=1/1%), non-epileptic seizures (n=6/8%), psychogenic seizures (n=6/8%), psychogenic non-epileptic events (n=2/3%), and pseudo-epileptic seizures (n=1/1%). Six studies used an ILAE definition of epilepsy in studies that included children with epilepsy[34,49,81,82,85,88.]

Respondents in the Delphi survey were asked to indicate, "Which of the following terms do you feel is best when describing paroxysmal events thought to be psychogenic in origin in the pediatric population?" The most popular term selected by respondents to this question (n=28) was non-epileptic events (57%), followed by episodes (18%), seizures (14%), other (7%), attacks (4%), and spells (0%). Respondents were also asked to rank the best names for PNES with professionals, children and family and given the ten options. The most popular option to use with child, family and professionals was 'non-epileptic events." The entire rankings are in Supplement 7.

Recommendations for Assessment of Suspected PNES in Children

Table 3a shows the percent agreement for each of the recommendations subjected to the Delphi process for the assessment of suspected PNES in children. A visual representation of results for round 1 and 2 of the Delphi process are included in Supplement 8a.

Box 1 - Recommendations focusing on PNES assessment with respect to description and possible recording of events

1. **The process of assessment of children with suspected PNES should include taking a comprehensive description of the episodes/events**.
   (e.g., What does the episode look like? When did/does it happen? Who is present? Where does it happen?)

2. **A description of the event by parent, self or school report should be sought since it is useful in determining if events are psychogenic in nature and can contribute to a 'possible' diagnosis of PNES in children by a clinician experienced in the diagnosis of seizure disorders.**

3. **Parent home/school videorecording of events should be sought as it is very important in considering whether events are psychogenic in nature and can contribute to a 'probable' diagnosis of PNES in children by a clinician experienced in the diagnosis of seizure disorders**

4. **If available, video-EEG should be used in all children with suspected PNES and if no epileptic activity is detected during a typical event, then a 'clinically established' and 'documented' PNES diagnosis can be made by a clinician experienced in the diagnosis of seizure disorders** *(an exception may be a focal aware seizure that is not detectable on EEG)*

- ***PNES Semiology***

The scoping review identified 23 studies which included a description of PNES semiology (see Supplement 9). Only six[45,60,68,72,76,91] employed a formal classification system to classify PNES events based on previous classification systems – whilst the remainder used study specific descriptions of semiology or used both. The most used classification method in the included articles were the one from Seneviratne et al[102]. There are no data provided on the reliability of any PNES classification systems or in general on the classification of seizure symptoms. PNES semiology was compared in the following groups: younger vs. older children, children vs. adults, males vs. females and children with PNES vs. children with epileptic seizures. However, direct comparisons between studies are difficult due to the different methods used to describe semiology.

Despite the limited evidence regarding the use of semiology in the diagnosis PNES, there was consensus that the process of assessment of children with suspected PNES should include taking a comprehensive description of the episodes including gathering information about the episodes from the child, if possible, caregivers and school report. It was agreed that this information can contribute to a 'possible' diagnosis of PNES in children by a clinician experienced in the diagnosis of seizure disorders.

- ***PNES Diagnosis***

In the scoping review, the primary method of identifying PNES was vEEG in 84% (n=65) of studies, EEG in 5% (n=4), clinical judgement in 1% (n=1) and was not described in 9% (n=7) (See Table 4). In 65 studies, the majority or all children underwent vEEG but it was not always possible to determine if every single patient with PNES was diagnosed using vEEG.

The results of the Delphi suggested that vEEG, when available, is vital to make a 'clinically established' and 'documented' PNES diagnosis in children. None of the studies in the scoping review compared vEEG to non-vEEG methods of assessment (e.g., mobile phone videos, eyewitness testimony, normal EEG). However, the results of the Delphi survey led to the recommendation that home/school videorecording of events can also contribute to a 'probable' diagnosis of PNES by a clinician experienced in the diagnosis of seizure disorders.

- ***Risk Factors/Stressors for PNES***

Box 2 - Recommendations focused on the need to take a comprehensive developmental history and to ask about potential stressors and to evaluate for the presence of other functional symptoms

5. **It is important to ask children with suspected PNES about potential stressors in their life.**

(e.g., school/academic difficulties, family difficulties, bullying, previous physical/sexual abuse, trauma).

6. **The process of assessment of children with suspected PNES should include taking a comprehensive medical/developmental history.**
   (e.g., asking about other medical conditions, learning/behavior, schooling).

7. **With suspected PNES in children, it is important to ask about other symptoms of conversion disorder/functional neurological disorder.**
   (e.g., pain, sensory or motor)

The results of the 41 studies about factors associated with PNES are in Supplement 10. A range of terms were used to describe PNES risk factors including stressors and adversities. There was a lack of agreement regarding what constitutes a risk factor or how they are measured. Thus, comparisons between studies were difficult. Risk factors can be broadly categorised into school related difficulties (e.g., bullying, and academic difficulties), stress in the family environment (including parental divorce/separation/discord), sexual abuse, physical abuse, fear or rejection, loss/grief, emotional problems, and no identified/unknown cause. In the Delphi survey, there was consensus about the importance of asking about potential stressors in children with suspected PNES.

The scoping review did not identify any studies that examined the utility of a medical history to facilitate the assessment of PNES in children. However, in the Delphi survey, there was consensus that the process of assessing children with suspected PNES should include taking a comprehensive medical/developmental history which is likely to be a routine part of pediatric medical assessment. The scoping review did not identify any studies exploring the efficacy of asking about other symptoms of conversion disorder/functional neurological disorder. However, in the Delphi survey, there was consensus that it is important to ask about other symptoms of conversion disorder/functional neurological disorder.

- ***Provoking Techniques for PNES***

Box 3 - Recommendations highlighting the utility of standard assessment techniques and the need to avoid invasive provocation techniques

8. **The use of standard techniques (e.g., sleep deprivation, hyperventilation, photic stimulation) is appropriate in the assessment of suspected PNES in children to help differentiate between epileptic and nonepileptic events.**

9. **The use of invasive provocation techniques (e.g., saline injection) or deceit should not be employed in the assessment of PNES in children.**

The use of invasive provocation techniques was reported to have been used to elicit PNES in 11 of 77 (14%) studies[33,45,59,69,73,83,84,94,95,97,101] (see Table 4) in the scoping review. Some methods were used in more than one study. The methods used were injection of intravenous saline (8 studies), use of alcohol patch (2), use of body compression (1) and use of tuning fork (1).

While there was consensus by the Delphi participants that the use of standard techniques is appropriate in the assessment of suspected PNES in children to help differentiate between

epileptic and non-epileptic events, they also agreed that the use of invasive provocation techniques (e.g., saline injection) or deceit should not be employed in the assessment of PNES in children.

Recommendations for the Management of PNES in Children

Table 3b shows the percent agreement for each of the recommendations subjected to the Delphi process for the management of PNES in children. A visual representation of results for round 1 and 2 of the Delphi process are included in Supplement 8b.

Box 4 - Recommendations highlighting the need for the pediatric neurologist to collaborate with professionals from psychology/psychiatry at diagnosis and subsequent follow-up

10. **The involvement of both a pediatric neurologist/epileptologist and psychologist /psychiatrist is necessary when PNES is first diagnosed to coordinate management and follow-up.**

11. **A pediatric neurologist (or other professional with expertise in epilepsy) should remain involved for a period after the diagnosis of PNES to manage withdrawal of anti-seizure medications, ensure acceptance of diagnosis and avoid further inappropriate investigations.**

The results of the scoping review (see Table 5) show that whilst management approaches varied between studies, most studies involved multidisciplinary management and/or referral to a psychologist or psychiatrist.

Recommendations for the management of PNES in children based on the Delphi process include a clear focus on the need for involvement of both a pediatric neurologist/epileptologist and psychologist/psychiatrist when PNES is first diagnosed to coordinate management and follow-up.

Box 5 - Recommendations highlighting the need to communicate the PNES diagnosis in children in an effective manner and develop a management plan.

12. **In medical records/reports it should always be clear that PNES refer to events of a psychogenic/functional (and not physiologic) nature that are part of the broader classification of functional neurological disorder/conversion disorder.**

13. **It should be made clear to the child and their family/caregivers that events are not epileptic in nature and that anti-seizure medications are not appropriate treatment. ***
    (*Unless the child also has epilepsy in which case medications would still be appropriate for the epileptic seizures but not the PNES)

14. **In the case of children with both PNES and epileptic seizures, there is a need for the child, their family/caregiver and supporting educational and health professionals to be made aware of manifestation of both epileptic and non-epileptic events. Management plans for both should be available for all children.**

15. **A comprehensive plan (written document) should be developed in collaboration with the child and family to inform all relevant health and educational professionals in the child's network.**

16. **A comprehensive management plan for the events at home, school and other relevant locations with clear indications on what supporting adults should do should be developed and agreed upon by all relevant stakeholders.**

The majority of studies identified in the scoping review (see Table 5) do not explicitly identify the importance of effectively communicating the diagnosis of PNES but the development of management plans is highlighted by a number of studies[25,27,29,36,97.] There was a strong focus on the need to communicate the diagnosis in an effective manner and develop management plans arising from the Delphi survey. Recommendations include the need to ensure that the young person and their family/caregivers are aware that the events are not epileptic in nature and that anti-seizure medications are not appropriate treatment. Additionally, it is recommended that in medical records/reports, it should always be made clear that PNES refer to events of a psychogenic/functional (and not physiologic) nature that are part of the broader classification of functional neurological disorder/conversion disorder. In addition to recommendations on communication of the diagnosis, three recommendations focused on the need for the development of management plans as noted above.

Box 6 - Management recommendations highlighting the need: (1) for educational materials about PNES to be provided to the child and parent and (2) to specifically focus on psychological support/therapy for the child and family

17. **Children should always be given developmentally appropriate visual/written information about the nature and possible causes of PNES and possible management approaches.**

18. **Parents/caregivers should always be given appropriate written/visual information about the nature, possible causes and possible management approaches.**

19. **The decision on treatment modality for PNES in children should consider the child's age, cognitive ability, and family factors. For younger children there may need to be a focus on behavioral approaches and skill teaching. For older children and adolescents, cognitive behavioral therapy may be useful.**

20. **When considering treatment for children with PNES it is important to consider that the family may need psychological support (e.g., psychoeducation, counselling) and this should be made available, where appropriate.**

The results of studies which focused on the treatment of PNES in children are shown in Table 5. Whilst most studies emphasised the need for psychological support, only one RCT was identified. This study involved a comparison between two interventions, a novel cognitive intervention drawn on the principles of cognitive behavior therapy for pediatric PNES called Retraining and Control Therapy (ReACT) versus supportive therapy[35]. The ReACT intervention resulted in significantly greater PNES reduction than supportive therapy, with 100% of patients experiencing no PNES 7 days after ReACT[35]. Outcomes were reported in 19 (25%) of 77 studies whilst mean follow-up time ranged from 3 to 55 months (see Supplement

11). Better outcomes appeared to be associated with access to counselling or psychological interventions whilst worse outcomes appeared to be associated with longer symptom duration (see Supplement 11). In studies where it was reported (n=19 studies), PNES freedom ranged from 16-100% (based on intention to treat analysis i.e., where proportion of loss to follow up patients were reported on these were assumed to not be seizure free). The proportion of children lost to follow-up in 13 studies ranged from 0-39%. The proportion of children without any improvement in seizure frequency in 15 studies ranged from 0% to 65%.

In terms of PNES treatment in children, recommendations developed from the Delphi process focused on the need for psychoeducation for the child and parents as well as psychotherapeutic approaches and psychological support for the family. The recommendations in Box 6 capture the various recommended management approaches.

Results of scoping review and Recommendations for the Assessment and Management of Psychiatric and Cognitive Difficulties in Children with PNES

Table 3c shows the percent agreement for each of the recommendations subjected to the Delphi process for the management of PNES in children. A visual representation of results for round 1 and 2 of the Delphi process are included in Supplement 8c.

Box 7 - Recommendations regarding the assessment and management of psychiatric (mental health and neurodevelopmental) and cognitive difficulties in children with PNES

21. **All children with confirmed PNES should be screened for mental health (e.g., depression, anxiety, trauma) and neurodevelopmental (e.g., ADHD, autism spectrum disorder) difficulties.**

22. **It is recommended that children with confirmed PNES be assessed for learning/cognitive difficulties if it is thought that these difficulties are contributing to the child's PNES or other mental health problems.**

23. **Children with PNES who have confirmed mental health or behavioral difficulties should access evidence-based treatments/supports for depression, anxiety, ADHD etc.**

The results of the scoping review showed that mental health and behavioral comorbidities are frequently documented in children and adolescents with PNES. Table 6 displays the 27 studies which focused on the prevalence of psychiatric disorders in children with PNES. All studies that included measures of psychopathology are listed in Supplement 12. The conditions that were most often studied were depression and anxiety (both 22 studies) followed by post-traumatic stress disorder (PTSD) and ADHD (both 14 studies). The prevalence of any psychiatric disorders ranged from 17% to 100% (n=10 studies), 16 to 65% (n=23) for depression, 7% to 84% (n=21 studies) for anxiety, 3% to 26% (n=13 studies) for PTSD and 4% to 29% (13 studies) for ADHD. Eight of the studies administered the Kiddie Schedule for Affective Disorders and Schizophrenia (K-SADS) interview[103]. A population-based study from Denmark showed that children with PNES had significantly higher prevalence of psychiatric disorders than healthy controls but also higher than in children with epilepsy alone[30]. One controlled study showed that children with PNES had significantly higher level of depression, anxiety, PTSD but not ADHD than their sibling[66]. There are limited data on the prevalence of global or specific cognitive difficulties or difficulties with academic achievement in children with PNES. In some studies children with intellectual disability were excluded. There were

also limited data about the treatment of mental health or behavioral difficulties in children with PNES.

The results of the Delphi survey include recommendations to consider mental health and neurodevelopmental difficulties in children with PNES (see Box 7) in line with data from the scoping review.

Discussion

This scoping review and accompanying Delphi survey highlight what is known about the assessment and management of psychogenic non-epileptic seizures (PNES) in children and provides consensus-based recommendations for clinical practice. With respect to terminology, although PNES is the name most often used in published no consensus was reached based on responses to the Delphi survey. The synthesis of data from the scoping review suggests that vEEG is commonly used at least in research studies to confirm a PNES diagnosis, although no studies comparing vEEG to other methods were identified. The semiology of PNES was not reported in a consistent manner across studies and thus there are limited conclusions that can be drawn about the utility of semiology in the assessment and diagnosis of PNES in children. Most respondents to the Delphi survey agreed with the importance of taking a comprehensive history of the events and a developmental history as well as asking about childhood stressors. Parent and school report of events, recording of the events and use of vEEG were also seen as important in the assessment process. Whilst there was agreement about the use of standard techniques to elicit PNES in children, there was consensus that invasive techniques such as saline injection should not be used to elicit PNES in children. Management of PNES in children would appear to predominantly involve multidisciplinary management and/or referral to a psychologist or psychiatrist although there was only one RCT identified in the scoping review addressing this topic. The results of the Delphi process suggest that the involvement of both neurology and psychology/psychiatry is important at diagnosis and for the short-term follow-up. Additionally, there is a need for a plan to inform relevant stakeholders of the child's diagnosis as well as the development of a written management plan. Children themselves need to be provided with developmentally appropriate information about PNES. Management of PNES should include behavioral approaches and skill teaching for young children, cognitive behavioral therapy for older children and adolescents and the wider family should be offered psychological support. The results of the scoping review suggest that children with PNES have a high prevalence of psychiatric issues. Respondents to the Delphi survey agreed that screening for psychiatric and cognitive difficulties should be part of the routine care for children with PNES.

Regarding assessment of PNES, there was agreement in the Delphi survey that, when available, vEEG should be used in all children with suspected PNES. If no epileptic activity is detected during a typical event, then a 'clinically established' and 'documented' PNES diagnosis can be made by a clinician experienced in the diagnosis of seizure disorders in line with Le France et al (2013)[15]. It must be noted that the scoping review did not identify any studies in children that compared vEEG with other methods highlighting a need for more research in this area. Additionally, respondents felt that home/school videorecording of events is important in considering whether events are psychogenic in nature and can contribute to a 'probable' diagnosis of PNES by a clinician experienced in the diagnosis of seizure disorders. This may be particularly important in resource limited settings where vEEG is not readily available or where children do not experience events during vEEG assessment. There was agreement among respondents to the Delphi survey that the process of assessment of children with suspected PNES should include taking a comprehensive description of the episodes/events

including what the episode(s) look like. The scoping review found that semiology of seizures are not recorded consistently across studies. Some differences were noted between males and females and younger and older children in some studies suggesting a need for more research and for the use of a standardized method of recording event types. The Delphi survey respondents agreed that asking about stressors is important, but the scoping review did not find that stressors or other related constructs were asked about in a consistent manner. The development of a standard way of asking about stressors may facilitate a better understanding of the precipitants of PNES in children. Delphi respondents highlighted the need with suspected PNES in children, to ask about other symptoms of conversion disorder/functional neurological disorder (e.g., pain, sensory or motor) and this is in line with previous research suggesting that children with PNES often have multiple functional neurological disorder symptoms[52]. There was unanimous agreement that the use of invasive provocation techniques (e.g., saline injection) or deceit should not be employed to elicit PNES in children despite this being used in 11 studies in the scoping review.

The scoping review identified only one published RCT focused on the treatment of PNES in children[35]. Most other studies focusing on interventions and management included multidisciplinary management and/or referral to a psychologist or psychiatrist. With respect to psychological interventions, a systematic review highlighted that there are relatively few studies evaluating the effectiveness of psychological interventions for children with PNES[104]. There was, however, much commonality in treatment components across the studies including cooperation or collaboration between physical and mental health services, the assessment and treatment of comorbidities and support for parents and liaison with school[104]. The results of the Delphi survey also highlight the need for psychological support for the child and the family/caregivers with modality depending on the child's age, cognitive ability, and family/caregiver circumstances. Respondents to the Delphi survey also highlighted the importance of ensuring that the child and family/caregivers know that the events are not epileptic and, in this respect, the continued involvement of a child neurologist after the diagnosis of PNES was also endorsed. For children with both epileptic and non-epileptic seizures it was highlighted that there is a need for family/caregiver and supporting educational and health professionals to be made aware of manifestation of both epileptic and non-epileptic events and that management plans for both should be available for all children**.** The importance of providing written or visual information about the nature and possible causes of PNES was also emphasised. Previous qualitative studies highlight that children with PNES and their families want access to educational resources and support groups[9]. Another aspect of management highlighted by the Delphi survey was the need to develop written plans to inform the child's network of the diagnosis and how to manage the psychogenic non-epileptic seizures when they occur.

The results of the scoping review indicate that children with PNES are at high risk of mental health difficulties including depression, anxiety, trauma, and ADHD. There are fewer studies on the prevalence of autism spectrum disorder but there may also be an increased risk of PNES in these children[44,105]. The results of the Delphi survey highlight the need to consider these difficulties in children with PNES. It is important to note that there is some evidence that the validity of self-report screening measures in this population is not optimal highlighting that clinical judgement will be vital in this population[106,107]. There are limited data on the prevalence of cognitive difficulties in PNES in the pediatric population and children with intellectual disability are often excluded from research studies. More studies are needed to explore possible specific cognitive difficulties (e.g., executive functioning difficulties) as well assess prevalence

and manifestations of PNES in children with intellectual disability. Respondents to the Delphi survey highlighted that children with PNES who have confirmed mental health or behavioural difficulties should access evidence-based treatment for depression, anxiety and ADHD. In this regard integrated approaches to the treatment of mental health and PNES (e.g., McFarlane et al[107]) are likely to be particularly promising in that there is likely to be expertise from both neurology and psychology/psychiatry available.

Future Research Directions

The results of the scoping review highlight that there are several areas where further research is warranted. With respect to assessment, studies focusing on a comparison between the validity of vEEG versus other methods (e.g., mobile app videos) would be welcomed given that vEEG is unlikely to be available in all settings. Agreement regarding the classification of the semiological presentation in PNES could allow for a more standardized means of reporting data and enable comparisons between research groups and studies. Additionally, agreement regarding classifications of PNES could also facilitate comparisons with semiology of epileptic seizures in children. In terms of outcome, there is a need for agreed outcomes measures beyond seizure freedom that can be used in PNES research studies in children. There is also a lack of well-validated outcomes measures in functional neurological disorders in general. The development of such measures could lead to increased consistency in outcome measurement and facilitate comparison of treatment effects across treatment modalities[108].

With respect to interventions and management of PNES in children, there is a need for more well-designed intervention studies including parent training, psychoeducation, and psycho-therapy including cognitive behavioral therapy and other psychotherapeutic approaches such as acceptance commitment therapy (ACT). Research on interventions in adults with PNES is less scarce than in children, and a recently published RCT in adults highlights the need to focus on measures such as psychological distress and quality of life as well as seizure freedom[109]. The impact of PNES on the wider family network including caregivers and siblings also needs more attention. Longitudinal studies looking at PNES in children need to focus not only on seizure outcome but also on quality of life, stigma and mental health outcomes. In a population-based study of children with PNES 14.2% had epilepsy and this this is an important subgroup. There is a need for more studies which compare children with PNES alone with children with PNES and epilepsy, to better understand if there are differences which may impact on assessment, intervention and outcomes.

Limitations

There are a number of limitations that need to be considered when interpreting the results of the scoping review and Delphi process. We were not able to locate a small number of possibly relevant articles in full text (n=8) or translate articles into English (n=4). The response rate to our Delphi Survey was only 42%. However, participants were from all ILAE regions, and were from multidisciplinary fields. Finally, recommendations were guided by expert opinion due to the limited evidence rather than a process such as the GRADE process which is commonly used to inform the strength of recommendations in guideline development.

Conclusions

There is currently a lack of guidelines for the assessment and management of PNES in children. Our scoping review highlighted major gaps in research in this area in children. The findings of our Delphi survey provide consensus recommendations and highlight the need for a systematic approach to the assessment of PNES in children. With respect to management, the responses to the Delphi survey highlighted the need for close collaboration between neurology and

psychology/ psychiatry, the need to have a comprehensive plan for informing all relevant health and educational professionals in the child's network and to manage events at home and other relevant locations. Efficacious treatment for PNES in children is likely to include psychological intervention and/or psychoeducation for the child and family. In addition, younger children are likely to benefit from a focus on behavioural approaches and skill teaching. For older children and adolescents, cognitive behavioral therapy may be useful**.** The scoping review highlighted that there is a significant need for more high-quality research to develop the evidence base for this population.

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Other authors have no conflicts of interest to declare.

Author Contributions

CR, KV, JMW, SA, MM, EW, MLS, CYF and MK were responsible for review conceptualisation. CR had responsibility for the review planning and execution. SW, CR, KV, NJ were responsible for the systematic search. CR, EJ and SK screened abstracts and full texts. CR, KV, SMK, MLS, SA, CYF, JMW, MK and EW were responsible for data extraction. CR and FM were responsible for tabulating data. CR with support from NJ and KW were responsible for Delphi Survey development and analysis. CR and NJ were responsible for choosing methods of data visualization. CR wrote initial draft with support of KV and NJ. All authors provided feedback on the initial draft via comments and suggestions.

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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.

References

[1]LaFrance Jr WC, Devinsky O. The treatment of nonepileptic seizures: historical perspectives and future directions. Epilepsia. 2004 Jun;45:15-21.
[2]Reilly C, Menlove L, Fenton V, Das KB. Psychogenic nonepileptic seizures in children: a review. Epilepsia. 2013 Oct;54(10):1715-24.
[3]Hansen AS, Rask CU, Rodrigo-Domingo M, Pristed SG, Christensen J, Nielsen RE. Incidence rates and characteristics of pediatric onset psychogenic nonepileptic seizures. Pediatric research. 2020 Nov;88(5):796-803.
[4]Villagrán A, 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 Jun 2.
[5]American Psychiatric Association, American Psychiatric Association. Diagnostic and statistical manual of mental disorders: DSM-5. Arlington, VA. 2013.
[6]World Health Organization. The ICD-10 classification of mental and behavioural disorders: clinical descriptions and diagnostic guidelines. World Health Organization; 1992.
[7]Valente KD, Alessi R, Vincentiis S, Dos Santos B, Rzezak P. Risk factors for diagnostic delay in psychogenic nonepileptic seizures among children and adolescents. Pediatric neurology. 2017 Feb 1;67:71-7.
[8]Jungilligens J, Michaelis R, Popkirov S. Misdiagnosis of prolonged psychogenic non-epileptic seizures as status epilepticus: epidemiology and associated risks. Journal of Neurology, Neurosurgery & Psychiatry. 2021 Dec 1;92(12):1341-5.
[9]McWilliams A, Reilly C, McFarlane FA, Booker E, Heyman I. Nonepileptic seizures in the pediatric population: A qualitative study of patient and family experiences. Epilepsy & Behavior. 2016 Jun 1;59:128-36.
[10]Morgan LA, Dvorchik I, Williams KL, Jarrar RG, Buchhalter JR. Parental ranking of terms describing nonepileptic events. Pediatric neurology. 2013 May 1;48(5):378-82
[11]Asadi-Pooya AA, Brigo F, Mildon B, Nicholson TR. Terminology for psychogenic nonepileptic seizures: making the case for "functional seizures". Epilepsy & Behavior. 2020 Mar 1;104:106895
[12]Kerr WT, Stern JM. We need a functioning name for PNES: Consider dissociative seizures. Epilepsy & Behavior: E&B. 2020 Mar 8;105:107002.
[13]Beghi M, Peroni F, Cornaggia CM. Reply to: We need a functioning name for PNES: Considering dissociative seizures. Epilepsy & behavior: E&B. 2020 Aug;109:107084.
[14]LaFrance Jr WC, Reuber M, Goldstein LH. Management of psychogenic nonepileptic seizures. Epilepsia. 2013 Mar;54:53-67.
[15]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 Nov;54(11):2005-18.
[16]Asadi-Pooya AA. The true prevalence of psychogenic nonepileptic seizures is much higher than this. Epilepsia. 2021 Nov;62(11):2875-6.
[17]Asadi-Pooya AA, Bahrami Z. Risk factors for the use of antiepileptic drugs in patients with psychogenic nonepileptic seizures. Epilepsy Behav. 2019;90:119–21.
[18]McWilliams A, Reilly C, Heyman I. Non-epileptic seizures in children: Views and approaches at a UK child and adolescent psychiatry conference. Seizure. 2017 Dec 1;53:23-5.
[19] Wichaidit BT, Østergaard JR, Rask CU. Diagnostic practice of psychogenic nonepileptic seizures (PNES) in the pediatric setting. Epilepsia. 2015 Jan;56(1):58-65.
[20]Munn Z, Peters MD, Stern C, Tufanaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. BMC medical research methodology. 2018 Dec;18(1):1-7.

[21]Armstrong R, Hall BJ, Doyle J, Waters E. 'Scoping the scope'of a cochrane review. Journal of public health. 2011 Mar 1;33(1):147-50.
[22]Egger M, Schneider M, Smith GD. Spurious precision? Metaanalysis of observational studies. BMJ. 1998;316(7125):140.
[23] Snilstveit B, Oliver S, Vojtkova M. Narrative approaches to systematic review and synthesis of evidence for international development policy and practice. Journal of development effectiveness. 2012 Sep 1;4(3):409-29.
[24]Nair R, Aggarwal R, Khanna D. Methods of formal consensus in classification/diagnostic criteria and guideline development. In Seminars in arthritis and rheumatism 2011 Oct 1 (Vol. 41, No. 2, pp. 95-105). WB Saunders.
[25]Helmer-Hirschberg, O. The use of the delphi technique in problems of educational innovations. Santa Monica, Calif.: Rand; 1966.
26
[26]Jones J, Hunter D. Consensus methods for medical and health services research. BMJ. 1995 Aug 5; 311(7001):376–380.
[27]Fredwall, M., Terry, D., Enciso, L., Burch, M. M., Trott, K., & Albert, D. V. (2021). Short-term outcomes in pediatric and adolescent patients with psychogenic nonepileptic events seen by telemedicine during the COVID-19 pandemic. *Epilepsy & Behavior*, *117*, 107739.
[28]Thabit, M. N., Sadek, A. A., Motawe, E. S., Ali, R. A. E., & Mohamed, M. M. (2021). Non-epileptic paroxysmal events in paediatric patients: A single tertiary centre study in Egypt. *Seizure*, *86*, 123-128.
[29]Fredwall, M., Terry, D., Enciso, L., Burch, M., Trott, K., & Albert, D. V. (2021). Outcomes of children and adolescents 1 year after being seen in a multidisciplinary psychogenic nonepileptic seizures clinic. *Epilepsia*, *62*(10), 2528-2538.
[30]Hansen, A. S., Rask, C. U., Christensen, A. E., Rodrigo-Domingo, M., Christensen, J., & Nielsen, R. E. (2021). Psychiatric Disorders in Children and Adolescents With Psychogenic Nonepileptic Seizures. *Neurology*.
[31]Sawchuk, T., Buchhalter, J., & Senft, B. (2020). Psychogenic non-epileptic seizures in children–psychophysiology & dissociative characteristics. *Psychiatry Research*, *294*, 113544.
[32]Zhang, L. P., Jia, Y., Huang, H., Li, D. W., & Wang, Y. P. (2021). Clinical Classifications of Children With Psychogenic Non-epileptic Seizure. *Frontiers in Pediatrics*, 996.
[33]Kaczmarek, I., Starczewska, M., Winczewska-Wiktor, A., & Steinborn, B. (2020). Sensitivity and specificity of induction of psychogenic non-epileptic seizures in children and adolescents. *Seizure*, *80*, 278-280.
[34]Masi, G., Madonia, U., Ferrari, A., Sicca, F., Brovedani, P., D'Acunto, G., ... & Lenzi, F. (2020). Psychopathological features in referred adolescents with psychogenic nonepileptic seizures with or without epilepsy. *Epilepsy & Behavior*, *112*, 107431.
[35]Fobian AD, Long DM, Szaflarski JP. Retraining and control therapy for pediatric psychogenic non-epileptic seizures. Annals of Clinical and Translational Neurology. 2020 Aug;7(8):1410-9.
[36]Flewelling, K. D., Koehler, A., Shaffer, J., & Dill, E. J. (2020). Medical and psychosocial outcomes of youth with psychogenic nonepileptic seizures: An observational study. *Epilepsy & Behavior*, *112*, 107383.
[37]Terry, D., Enciso, L., Trott, K., Burch, M., & Albert, D. V. (2020). Outcomes in children and adolescents with psychogenic nonepileptic events using a multidisciplinary clinic approach. *Journal of Child Neurology*, *35*(13), 918-923.
[38]Flewelling, K. D., Koehler, A., Shaffer, J., & Dill, E. J. (2020). Correlates of health-related quality of life in youth with psychogenic non-epileptic seizures. *Seizure*, *83*, 203-207.

[39] [a]Sawchuk, T., Asadi-Pooya, A. A., Myers, L., Valente, K. D., Restrepo, A. D., D'Alessio, L., ... & Buchhalter, J. (2020). Clinical characteristics of psychogenic nonepileptic seizures across the lifespan: an international retrospective study. *Epilepsy & Behavior*, *102*, 106705
[40]Myers, L., Trobliger, R., Bortnik, K., Zeng, R., Segal, E., & Lancman, M. (2019). Dissociation and other clinical phenomena in youth with psychogenic non-epileptic seizures (PNES) compared to youth with epilepsy. *Seizure*, *70*, 49-55.
[41]Asadi-Pooya, A. A., AlBaradie, R., Sawchuk, T., Bahrami, Z., Al_Amer, A., & Buchhalter, J. (2019). Psychogenic nonepileptic seizures in children and adolescents: an international cross-cultural study. *Epilepsy & Behavior*, *90*, 90-92.
[42]Gowda, V. K., Amoghimath, R., Benakappa, N., & Shivappa, S. K. (2019). Spectrum of nonepileptic paroxysmal events in children from Southern India. *Journal of neurosciences in rural practice*, *10*(04), 608-612.
[43]Uzun, O., Akdemir, D., Topcu, M., & Ozsungur, B. (2019). Childhood Traumas, Attachment and Alexithymia in Adolescents with Psychogenic Nonepileptic Seizure Type of Conversion Disorder.
[44]McWilliams, A., Reilly, C., Gupta, J., Hadji-Michael, M., Srinivasan, R., & Heyman, I. (2019). Autism spectrum disorder in children and young people with non-epileptic seizures. *Seizure*, *73*, 51-55.
[45]Madaan, P., Gulati, S., Chakrabarty, B., Sapra, S., Sagar, R., Mohammad, A., ... & Tripathi, M. (2018). Clinical spectrum of psychogenic non epileptic seizures in children; an observational study. *Seizure*, *59*, 60-66.
[46]Kandler, R., Lawrence, S., Pang, C., Lai, M., & Whitehead, K. (2018). Optimising the use of EEG in non-epileptic attack disorder: results of a UK national service evaluation. *Seizure*, *55*, 57-65.
[47]Bursch, B., Forgey, M., Emerson, N. D., Siddarth, P., Weisbrot, D. M., Shaw, R. J., ... & Plioplys, S. (2018). Sibling-controlled study of parental bonding, coping, and urgent health-care use in families with children with nonepileptic seizures. *Journal of pediatric psychology*, *43*(10), 1128-1137.
[48]Luthy, S. K., Moss, A. F., Torok, M. R., McLeod, L., & Wilson, K. M. (2018). Characteristics of children hospitalized for psychogenic nonepileptic seizures due to conversion disorder versus epilepsy. *Hospital pediatrics*, *8*(6), 321-329.
[49]Inaida, S., Kanemoto, K., Tanaka, S., Gouji, Y., Oshima, T., Matsumoto, R., ... & Kawakami, K. (2018). Psychogenic non-epileptic seizures in Japan: Trends in prevalence, delay in diagnosis, and frequency of hospital visits. *Epilepsy & Seizure*, *10*(1), 73-86.
[50] Kozlowska, K., Rampersad, R., Cruz, C., Shah, U., Chudleigh, C., Soe, S., ... & Carrive, P. (2017). The respiratory control of carbon dioxide in children and adolescents referred for treatment of psychogenic non-epileptic seizures. *European child & adolescent psychiatry*, *26*(10), 1207-1217.
[51]Kozlowska, K., Chudleigh, C., Cruz, C., Lim, M., McClure, G., Savage, B., ... & Gill, D. (2018). Psychogenic non-epileptic seizures in children and adolescents: Part II–explanations to families, treatment, and group outcomes. *Clinical child psychology and psychiatry*, *23*(1), 160-176.
[52]Kozlowska, K., Chudleigh, C., Cruz, C., Lim, M., McClure, G., Savage, B., ... & Gill, D. (2018). Psychogenic non-epileptic seizures in children and adolescents: Part I–Diagnostic formulations. *Clinical child psychology and psychiatry*, *23*(1), 140-159.
[53]Doss, J., Caplan, R., Siddarth, P., Bursch, B., Falcone, T., Forgey, M., ... & Plioplys, S. (2017). Risk factors for learning problems in youth with psychogenic non-epileptic seizures. *Epilepsy & Behavior*, *70*, 135-139

[55]Mohamed, I. N., Elseed, M. A., Hamed, A. A., Abdel-Rahman, M. E., El-Sadig, S. M., Omer, I. M., ... & Salih, M. A. (2017). Prevalence of epilepsy in 74,949 school children in Khartoum State, Sudan. *Paediatrics and international child health*, *37*(3), 188-192.
[56]Umesh, S., Tikka, S. K., Goyal, N., Sinha, V. K., & Nizamie, S. H. (2017). Aberrant gamma band cortical sources and functional connectivity in adolescents with psychogenic non-epileptic seizures: a preliminary report. *Psychiatry research*, *247*, 51-54.
[57]Plioplys, S., Doss, J., Siddarth, P., Bursch, B., Falcone, T., Forgey, M., ... & Caplan, R. (2016). Risk factors for comorbid psychopathology in youth with psychogenic nonepileptic seizures. *Seizure*, *38*, 32-37.
[58]Narita, Y., Hamano, S. I., Kuroda, M., & Kikuchi, K. (2016). The clinical features of patients suffering from psychogenic non-epileptic seizures with or without epileptic seizures. *No to hattatsu= Brain and development*, *48*(6), 425-429.
[59]Park, E. G., Lee, J., Lee, B. L., Lee, M., & Lee, J. (2015). Paroxysmal nonepileptic events in pediatric patients. *Epilepsy & Behavior*, *48*, 83-87.
[60]Say, G. N., Taşdemir, H. A., & İnce, H. (2015). Semiological and psychiatric characteristics of children with psychogenic nonepileptic seizures: gender-related differences. *Seizure*, *31*, 144-148.
[61]Rawat, V. S., Dhiman, V., Sinha, S., Sagar, K. J. V., Thippeswamy, H., Chaturvedi, S. K., ... & Satishchandra, P. (2015). Co-morbidities and outcome of childhood psychogenic non-epileptic seizures—An observational study. *Seizure*, *25*, 95-98.
[62]Yadav, A., Agarwal, R., & Park, J. (2015). Outcome of psychogenic nonepileptic seizures (PNES) in children: a 2-year follow-up study. *Epilepsy & Behavior*, *53*, 168-173.
[63]Çitilcioğlu, H. B., Matyar, S., Akgül, O., Bucak, İ. H., & Turgut, M. Serum prolactin levels in differential diagnosis of pediatric epileptic and nonepileptic (pseudo-SEIZURES) seizures.
[64]Sawchuk, T., & Buchhalter, J. (2015). Psychogenic nonepileptic seizures in children—Psychological presentation, treatment, and short-term outcomes. *Epilepsy & behavior*, *52*, 49-56.
[65]Say, G. N., Tasdemir, H. A., Akbas, S., Yüce, M., & Karabekiroglu, K. (2014). Self-esteem and psychiatric features of Turkish adolescents with psychogenic non-epileptic seizures: a comparative study with epilepsy and healthy control groups. *The International Journal of Psychiatry in Medicine*, *47*(1), 41-53
[66]Plioplys, S., Doss, J., Siddarth, P., Bursch, B., Falcone, T., Forgey, M., ... & Caplan, R. (2014). A multisite controlled study of risk factors in pediatric psychogenic nonepileptic seizures. *Epilepsia*, *55*(11), 1739-1747.
[67]Aich, T. K. (2014). Absent posterior alpha rhythm: An indirect indicator of seizure disorder?. *Indian journal of psychiatry*, *56*(1), 61.
[68]Wadwekar, V., Nair, P. P., Murgai, A., Thirunavukkarasu, S., & Thazhath, H. K. (2014). Semiologic classification of psychogenic non epileptic seizures (PNES) based on video EEG analysis: do we need new classification systems?. *Seizure*, *23*(3), 222-226.
[69]Yi, Y. Y., Kim, H. D., Lee, J. S., Cheon, K. A., & Kang, H. C. (2014). Psychological problems and clinical outcomes of children with psychogenic non-epileptic seizures. *Yonsei medical journal*, *55*(6), 1556-1561.
[70]Li, R., Liu, K., Ma, X., Li, Z., Duan, X., An, D., ... & Chen, H. (2015). Altered functional connectivity patterns of the insular subregions in psychogenic nonepileptic seizures. *Brain topography*, *28*(4), 636-645.
[71]Abdel Kader, A. A., & Basheer, M. A. (2014). Semiology of Psychogenic Non-Epileptic Seizures in Adults and Children. *Egyptian Journal of Neurology, Psychiatry & Neurosurgery*, *51*(2).

[72]Dhiman, V., Sinha, S., Rawat, V. S., Vijaysagar, K. J., Thippeswamy, H., Srinath, S., ... & Satishchandra, P. (2014). Children with psychogenic non-epileptic seizures (PNES): a detailed semiologic analysis and modified new classification. *Brain and Development*, *36*(4), 287-293.
[73]Yılmaz, Ü., Serdaroğlu, A., Gürkaş, E., Hirfanoğlu, T., & Cansu, A. (2013). Childhood paroxysmal nonepileptic events. *Epilepsy & Behavior*, *27*(1), 124-129.
[74]Akdemir, D., Uzun, Ö., Özsungur, B. P., & Topçu, M. (2013). Health-related quality of life in adolescents with psychogenic nonepileptic seizures. *Epilepsy & Behavior*, *29*(3), 516-520.
[75]Alessi, R., Vincentiis, S., Rzezak, P., & Valente, K. D. (2013). Semiology of psychogenic nonepileptic seizures: age-related differences. *Epilepsy & Behavior*, *27*(2), 292-295.
[76]Szabó, L., Siegler, Z., Zubek, L., Liptai, Z., Körhegyi, I., Bánsági, B., & Fogarasi, A. (2012). A detailed semiologic analysis of childhood psychogenic nonepileptic seizures. *Epilepsia*, *53*(3), 565-570.
[77]Kim, S. H., Kim, H., Lim, B. C., Chae, J. H., Kim, K. J., Hwang, Y. S., & Hwang, H. (2012). Paroxysmal nonepileptic events in pediatric patients confirmed by long-term video-EEG monitoring—Single tertiary center review of 143 patients. *Epilepsy & Behavior*, *24*(3), 336-340.
[78]Kutluay, E., Selwa, L., Minecan, D., Edwards, J., & Beydoun, A. (2010). Nonepileptic paroxysmal events in a pediatric population. *Epilepsy & Behavior*, *17*(2), 272-275.
[79]Hirfanoğlu, T., Serdaroğlu, A., Cansu, A., & Bilir, E. (2010). Outcome of Prolonged Video-EEG Monitoring in Children with Epilepsy.
[80]Salpekar, J. A., Plioplys, S., Siddarth, P., Bursch, B., Shaw, R. J., Asato, M. R., ... & Caplan, R. (2010). Pediatric psychogenic nonepileptic seizures: a study of assessment tools. *Epilepsy & Behavior*, *17*(1), 50-55.
[81]Verrotti, A., Agostinelli, S., Mohn, A., Manco, R., Coppola, G., Franzoni, E., ... & Curatolo, P. (2009). Clinical features of psychogenic non-epileptic seizures in prepubertal and pubertal patients with idiopathic epilepsy. *Neurological Sciences*, *30*(4), 319-323.
[82]Chinta, S. S., Malhi, P., Singhi, P., & Prabhakar, S. (2008). Clinical and psychosocial characteristics of children with nonepileptic seizures. *Annals of Indian Academy of Neurology*, *11*(3), 159.
[83]Kacinski, M., Leskiewicz, M., Jaworska-Feil, L., Zajac, A., Kubik, A., Budziszewska, B., & Lason, W. (2007). Pseudo-epileptic seizures in children are not associated with enhanced plasma level of allopregnanolone. *Pharmacological Reports*, *59*(6), 683.
[84]Patel, H., Scott, E., Dunn, D., & Garg, B. (2007). Nonepileptic seizures in children. *Epilepsia*, *48*(11), 2086-2092.
[85]Vincentiis, S., Valente, K. D., Thomé-Souza, S., Kuczinsky, E., Fiore, L. A., & Negrão, N. (2006). Risk factors for psychogenic nonepileptic seizures in children and adolescents with epilepsy. *Epilepsy & Behavior*, *8*(1), 294-298.
[86]Witgert, M. E., Wheless, J. W., & Breier, J. I. (2005). Frequency of panic symptoms in psychogenic nonepileptic seizures. *Epilepsy & Behavior*, *6*(2), 174-178.
[87]Bhatia, M. S., & Sapra, S. (2005). Pseudoseizures in children: a profile of 50 cases. *Clinical Pediatrics*, *44*(7), 617-621.
[88]Ahmed, M. A. S., Martinez, A., Mariam, S., & Whitehouse, W. (2004). Chvostek's sign and hypocalcaemia in children with seizures. *Seizure*, *13*(4), 217-222.
[89]Pakalnis, A., & Paolicchi, J. (2003). Frequency of secondary conversion symptoms in children with psychogenic nonepileptic seizures. *Epilepsy & Behavior*, *4*(6), 753-756.
[90]Kotagal, P., Costa, M., Wyllie, E., & Wolgamuth, B. (2002). Paroxysmal nonepileptic events in children and adolescents. *Pediatrics*, *110*(4), e46-e46.
[91]Gudmundsson, O., Prendergast, M., Foreman, D., & Cowley, S. (2001). Outcome of pseudoseizures in children and adolescents: a 6-year symptom survival analysis. *Developmental medicine and child neurology*, *43*(8), 547-551.

[92]Pakalnis, A., & Paolicchi, J. (2000). Psychogenic seizures after head injury in children. *Journal of child neurology*, *15*(2), 78-80.
[93]Irwin, K., Edwards, M., & Robinson, R. (2000). Psychogenic non-epileptic seizures: management and prognosis. *Archives of disease in childhood*, *82*(6), 474-478.
[94]Wyllie, E., Glazer, J. P., Benbadis, S., Kotagal, P., & Wolgamuth, B. (1999). Psychiatric features of children and adolescents with pseudoseizures. *Archives of pediatrics & adolescent medicine*, *153*(3), 244-248.
[95]Tamer, S. K. (1997). The pediatric non-epileptic seizure. *The Indian Journal of Pediatrics*, *64*(5), 671-676.
[96]Selbst, S. M., & Clancy, R. (1996). Pseudoseizures in the pediatric emergency department. *Pediatric emergency care*, *12*(3), 185-188.
[97]Kramer, U., Carmant, L., Riviello, J. J., Stauffer, A., Helmers, S. L., Mikati, M. A., & Holmes, G. L. (1995). Psychogenic seizures: video telemetry observations in 27 patients. *Pediatric neurology*, *12*(1), 39-41.
[98]Lancman, M. E., Asconapé, J. J., Graves, S., & Gibson, P. A. (1994). Psychogenic seizures in children: long-term analysis of 43 cases. *Journal of Child Neurology*, *9*(4), 404-407.
[99]Valdizan, J. R., Vergara, J. M., Rodriguez, J. P., Guallar, A., & Garcia, C. (1992). Nocturnal prolactin and growth hormone levels in children with complex partial and generalized tonic-clonic seizures. *Acta neurologica scandinavica*, *86*(2), 139-141.
[100]Wyllie, E., Friedman, D., Lüders, H., Morris, H., Rothner, D., & Turnbull, J. (1991). Outcome of psychogenic seizures in children and adolescents compared with adults. *Neurology*, *41*(5), 742-744.
[101]Wyllie, E., Friedman, D., Rothner, A. D., Luders, H., Dinner, D., Morris III, H., ... & Kotagal, P. (1990). Psychogenic seizures in children and adolescents: outcome after diagnosis by ictal video and electroencephalographic recording. *Pediatrics*, *85*(4), 480-484.
[102]Seneviratne, U., Reutens, D., & D'Souza, W. (2010). Stereotypy of psychogenic nonepileptic seizures: Insights from video-EEG monitoring. *Epilepsia*, *51*(7), 1159-1168.
[103]Kaufman J, Birmaher B, Brent D, et al. Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL): initial reliability and validity data. J Am Acad Child Adolesc Psychiatry 1997;36:980–988.
[104]Velani, H., & Gledhill, J. (2021). The effectiveness of psychological interventions for children and adolescents with non-epileptic seizures. *Seizure*, *93*, 20-31.
[105]Freedman, D. A., Terry, D., Enciso, L., Trott, K., Burch, M., & Albert, D. V. (2022). Brief Report: Psychogenic Nonepileptic Events in Pediatric Patients with Autism or Intellectual Disability. *Journal of Autism and Developmental Disorders*, 1-5.
[106]Kozlowska, K., Cruz, C., Davies, F., Brown, K. J., Palmer, D. M., McLean, L., ... & Williams, L. M. (2016). The utility (or not) of self-report instruments in family assessment for child and adolescent conversion disorders?. *Australian and New Zealand Journal of Family Therapy*, *37*(4), 480-499.
[107]McFarlane, F. A., Allcott-Watson, H., Hadji-Michael, M., McAllister, E., Stark, D., Reilly, C., ... & Heyman, I. (2019). Cognitive-behavioural treatment of functional neurological symptoms (conversion disorder) in children and adolescents: a case series. *European Journal of Paediatric Neurology*, *23*(2), 317-328.
[108]Goldstein, L. H., Robinson, E. J., Mellers, J. D., Stone, J., Carson, A., Reuber, M., ... & Samarasekera, S. (2020). Cognitive behavioural therapy for adults with dissociative seizures (CODES): a pragmatic, multicentre, randomised controlled trial. *The Lancet Psychiatry*, *7*(6), 491-505.
[109] Pick, S., Anderson, D. G., Asadi-Pooya, A. A., Aybek, S., Baslet, G., Bloem, B. R., ... & Nicholson, T. R. (2020). Outcome measurement in functional neurological disorder: a

systematic review and recommendations. *Journal of Neurology, Neurosurgery & Psychiatry*, *91*(6), 638-649.
[110] Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Leventhal, B. L., DiLavore, P. C., ... & Rutter, M. (2000). The Autism Diagnostic Observation Schedule—Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of autism and developmental disorders*, *30*(3), 205-223.
[111] Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: a revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of autism and developmental disorders*, *24*(5), 659-685.
[112] Gillberg, C., Gillberg, C., Råstam, M., & Wentz, E. (2001). The Asperger Syndrome (and high-functioning autism) Diagnostic Interview (ASDI): a preliminary study of a new structured clinical interview. *Autism*, *5*(1), 57-66.
[113] American Psychiatric Association. (2000). Diagnostic and statistical manual of mental disorders, text revision (DSM-IV-TR®).
[114] American Psychiatric Association. (1994). Diagnostic and Statistical Manual of Mental Disorders,(DSM-IV), Washington DC American Psychiatric Association, 1994. *Tradução portuguesa. Fernandes JC edt, Climepsi Editores, Lisboa*.
[115]Puig-Antich, J., & Chambers, W. (1978). The schedule for affective disorders and schizophrenia for school-age children (Kiddie-SADS). *New York: New York State Psychiatric Institute*.
[116] Hubsch, C., Baumann, C., Hingray, C., Gospodaru, N., Vignal, J. P., Vespignani, H., & Maillard, L. (2011). Clinical classification of psychogenic non-epileptic seizures based on video-EEG analysis and automatic clustering. *Journal of Neurology, Neurosurgery & Psychiatry*, *82*(9), 955-960.
[117] Betts, T. A. (1991). Pseudoseizures (non-epileptic attack disorder). *Women and epilepsy*, 243-258.

Figure 1: Search Process for studies focusing on PNES in children

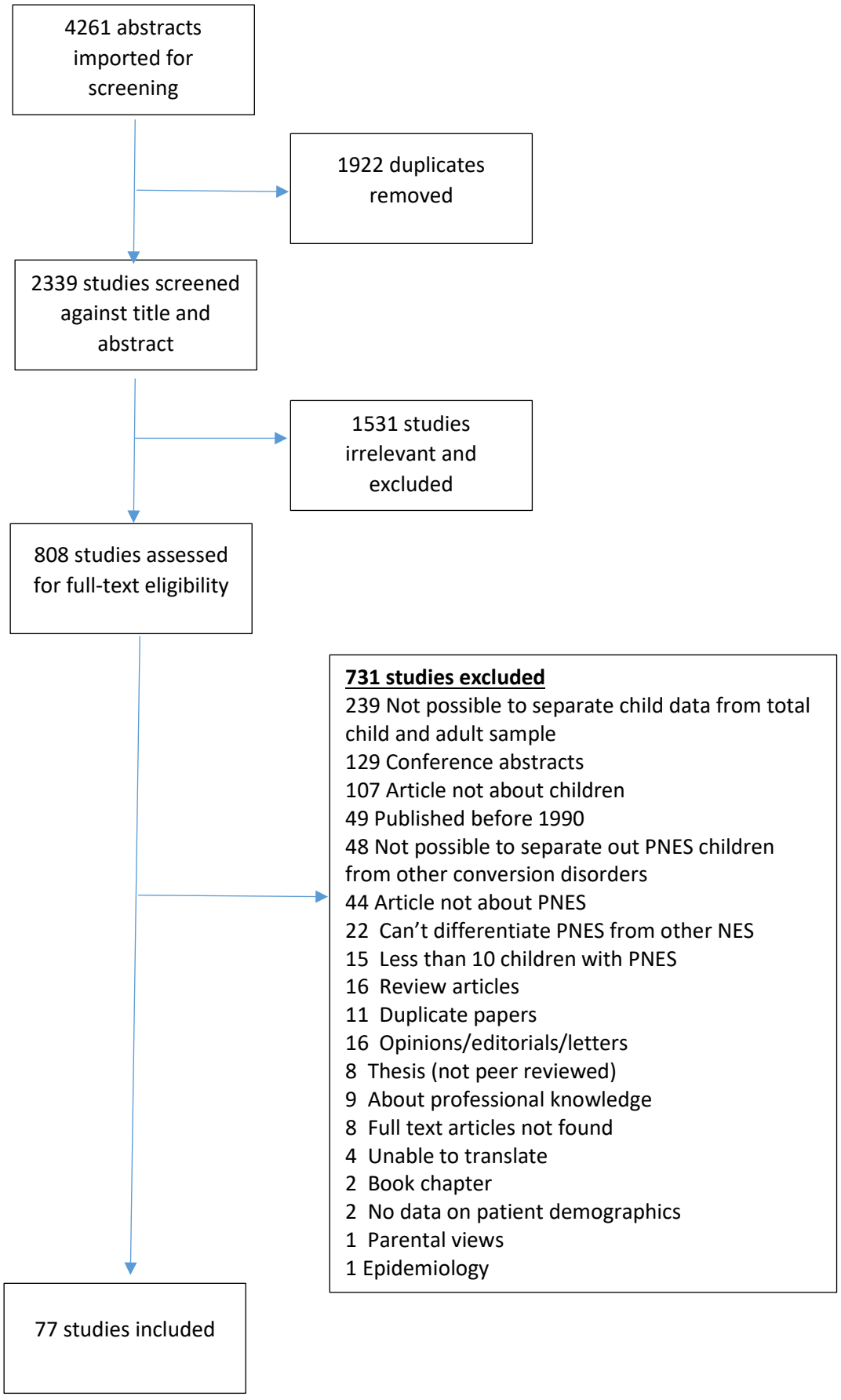

Table 1: Characteristics of studies on psychogenic non-epileptic seizures (PNES) in children 1st January 1990 to 13th September 2021

Author	Year	Retrospective or Prospective	Study Design	Location - Country	Location - Region	Ascertainment	Sample Size	Age range (Mean)	Gender m/f/o
Fredwall et al[27a]	2021	Prospective	Cross-Sectional	US	Ohio	Hospital	23	8-19 (14)	8/15
Thabit et al[28]	2021	Prospective	Cross-Sectional	Egypt	Sohag	Hospital	21	6-17 (8.90)	5/16
Fredwall et al[29b]	2021	Retrospective	Cross-Sectional	US	Ohio	Hospital	125	NR (NR)	30/94/1
Hansen et al[30a]	2021	Retrospective	Cross-Sectional	Demark	Whole Country	Population-based	384	5-17 (NR)	70/314
Hansen et al[3b]	2020	Retrospective	Cross-Sectional	Demark	Whole Country	Population-based	386	5-17 (NR)	64/322
Sawchuck et al[31b]	2020	Retrospective	Cross-sectional	Canada	Calgary	Hospital	33	10-17 (14.4)	10/23
Zhang et al[32]	2021	Retrospective	Cross-sectional	China	Beijing	Hospital	88	4-14 (NR)	54/34
Kaczmarek et al[33]	2020	Retrospective	Cross-Sectional	Poland	Poznan	Hospital	158	NR (14.40)	25/133
Masi et al[34]	2020	Prospective	Cross-Sectional	Italy	Pisa	Hospital	22	12-21 (15.71)	8/14
Fobian et al[35]	2020	Prospective	RCT	US	Alabama	Hospital	29	NR (15.1)	8/21
Flewelling et al[a36]	2020	Retrospective	Cross-sectional	US	Colorado	Hospital	19	9-17 (13.95)	2/17
Terry et al[37]	2020	Prospective	Cross-Sectional	US	Ohio	Hospital	101	NR (14.80)	26/75
Flewelling et al[38]	2020	Retrospective	Cross-sectional	US	Colorado	Hospital	37	8-18 (14.08)	5/32
Sawchuck et al[a39]	2020	Retrospective	Cross-sectional	Multinational	Multicentre	Hospital	178	4-18 (NR)	65/113

Myers et al[40]	2019	Prospective	Cross-sectional	US	Hackensack	Hospital	15	NR (14.3)	4/11
Asadi-Pooya et al[41]	2019	Retrospective	Cross-Sectional	Multinational	Multicentre	Hospital	51	8-16 (13.4)	19/32
Gowda et al[42]	2019	Prospective	Cross-Sectional	India	Karantaka	Hospital	37	5-18 (10.5)	26/11
Uzun et al[43]	2019	Prospective	Cross-sectional	Turkey	Ankara	Hospital	42	12-18 (14.80)	7/35
McWilliams et al[44]	2019	Prospective	Other	UK	London	Hospital	59	NR	22/37
Madanna et al[45]	2018	Prospective	Other	India	New Delhi	Hospital	80	6-16 (10.50)	45/35
Kandler et al[46]	2018	Prospective	Cross-sectional	UK	Multicentre	Hospital	44	NR	NR
Bursch et al[47]	2018	Prospective	Cross-sectional	US	Multicentre	Hospital	47	8-18 (14.60)	13/34
Luthy et al[49]	2018	Retrospective	Cross Sectional	US	Multicentre	Hospital	399	8-20 (NR)	110/289
Inaida et al[50]	2018	Retrospective	Other	Japan	National Database	Other	69	0-18	25/44
Kozlowska et al[51]	2018	Prospective	Other	Australia	Multicentre	Hospital	60	8-18 (13.45)	18/42
Kozlowska et al[52]	2018	Prospective	Other	Australia	Multicentre	Hospital	60	8-18 (13.45)	18/42
Kozlowska et al[53]	2017	Prospective	Other	Australia	Sydney	Hospital	60	8-17 (13.45)	18/42
Doss et al[54]	2017	Prospective	Case-control	US	Multicentre	Hospital	55	8-18 (14.80)	16/39
Mohamed et al[55]	2017	Prospective	Cross-sectional	Sudan	Khartoum	Population	15	6-14 (NR)	NR
Umesh et al[56]	2017	Prospective	Case-control	India	Kanke	Hospital	15	NR (17.20)	4/11

Valente et al[7]	2017	Prospective	Cross-sectional	Brazil	Sao Paolo	Hospital	53	7-17 (12.81)	21/32
Pliolpys et al[57]	2016	Prospective	Case-control	US	Multicentre	Hospital	55	8-18 (14.80)	16/39
McWilliams et al[9]	2016	Prospective	Cross-sectional	UK	London	Hospital	10	6-19 (14.80)	6/4
Narita et al[58]	2016	Prospective	Other	Japan	Unknown	Hospital	15	NR (9.3)	2/13
Park et al[59]	2015	Retrospective	Case-control	South Korea	Seoul	Hospital	33	NR	NR
Say et al[60]	2015	Retrospective	Case-control	Turkey	Samsun	Hospital	62	11-18 (14.19)	18/44
Rawat et al[61]	2015	Retrospective	Cross-sectional	India	South India	Hospital	34	8-16 (12)	NR
Yadav at al[62]	2015	Retrospective	Cross-sectional	US	Cleveland	Hospital	90	5-18 (14)	32/58
Citilcioğlu et al[63]	2015	Prospective	Case-control	Turkey	Adana	Hospital	50	NR (11)	18/32
Sawchuck et al[64]	2015	Retrospective	Cross-sectional	Canada	Calgary	Hospital	29	NR	7/22
Say et al[65]	2014	Prospective	Case-control	Turkey	Samsun	Hospital	34	11-18 (14.26)	11/23
Plioplys et al[66]	2014	Prospective	Case-control	US	Multicentre	Hospital	55	8-18 (14.80)	16/39
Aich et al[67]	2014	Retrospective	Case-Control	Nepal	Bhairahawa	Hospital	53	5-17 (12.20)	21/32
Wadwekar et al[68]	2014	Retrospective	Cross-sectional	India	Pondicherry	Hospital	23 of 54 were children	NR	NR
Yi et al[69]	2014	Retrospective	Cross-sectional	South-Korea	Seoul	Hospital	25	8-19 (13.82)	11/14

Li et al[70]	2014	Retrospective	Case-control	China	Chengdu	Hospital	11	13-17 (14.8)	2/9
Abdel Kader et al[71]	2014	Retrospective	Cross-sectional	Egypt	Cairo	Hospital	11	NR NR	7/4
Dhiman et al[72]	2013	Retrospective	Cross-sectional	India	Karantaka	Hospital	56	2-17 (12.30)	26/30
Yilmaz et al[73]	2013	Retrospective	Cross-sectional	Turkey	Ankara	Hospital	54	NR (11.35)	18/36
Akmedir et al[74]	2013	Prospective	Case-Control	Turkey	Hacettepe	Hospital	34	12-17 (15)	7/27
Alessi et al[75]	2013	Retrospective	Case-Control	Brazil	Sao Paulo	Hospital	42	6-17 (12)	22/20
Szabo et al[76]	2012	Retrospective	Cross-sectional	Hungary	Budapest	Hospital	27	8-18 (11.60)	6/21
Kim et al[77]	2012	Retrospective	Other	South Korea	Seoul	Hospital	15	3-19 (NR)	NR
Kutluay et al[78]	2010	Retrospective	Cross-sectional	US	Ann Arbour	Hospital	36	6-17 (13.5)	13/23
Hirfanoglu et al[79]	2010	Prospective	Cross-sectional	Turkey	Ankara	Hospital	31	NR	NR
Salpekar et al 2010[80]	2010	Prospective	Case-control	US	Multicentre	Hospital	24	10-17 (14)	10/14
Verrotti et al[81]	2009	Retrospective	Cross-sectional	Italy	Multicentre	Hospital	36	6-17 (NR)	10/26
Chinta et al[82]	2008	Prospective	Case-control	India	North India	Hospital	17	7-13 (10.70)	4/13
Kacinski et al[83]	2007	Prospective	Other	Poland	Krakow	Hospital	45	11-19	4/41
Patel et al[84]	2007	Retrospective	Cross-sectional	US	Indianapolis	Hospital	59	5-20 (13.14)	22/37
Vincentiis et al[85]	2006	Prospective	Cross-sectional	Brazil	Sao Paulo	Hospital	21	5-18 (13.1)	12/9

Witgert et al[86]	2005	Retrospective	Case-control	US	Houston	Hospital	18	13-18 (16.33)	4/14
Bhatia & Sapra[87]	2005	Prospective	Cross-Sectional	India	Delhi	Hospital	50	6-12 (8.50)	22/28
Ahmed et al[88]	2004	Prospective	Cross-Sectional	UK	Nottingham	Hospital	30	NR	NR
Pakalnis & Paolicchi[89]	2003	Retrospective	Cross-sectional	US	Columbus	Hospital	22	7-17 (13.50)	3/19
Kotogal et al[90]	2002	Retrospective	Cross-sectional	US	Cleveland	Hospital	62	5-18 (NR)	28/32
Gudmundsson et al[91]	2001	Retrospective	Cross-sectional	UK	Birmingham	Hospital	17	8-15 (12.90)	2/15
Pakalanis & Paolicchi[92]	2000	Retrospective	Cross-sectional	US	Columbus	Hospital	16	5-18 (10.50)	11/5
Irwin et al[93]	2000	Retrospective	Cross-sectional	UK	London	Hospital	35	6-18 (14.10)	11/24
Wyllie et al[94]	1999	Retrospective	Cross-sectional	US	Cleveland	Hospital	34	9-18 (14)	9/25
Tamer et al[95]	1997	Retrospective	Cross-Sectional	India	Bhilal	Hospital	22	5-18 (NR)	10/12
Selbst & Clancy[96]	1996	Retrospective	Cross-sectional	US	Philadelphia	Hospital	10	6-17 (12.4)	3/7
Kramer et al[97]	1995	Retrospective	Cross-sectional	US	Boston	Hospital	27	6-17 (12.6)	9/18
Lancman et al[98]	1994	Retrospective	Other	US	North Carolina	Hospital	43	NR	11/32
Valdizan et al[99]	1992	Prospective	Cross-Sectional	Spain	Zaragoza	Hospital	17	6-13 (9.27)	NR
Wyllie et al[100]	1991	Retrospective	Case-control	US	Cleveland	Hospital	18	8-18 (14.50)	6/12
Wyllie et al[101]	1990	Prospective	Cross-sectional	US	Cleveland	Hospital	21	8-18 (14.50)	6/15

NR= Not Reported, RCT= Randomized Control Trial, m/f/o = male/female/other

Table 2: Characteristics of studies of psychogenic non-epileptic seizures (PNES)DSM in children with respect to age of onset, diagnosis, number with epilepsy and definition of PNES.

Author	Year	Mean Age of onset	Mean Age of diagnosis	Delay in diagnosis	Number (%) with epilepsy	Controls without PNES	PNES Definition	PNES terminology	Epilepsy definition
Fredwall et al[27a]	2021	NR	NR	NR	7 (30%)	No	None/Other	Psychogenic Nonepileptic events	NR
Thabit et al[28]	2021	NR	NR	NR	0 (0%)	No	None/Other	PNES	NR
Fredwall et al[29]	2021	NR	NR	NR	23 (18%)	No	Le France et al.	PNES	NR
Hansen et al[30]	2021	NR	NR	NR	54 (14%)	Yes	ICD +Le France et al.	PNES	ICD
Hansen et al[3]	2021	NR	NR	NR	0 (%)	No	Other/Other	PNES	NA
Sawchuck et al[31]	2020	NR	NR	NR	55(14%)	No	ICD +Le France et al	PNES	ICD
Zhang et al[32]	2020	NR	NR	NR	10 (33%)	No	Le France et al.	PNES	NR
Kaczmarek et al[33]	2020	NR	NR	NR	0 (%)	Yes	ICD	PNES	NR
Masi et al[34]	2020	NR	NR	NR	7(32%)	Yes	Le France et al.	PNES	ILAE
Fobian et al[35]	2020	NR	NR	0.75	3 (10%)	No	None/Other	PNES	NR
Flewelling et al[36]	2020	NR	NR	NR	9(47%)	No	None/Other	PNES	NR
Terry et al[37]	2020	14.20	14.80	0.60	22 (22%)	No	Le France et al.	Psychogenic non epileptic events	NR
Flewelling et al[38]	2020	NR	NR	NR	17 (46%)	No	None/Other	PNES	NR
Sawchuck et al[39]	2020	NR	NR	NR	0 (0%)	No	None/Other	PNES	NA

Myers et al[40]	2019	NR	NR	NR	0 (0%)	Yes	None/Other	PNES	NA
Asadi-Pooya et al[41]	2019	12.30	13.40	1.00	13(25%)	No	None/Other	PNES	NR
Gowda et al[42]	2019	NR	NR	NR	0(0%)	No	None/Other	Psychogenic seizures	NR
Uzun et al[43]	2019	NR	NR	NR	0 (0%)	Yes	None/Other	PNES	NA
McWilliams et al[44]	2019	12.50	NR	NR	22 (38%)	No	None/Other	NEAD	NR
Madanna et al[45]	2018	NR	NR	NR	0(0%)	No	None/Other	PNES	NA
Kandler et al[46]	2018	NR	NR	NR	NR	No	None/Other	NEAD	Clinical with EEG change but not clearly stated
Bursch et al[47]	2018	13.64	NR	NR	13 (28%)	Yes	DSM	PNES	NR
Luthy et al[49]	2018	NR	NR	NR	0	Yes	ICD	PNES	ILAE
Inaida et al[50]	2018	NR	NR	NR	31 (45%)	No	ICD	PNES	NR
Kozlowska et al[51]	2018	NR	NR	NR	7 (12%)	Yes	None/Other	PNES	NR
Kozlowska et al[52]	2017	NR	NR	NR	7 (12%)	No	None/Other	PNES	NR
Kozlowska et al[53]	2017	NR	NR	NR	7 (12%)	Yes	None/Other	PNES	NA
Doss et al[54]	2017	14.30	NR	NR	16(29%)	Yes	DSM	PNES	NR
Mohamed et al[55]	2017	NR	NR	NR	0 (0%)	No	None/Other	PNES	NA
Umesh et al[56]	2017	NR	NR	NR	0 (0%)	Yes	ICD	PNES	NR
Valente et al[7]	2017	11.17	NR	1.48	21 (40%)	No	None/Other	PNES	Clinical Opinion
Pliolpys et al[57]	2016	14.30	NR	NR	16 (29%)	Yes	DSM	PNES	NR
McWilliams et al[9]	2016	NR	NR	NR	3 (30%)	No	None/Other	NES	NR

Narita et al[58]	2016	NR	NR	NR	6 (40%)	No	None/Other	PNES	NR
Park et al[59]	2015	NR	NR	NR	0	Yes	None/Other	PNES	Other
Say et al[60]	2015	NR	NR	0.85	24 (40%)	No	None/Other	PNES	NR
Rawat et al[61]	2015	9.00	12.00	0.83	8 (24%)	No	DSM	PNES	NR
Yadav at al[62]	2015	NR	NR	0.25	19 (21%)	No	None/Other	PNES	NR
Citilcioğlu et al[63]	2015	NR	NR	NR	0(0%)	Yes	None/Other	NES	NR
Sawchuck et al[64]	2015	NR	NR	NR	7 (24%)	No	DSM	PNES	NR
Say et al[65]	2014	NR	NR	NR	0	Yes	None/Other	PNES	2 unprovoked epileptic seizures
Plioplys et al[66]	2014	14.30	NR	NR	16(29%)	Yes	DSM	PNES	NR
Aich et al[67]	2014	NR	NR	NR	0(0%)	Yes	None/Other	Pseudoseizures	NR
Wadwekar et al[68]	2014	NR	NR	NR	6(9%)	No	None/Other	PNES	NR
Yi et al[69]	2014	NR	13.71	0.80	8 (32%)	No	None/Other	PNES	NR
Li et al[70]	2014	NR	NR	NR	0 (0%)	Yes	None/Other	PNES	NA
Abdel Kader et al[71]	2014	NR	NR	NR	0 (0%)	Yes	None/Other	PNES	NA
Dhiman et al[72]	2013	8.90	11.90	3.20	9 (16%)	No	None/Other	PNES	NR
Yilmaz et al[73]	2013	13.79	NR	2.49	6 (11%)	Yes	None/Other	PNES	NR
Akmedir et al[74]	2013	13.60	NR	1.28	0(0%)	Yes	None/Other	PNES	NA
Alessi et al7[5]	2013	NR	NR	NR	17	Yes	None/Other	PNES	Previous or current history of seizures based on EEG
Szabo et al[76]	2012	11.60	NR	NR	9 (33%)	No	None/Other	PNES	NR
Kim et al[77]	2012	NR	NR	NR	4 (27%)	Yes	None/Other	PNES	NR
Kutluay et al[78]	2010	NR	NR	NR	9 (25%)	Yes	None/Other	PNES	NR

Hirfanoglu et al[79]	2010	NR	NR	NR	0(0)%	No	None/Other	Pseudoseizures	NA
Salpekar et al 2010[80]	2010	NR	NR	NR	2 (8.3%)	Yes	None/Other	PNES	NR
Verrotti et al[81]	2009	NR	NR	NR	36(100%)	No	None/Other	PNES	ILAE
Chinta et al[82]	2008	NR	NR	NR	0 (0%)	Yes	DSM	NES	ILAE
Kacinski et al[83]	2007	NR	R	NR	0(0%)	No	None/Other	Other	NA
Patel et al[84]	2007	12.90	13.40	0.54	26 (38%)	No	None/Other	NES	NR
Vincentiis et al[85]	2006	NR	NR	NR	19 (91%)	No	None/Other	PNES	ILAE
Witgert et al[86]	2005	NR	NR	NR	0 (0%)	Yes	None/Other	PNES	NA
Bhatia & Sapra[87]	2005	NR	NR	NR	0	No	ICD	Pseudoseizures	NA
Ahmed et al[88]	2004	NR	NR	NR	0	Yes	None/Other	Pseudoseizures	ILAE
Pakalnis & Paolicchi[89]	2003	NR	NR	NR	5(23%)	No	None/Other	PNES	NR
Kotogal et al[90]	2002	NR	NR	NR	11 (18%)	Yes	None/Other	Psychogenic Seizures	NR
Gudmundsson et al[91]	2001	11.10	12.90	0.90	0 (0%)	No	None/Other	Pseudoseizures	NA
Pakalanis & Paolicchi[92]	2000	NR	NR	NR	3 (19%)	No	None/Other	Psychogenic Seizures	NR
Irwin et al[93]	2000	NR	NR	NR	11 (31%)	No	None/Other	PNES	NR
Wyllie et al[94]	1999	NR	NR	0.95	4 (12%)	No	DSM	Pseudoseizures	NR
Tamer et al[95]	1997	NR	NR	NR	9(41%)	No	None/Other	NES	NR
Selbst & Clancy[96]	1996	NR	NR	NR	0	No	None/Other	Pseudoseizures	NR
Kramer et al[97]	1995	12.00	NR	1.10	4 (15%)	No	None/Other	Psychogenic Seizures	NR

Lancman et al[98]	1994	12.40	NR	3.50	0 (0%)	No	None/Other	Psychogenic Seizures	NA
Valdizan et al[99]	1992	NR	NR	NR	0	Yes	None/Other	Pseudoseizures	NR
Wyllie et al[100]	1991	NR	NR	NR	0 (0%)	Yes	None/Other	PNES	NA
Wyllie et al[101]	1990	13.80	NR	0.57	0 (0%)	No	None/Other	Psychogenic Seizures	NA

DSM – Diagnostic and Statistical Manual of Mental Disorders, ICD= International Classification of Disease, ILAE= International League Against Epilepsy NA=Not applicable = as patients with epilepsy were not included, NEAD = non-epileptic attack disorder, NR=Not reported.

Table 3a: Level of agreement of round 1 and round 2 of the Delphi survey –Assessment of suspected psychogenic non-epileptic seizures (PNES) in children

Recommendation - Assessment of suspected PNES in children	Round 1	Round 2
R1: The word psychogenic is useful when describing children who have seizure like events which are thought to be functional in nature. R2: The term psychogenic can be perceived negatively or be stigmatizing and should only be used with young people and their family/caregivers if it is felt to be helpful to explain the psychological nature of these events.	**50%**	**69%**
The process of assessment of young people with suspected PNES should include taking a comprehensive description of the episodes/events – (e.g. What does the episode look like? When did/does it happen? Who is present? Where does it happen?)	**100%**	**NA**
The process of assessment of young people with suspected PNES should include taking a comprehensive medical/developmental history (e.g., asking about other medical conditions, learning/behavior, schooling).	**100%**	**NA**
It is important to ask about potential stressors in the young person's life (e.g., school/academic difficulties, family difficulties, bullying, previous physical/sexual abuse, trauma).	**96%**	**NA**
With suspected PNES in young people, it is important to ask about other symptoms of conversion disorder/functional neurological disorder (e.g., pain, sensory or motor).	**96%**	**NA**
Parent, self or school report of events are useful in determining if events are psychogenic in nature and can contribute to a 'possible' diagnosis of PNES by a clinician experienced in diagnosis of seizure disorders.	**89%**	**NA**
Parent home/school video-recording of events is very important in considering whether events are psychogenic in nature and can contribute to a 'probable' diagnosis of PNES by a clinician experienced in diagnosis of seizure disorders.	**89%**	**NA**
If available, Video-EEG should be used with all young people with suspected PNES and if no epileptic activity is detected during a typical event, then a 'clinically established' and 'documented' PNES diagnosis can be made by a clinician experienced in diagnosis of seizure disorders.	**85%**	**NA**

R1: The use of standard techniques (e.g., sleep deprivation, hyperventilation, photic stimulation) is appropriate in an attempt to elicit PNES in children R2: The use of standard techniques (e.g., sleep deprivation, hyperventilation, photic stimulation) is appropriate in the assessment of suspected PNES in children to help differentiate between epileptic and nonepileptic events.	**61%**	**81%**
R1: The use of invasive provocation techniques (e.g., saline injection) or deceit should not be employed to elicit PNES in young people. R2: The use of invasive provocation techniques (e.g., saline injection) or deceit should not be employed in the assessment of PNES in young people	**71%**	**100%**

Table 3b: Level of agreement of round 1 and round 2 of the Delphi survey –Diagnosis and management of PNES in children

Recommendation - Assessment of suspected PNES in children	**Round 1**	**Round 2**
The involvement of both a pediatric neurologist/epileptologist and psychologist /psychiatrist is necessary when PNES is first diagnosed to coordinate management and follow-up.	**89%**	**NA**
It should be made clear to the young person and their family/caregivers that events are not epileptic in nature and that anti-seizure medications are not appropriate treatment.* (*Unless child also has epilepsy in which case medications would still be appropriate for the epileptic seizures but not the PNES)	**92%**	**NA**
R1: The child (if developmentally and age appropriate) and their parents should be informed of the diagnosis of PNES separately.	**36%**	**NA**
In medical records/reports it should always be made clear that PNES refer to events of a psychogenic/functional (and not physiologic) nature that are part of the broader classification of functional neurological disorder/conversion disorder	**89%**	**NA**
A comprehensive plan (written document) should be developed in collaboration with the child and family to inform all relevant health and educational professionals in the child's network.	**82%**	**NA**
A pediatric neurologist (or other professional with expertise in epilepsy) should remain involved for a period of time after the diagnosis of PNES to manage withdrawal of anti-seizure	**89%**	**NA**

medications, ensure acceptance of diagnosis and avoid further inappropriate investigations.		
A comprehensive management plan for the events at home, school and other relevant locations with clear indications on what supporting adults should do should be developed and agreed upon by all relevant stakeholders.	**96%**	**NA**
In the case of young people with both PNES and epileptic seizures, there is a need for the young person, their families/caregivers and supporting educational and health professionals to be made aware of manifestation of both epileptic and non-epileptic events. Management plans for both should be available for all children.	**93%**	**NA**
Young people should always be given developmentally appropriate visual/written information about the nature and possible causes of PNES and possible management approaches.	**86%**	**NA**
Parents/caregivers should always be given appropriate written/visual information about the nature, possible causes and possible management approaches.	**86%**	**NA**
The decision on treatment modality for PNES in children should take into account the child's age, cognitive ability and family factors. For younger children there may need to be a focus on behavioral approaches and skill teaching. For older children and adolescents, cognitive behavioral therapy may be useful**.**	**93%**	**NA**
R1: Family therapy/counselling should be offered to all families of children with PNES. R2: When considering treatment for children with PNES it is important to consider that the family may need psychological support (e.g., psychoeducation, counselling) and this should be made available, where appropriate	**68%**	**100%**

Table 3c: Level of Agreement of Round 1 and Round 2 of the Delphi survey –Assessment and management of psychopathology in children with PNES

Recommendation - Assessment of suspected PNES in children	**Round 1**	**Round 2**
All young people with confirmed PNES should be screened for mental health (e.g., depression, anxiety, trauma) and neurodevelopmental (e.g., ADHD, autism spectrum disorder) difficulties.	**93%**	**NA**
R1: All young people with confirmed PNES should be assessed for learning/cognitive difficulties.	68%	**92%**

R2: It is recommended that young people with confirmed PNES be assessed for learning/cognitive difficulties if it is thought that these difficulties are contributing to the child's PNES or other mental health problems.		
Young people with PNES who have confirmed mental health or behavioral difficulties should access evidence-based treatments/supports for depression, anxiety ADHD, etc.	93%	**NA**

R1= Round 1 R2= Round 2

Table 4: Assessment methods for psychogenic non-epileptic seizures (PNES) in children

Author	Year	Primary assessment method	Semiology reported	Invasive provocation methods	Other* assessment methods employed
Fredwall et al[27a]	2021	Video EEG	Yes	NA	No
Thabit et al[28]	2021	Video EEG	No	NA	No
Fredwall et al29[b]	2021	Video EEG	Yes	NA	No
Hansen et al[30a]	2021	Video EEG	Yes	NA	No
Hansen et al[3b]	2021	Video EEG	Yes	NA	No
Sawchuck et al[31b]	2020	Video EEG	No	NA	No
Zhang et al[32]	2020	Video EEG	No	NA	No
Kaczmarek et al[33]	2020	Video EEG	No	After hyperventilation and photic stimulation, the patient was informed that a swab with a "medicine" would be applied to their left forearm and that the substance on it could trigger a seizure episode or other sensations". Then a cotton swab (approximate dimensions 5 cm × 5 cm) with water was applied to the left forearm, and the technician asked the patient to report all sensations. The trial ended with the removal of a cotton swab from the forearm.	See provocation methods: Sensitivity of placebo test for the diagnosis of PNES was 81.1%, specificity 79.8%, positive predictive value 89.6% and negative predictive value 66.3%
Masi et al[34]	2020	Video EEG	No	NA	NA
Fobian et al[35]	2020	Video EEG	No	NA	NA

Flewelling et al[a36]	2020	Video EEG	No	NA	NA
Terry et al[37]	2020	Video EEG	Yes	NA	NA
Flewelling et al[b38]	2020	Video EEG	No	NA	NA
Sawchuck et al[a39]	2020	Video EEG	Yes	NA	NA
Myers et al[40]	2019	Video EEG	No	NA	NA
Asadi-Pooya et al[41]	2019	Video EEG	No	NA	NA
Gowda et al[42]	2019	Video EEG	No	NA	NA
Uzun et al[43]	2019	NR	No	NA	NA
McWilliams et al[44]	2019	Video EEG	No	NA	NA
Madanna et al[45]	2018	Video EEG	Yes	In case of failure of spontaneous of event within one hour of vEEG recording, induction protocols were used in a sequential manner; verbal suggestion followed by placement of a tuning fork followed by body part compression (with hand to induce the event without eliciting pain or discomfort).	Yes - Induction: see provocation methods
Kandler et al[46]	2018	Video EEG	No	NA	NA
Bursch et al[47]	2018	Video EEG	No	NA	NA
Luthy et al[49]	2018	Video EEG	No	NA	NA
Inaida et al[50]	2018	Clinical Judgement	No	NA	NA

Kozlowska et al[51]	2018	Video EEG	No	NA	NA
Kozlowska et al[52]	2017	Video EEG	Yes	NA	NA
Kozlowska et al[53]	2017	Video EEG	No	NA	NA
Doss et al[54]	2017	Video EEG	No	NA	NA
Mohamed et al[55]	2017	NR	No	NA	NA
Umesh et al[56]	2017	Normal EEG	No	NA	NA
Valente et al[7]	2017	Video EEG	No	NA	NA
Pliolpys et al[57]	2016	Video EEG	No	NA	NA
McWilliams et al[9]	2016	Video EEG	No	NA	NA
Narita et al[58]	2016	Unknown	No	NA	NA
Park et al[59]	2015	Video EEG	No	For the patients who were suspected of having psychogenic nonepileptic seizures, when typical episodes were not observed, an attempt was made to induce the event by injecting 1 to 2 ml of saline intravenously after obtaining parental consent	NA
Say et al[60]	2015	Video EEG	Yes	NA	NA
Rawat et al[61]	2015	Video EEG	No	NA	NA
Yadav at al[62]	2015	Video EEG	Yes	NA	NA
Citilcioğlu et al[63]	2015	Normal EEG	No	NA	Serum prolactin levels in the differential diagnosis of

					epileptic and nonepileptic seizures.
Sawchuck et al[64]	2015	Video EEG	No	NA	NA
Say et al[65]	2014	Video EEG	No	NA	NA
Plioplys et al[66]	2014	Video EEG	No	NA	NA
Aich et al[67]	2014	Not described	No	NA	NA
Wadwekar et al[68]	2014	Video EEG	Yes	NA	NA
Yi et al[69]	2014	Video EEG	Yes	Patient was told that an intravenous drug that will induce a seizure will be injected. Saline solution was intravenously injected a millilitre at a time to a maximum of 10 mls. We then asked the patient if they felt anything. After observation of the spell, we explained that the action of the solution was over, to stop the event. After the test, we presented the family with the video to ensure the similarity of the recorded event with typical ones	NA
Li et al[70]	2014	Video EEG	No	NA	NA
Abdel Kader et al[71]	2014	Video EEG	No	NA	NA
Dhiman et al[72]	2013	Video EEG	No	NA	NA
Yilmaz et al[73]	2013	Video EEG	Yes	In patients suspected of having psychogenic seizures, when typical episodes were not observed, an attempt was made to induce the event by verbal suggestion, hyperventilation, and/or the injection of 1- to 2-ml saline intravenously after obtaining parental consent	NA

Akmedir et al[74]	2013	Video EEG	No	NA	NA
Alessi et al7[5]	2013	Video EEG	Yes	NA	NA
Szabo et al[76]	2012	Video EEG	Yes	NA	NA
Kim et al[77]	2012	Video EEG	Yes	NA	NA
Kutluay et al[78]	2010	Video EEG	No	NA	NA
Hirfanoglu et al[79]	2010	Video EEG	No	NA	NA
Salpekar et al 2010[80]	2010	Video EEG	No	NA	NA
Verrotti et al[81]	2009	Video EEG	Yes	NA	NA
Chinta et al[82]	2008	Video EEG	Yes	NA	NA
Kacinski et al[83]	2007	Video EEG	No	Intravenous saline Injection	Yes
Patel et al[84]	2007	Video EEG	Yes	Hyperventilation, photic stimulation, suggestion, and rarely application of an alcohol patch, alone or in combination.	No
Vincentiis et al[85]	2006	Video EEG	No	NA	NA
Witgert et al[86]	2005	Video EEG	No	NA	NA
Bhatia & Sapra[87]	2005	Video EEG	No	NA	NA
Ahmed et al[88]	2004	NR	No	NA	NA
Pakalnis & Paolicchi[89]	2003	Video EEG	No	NA	NA

Kotogal et al[90]	2002	Video EEG	Yes	NA	NA
Gudmundsson et al[91]	2001	Normal EEG	Yes	NA	NA
Pakalanis & Paolicchi[92]	2000	Video EEG	No	NA	NA
Irwin et al[93]	2000	Video EEG	Yes	NA	NA
Wyllie et al[94]	1999	Video EEG	No	Saline injection in 3 patients – but this method was subsequently abandoned.	NA
Tamer et al[95]	1997	Clinical Judgement	No	Induction by provocation and suggestion was attempted in some cases by injecting 10 ml of normal saline slowly during EEG record to observe simultaneous clinical and EEG abnormality. Provocation attempted by i.v. saline while doing EEG was possible in 7(31.8%) older children.	NA
Selbst & Clancy[96]	1996	Clinical Judgment	No	NA	NA
Kramer et al[97]	1995	Video EEG	Yes	When a typical event was observed on the first day of recording, PNES was induced by intravenous injection of normal saline (1 ml).	Yes
Lancman et al[98]	1994	Video EEG	Yes	NA	NA
Valdizan et al[99]	1992	NR	No	NA	Measurement of nocturnal prolactin - The results obtained show a clear difference between epileptic patients and the PNES group, due to an increase in mean prolactin values in epileptic patients, both in delta 1 and delta 2 of sleep.

Wyllie et al[100]	1991	Video EEG	No	NA	NA
Wyllie et al[101]	1990	Video EEG	No	If patients did not have a spontaneous episode during video-EEG recording, then the electroencephalographer attempted to induce a spell with suggestion and intravenous saline injection. Fifteen patients (71%) had their recorded seizures spontaneously, and six (29%) had seizures in response to suggestion and intravenous saline injection.	No

NA= Not Applicable, NR= Not Reported, *i.e., non-EEG methods

Table 5 Management of psychogenic non-epileptic seizures (PNES) in children

Author	Year	Sample Size	Description of Management/Intervention
Fredwall et al[27]	2021	23	• The MDT clinic team consists of epilepsy and psychology clinicians, a social worker, a nurse, and administrative support staff. Patient appointments in this clinic are joint visits that serve as a bridge between neurology and psychology. During the visit, first an epilepsy practitioner confirms the diagnosis of PNEE; then the psychology provider assesses psychological risk factors, provides an action plan to address PNEE, as well as makes a recommendation for counselling. The team social worker provides support to families facing barriers to care, including travel to clinic and other needs. The team nurse conducts follow-up phone calls at one-, three-, and twelve-months following the clinic visit. During Covid-19 pandemic clinics were provided via telemedicine.
Fredwell et al[29]	2021	125	• Multidisciplinary pediatric PNES clinic. Collaboration by neurology and psychology clinicians, social workers, and nurses supported patients in accessing needed health care and facilitated follow-up. This model resulted in most patients accepting the diagnosis of PNES and linking with counselling services.
Flewelling et al[38]	2020	37	• Upon a confirmed diagnosis of PNES through v-EEG, patients are referred to, a multidisciplinary clinic that provides brief treatment of PNES in youth. Patients and their families met with a nurse practitioner, epileptologist, neuropsychologist, and clinical psychologist who provided psychoeducation and appropriate recommendations.
Flewelling et al[36]	2020	19	• A multidisciplinary clinic that meets once monthly and provides brief treatment to youth with PNES and intractable epilepsy. At the initial visit, patients with PNES and their families were provided with a thorough explanation of the condition and met with a nurse practitioner, epileptologist, neuropsychologist, and clinical psychologist. These professionals established the patient's existing knowledge of PNES, presented v-EEG data, discussed psychological issues underlying the condition, examined the relationship between neuropsychological functioning and school-based episodes, explored initial reactions to the diagnosis, and made specific recommendations, including medication management, referrals to outpatient mental health, development of response plans for behavioral management of seizures, and guidance on reintegrating the patient into school and extracurricular activities. Patients were scheduled for follow-up appointments, occurring on average six months after the initial visit. During the follow-up appointment, a multidisciplinary team of epilepsy specialists reassessed the families

			understanding of the diagnosis, assessed barriers to following through with treatment, reinforced the need for psychological interventions, and supported patients and families in finding appropriate resources within the community.
Terry et al[37]	2020		• A clinic in which patients are seen by both an epilepsy provider and a psychologist. The purpose of this joint visit is to provide another opinion regarding the diagnosis and to serve as a bridge from neurology to mental health services. Patients are typically seen once in a joint appointment, then are transitioned to Behavioral Health for ongoing treatment and management. As part of ongoing quality improvement efforts for the clinic, a nurse, and social worker were added to the team to help with care coordination for patients seen in the clinic and to facilitate follow-up. The clinic tries to see patients expeditiously following a diagnosis of to solidify the diagnosis, provide education, and answer questions to help the family accept the diagnosis, and quickly move patients toward treatment and recovery.
Fobian et al[35]	2020	29	• Participants were randomized to receive either eight sessions of Retraining and Control Therapy (ReACT) or supportive therapy, and participants completed follow-up visits at 7- and 60-days posttreatment. ReACT aimed to retrain classically conditioned, involuntary PNES by targeting catastrophic symptom expectations and a low sense of control over symptoms using principles of habit reversal. Supportive therapy was based on the assumption that relief from stress or problems can be achieved by discussion with a therapist. • The intervention includes four steps (1) a clear etiological description based on the Integrated Etiological Summary Model (2) an individually tailored patient plan to retrain physical symptoms which challenges catastrophic symptom expectations and teaches patients to engage in behaviors incompatible with PNES similarly to habit reversal, an evidence-based behavioral treatment for retraining tics (3) a family plan to react to PNES in which they monitor the patient for safety but otherwise allow the patient to follow their plan to independently control the episode and a plan to return to school and social activities.
McWilliams et al[44]	2019	60*	• Intervention always involved child, family and school, and the psychological therapy employed a cognitive-behavioural framework tailored to the needs of a young person with AS.
Kozlowska et al[51]	2018	60	• Diagnostic formulation guided treatment approach which included o Individual therapy to manage PNES o Attend hospital school o Physiotherapy programme

			○ Outpatient treatment - both individual and family therapy and liaison with community mental health to address stressors • Diagnostic formulations were used to inform both the explanations about PNES that were given to them and their families and the clinical interventions that were used to help patients gain control over PNES. • Six different approaches ○ lay explanation and treatment interventions for dissociative PNES ○ lay explanation and treatment interventions for dissociative PNES triggered by hyperventilation ○ lay explanation and treatment interventions for innate defence responses presenting as PNES ○ lay explanation and treatment interventions for PNES associated with syncope triggered by vocal cord adduction in the context of distress ○ lay explanation and treatment interventions for non-epileptic seizures associated with syncope triggered by activation of the Valsalva manoeuvre in the context of distress ○ Lay explanation and treatment interventions for non-epileptic seizures associated with syncope triggered by reflex activation of the vagus nerve • All patients enrolled in the Mind–Body Programme engage in daily individual therapy to learn how to manage their PNES, attend the hospital school to commence reintegration back to school and complete a physiotherapy exercise programme to increase their body's capacity to manage changes in body state and to increase their physical resilience. The standard inpatient programme for children/ adolescents with PNES runs over a 2-week period. • Admissions are typically followed by outpatient treatment – both individual and family based – with community-based mental health services to address stressors that reside within the family and school systems and that function to trigger or perpetuate the patient's symptoms. The Psychological Medicine team continues, as needed, to support clinicians working in community-based services via telephone contact.

			• Of 53 patients with PNES who participated in the MyCalmBeat evaluation, 41 (77%) were able to utilize the biofeedback tool. • A total of 12 patients with PNES were unable to utilize MyCalmBeat because they were unable to decrease the initial starting respiratory rates of 20 breaths/min
McWilliams et al[9]	2016	10	• At the time of the study, some patients had been newly diagnosed with NES. Others were awaiting, were partway through, or had completed a course of cognitive behavior therapy-informed treatment.
Sawchuck et al 2015[64]	2015	29	• Treatment consisted of education around diagnosis and of individual psychological treatment, which, in most cases, was cognitive behavioral therapy up to 14 sessions. • Additional mental health services including psychiatric medication, family therapy, and admission to day or inpatient treatment were also utilized in a smaller number of cases.
Yi et al[69]	2014	25	• All patients were recommended to visit our psychiatrists after discussion about the diagnosis of PNES • Treatment plans varied depending on the results of the psychological consultation; psychotherapy in 9 patients, and a combination of psychotherapy and psychopharmacological therapy in 13 patients. The three remaining patients regularly visited our psychiatric clinic to assess their clinical status without treatment.
Rawat et al[61]	2015	34	• Psychosocial interventions included working with the family and child. Reassurance along with acknowledging of the symptoms and educating the parents with regard to the nature of the illness and ways of handling the paroxysmal attacks was the most common psychosocial intervention done in these children (34/34, 100%). • Individual therapy, involving cognitive behavioral therapy (CBT)/psychotherapy was used in 58.8% (20/34). • Psychotropic medications were used in 18 children (18/34, 52.9%). The most common medication class used was selective serotonin reuptake inhibitors (SSRI) (14/34; 41.2%), fluoxetine (n = 5/34; 14.7%), escitalopram (n = 5/34; 14.7%), sertraline (n = 3/34; 8.8%) and fluvoxamine (n = 1/34, 2.9%). Clonazepam was the most common non-SSRI drug prescribed (7/34, 20.6%). Combined intervention involving both pharmacological and psychosocial interventions and was used in 18 children (18/34 52.9%)

Chinta et al[82]	2008	17	• The treatment and management of these generally followed the approach highlighted by previous studies. This involved (i) shifting the focus of the parents from an organic to a psychosocial explanation of the symptoms; (ii) encouraging the child and parents to resume normal activities; (iii) ignoring or discouraging sick role behavior; and (iv) using problem solving coping techniques to tackle the child's difficulties, and (v) family counseling for enhancing parental competence to tackle problems and resolving family crises.
Vincentiis et al[85]	2006	21	• Once the diagnosis of PNES was established, all patients and their families were informed and referred for treatment. In 10 (47.6%) patients, psychoactive drug therapy was initiated, and in 3 (14.35%), dose adjustment of previously used drugs was needed. Reduction of AEDs was possible in 6 patients (28.6%).
Bathia & Sapra[87]	2005	50	• The patient were put on appropriate drug treatment (anxiolytic and/or antidepressants) and/or psychotherapy (explanation, reassurance, suggestion, confrontation and discussion of the problems associated with origin of pseudoseizure) for 3 months and followed up every 2 weeks for 3 months to assess the improvement.
Gudmundsson et al[91]	2001	17	• The treatment programme given was distraction by rehabilitation, which comprises milieu therapy and attendance at the hospital school together with anticonvulsant withdrawal. The aim of milieu therapy is to allow the child to escape with honour from sick role behaviour (Dubowitz and Hersov, 1976). The children are encouraged to function as normally as possible and engage in different activities to distract them from preoccupation with their predicament. If patients have seizures, fuss is avoided beyond making sure they are safe. They are not comforted. As soon as they have recovered, the children are encouraged to continue with the task that they were engaged upon when the seizure occurred. Parents are seen regularly for support and to empower them to cope with seizures appropriately outside the hospital. Individual work by nursing staff focuses on coping mechanisms, relaxation, anxiety and stress management as well as widening the child's support system of family, friends, and relatives. The children attend the hospital school all through their stay and continuous close liaison makes it possible to apply the same therapeutic principles there. The child's own school is visited to explain the condition and seizure management, to obtain previous academic records, and for continuity of course work on return from hospital. The rate of reintegration back to the child's own school is tailored individually. In our experience, gradual reintegration is likely to be more successful.

Irwin et al[93]	2000	35	• Management in most cases included early referral to a child psychologist. • All children with a primary diagnosis of epilepsy were also reviewed regularly by a paediatric neurologist. • Once the diagnosis of PNES was clear in the group without epilepsy, and had been accepted by the patient and family, the patient was discharged from the care of the neurologist to avoid the appearance of inconsistency. • In some cases, the diagnosis and management required admission to hospital, for example in all child relationship. Two of the eight cases with a history of abuse or violence were referred for pro longed management by a child psychiatrist. • Three of the 35 patients required a three month stay in a psychiatric unit
Tamer et al[95]	1997	22	• 10 children: Hospitalization, talking, discussing and by working them up diagnostically • 8 children: are being counselled in child guidance clinic with significant improvement • 6 children: Gradual withdrawal of AEDs and substitution with tranquillizers worked cases who were on polytherapy for so called intractable seizures. • 2 children who had epileptic seizures earlier but now have hanged responded to gradual withdrawal of AEDs • 2 children failed to come for follow-up
Kramer et al[97]	1995	27	• All patients were seen by psychiatrists, and a plan for psychiatric treatment following discharge was designed. • AEDs were discontinued in all patients.
Lancman et al[98]	1994	43	• Different interventions were used in our patients, so that we were not able to determine
Wyllie et al[100]	1991	18	• All patients were told that the attacks were e motional in basis, and were advised to remain off AEDs and obtain psychological counselling.
Wyllie et al[101]	1990	21	• After discussion of diagnosis and treatment, all but 1 of the patients agreed to remain without antiepileptic medication. Sixteen patients agreed to have psychiatric treatment: 13 patients had outpatient treatment for 3 to 36 (mean 9) months, 8 patients had inpatient treatment for 0.5 to 13 (mean 2) months, and 5 patients had both. Five patients were treated with psychotropic medication (imipramine, desipramine, or haloperidol). Few of the patients returned to their pediatric neurologist for follow-up care

AEDs = Antiepileptic Drugs, MDT= Multidisciplinary Team, Psychogenic nonepileptic events (PNEE), *Unclear if all patients received the intervention, PNES= Psychogenic Nonepileptic SeizuresDubowitz, V., & Hersov, L. (1976). Management of children with non-organic (hysterical) disorders of motor function. *Developmental Medicine & Child Neurology*, *18*(3), 358-368.

Table 6: Prevalence of Psychiatric Disorders in Children with psychogenic non-epileptic seizures (PNES)

Author and Year	Criteria/ Measure	Sample Size	Any	Depression	Anxiety	ADHD	Autism	PTSD	Psychosis	Eating Disorder	Bipolar Disorder	ODD	Other
Hansen et al 2021[30]	ICD-10[6]	384	39%	11%		12%*			7%	3%	NR	NR	• Adjustment disorder 12.5% • SSRD 9.1% • Personality Disorder 5% • Self-harm 1% • Substance use 5%
Fredwell et al 2021[27]	Parent report	23	70%	26%	35%	NR	NR	13%	NR	NR	NR	NR	NR
Fredwell et al 2021[29]	Parent Report	125	NR	16%	22%	7%	2%	8%	NR	NR	1%	NR	• Suicidal ideation/self-harm 6 (5%)
Sawchuck et al 2020[31]	DSM5[5]and/or chart review	33	NR	42%	67%	NR	NR	6%	NR	NR	NR	NR	NR
Masi et al 2020[34]	KSADS-PL[103]	22	NR	50%	NR	NR	NR	NR	NR	NR	NR	NR	NR
Terry et al 2020[37]	Medical Records	101	68%	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR
McWilliams et al 2019[44]	ADOS[110] ADI-R[111] ASDI[112] ICD-10[6]	59	50%	24%	NR	8.5%	16.9%	3%	NR	3%	NR	2%	• Tic disorder 5.1% • Social anxiety 14% • Other anxiety 14% • OCD 3% • Tic disorder 5%

Uzun et al 2019[43]	K-SADS-PL[103]	42	64%	26%	31%	24%	NR	NR	NR	7%	NR	2%	• Conduct disorder 5%
Myers et al 2019[40]	Psychiatric History	15	100%	40%	13%	13%	NR	7%	NR	NR	7%		• OCD 7% • Panic disorder 20% • Substance 13% • Suicide attempt 20% • Self-harm 7%
Luthy et al 2018[48]	DSM5/ICD[5]	399	41%	8%	27%	NR	NR	8%	NR	NR	10%	NR	NR
Madanna et al 2018[45]	DSM-IV-TR[113]	60	NR	14%	37%	NR	NR	12%	NR	NR	NR	1%	• Panic disorder 3% • Adjustment disorder 9% •
Kozlowska et al 2017[50]	DSM-IV[114]	60	NR	17%	37%	NR	NR	12%	NR	1%	NR	NR	• Panic disorder 12% • Behavioral disorder 5%
Valente et al 2017[7]	K-SADS-PL[103] DSM-IV[114] ICD-10[6]	53	NR	45%	36%	4%	NR	NR	NR	NR	NR	NR	• 15.1% somatoform disorders • 18.9% conduct disorder
Say et al 2015[60]	KSADS-PL[103]	62	NR	15%	20%	24%	NR	11%	NR	NR	NR	NR	• 12.9% Disruptive behaviours disorder
Yadav et al 2015[62]	Data collected at diagnosis	90	67%	36%	23%	11%	NR	6%	6%	NR	NR	NR	• ODD/PDD 6%

Plioplys et al 2014[66]	K-SADS-PL[103]	55	NR	44%	84%	29%	NR	26%	NR	NR	NR	NR	NR
Sawchuk & Buchhalter 2015[64]	Medical chart and/or DSM-IV[114]	29	NR	52%	21%	NR	NR	NR	NR	NR	NR	NR	• 28% attention, speech or learning disorder • 21% self-harm
Rawat et al 2015[61]	DSM-5[5]	34	NR	15%	NR	7%	NR	NR	NR	NR	NR	NR	NR
Say et al 2014[65]	KSADS-PL[103]	34	NR	65%	26%	NR	29%	18%	NR	NR	NR	9%	• Conduct disorder 6% • Alcohol/substance use disorder 15% • Suicide attempt 15% • OCD 6% • Generalized anxiety disorder 9% • Separation anxiety disorder 3% • Specific phobia 3%
Yi et al 2014[69]	DSM-IV[114]	25	NR	36%	12%	28%	NR	NR	4%	NR	4%	NR	• Adjustment disorder 8% • Conduct disorder 4%
Akmedir et al 2013 [74]	K-SAD-PL[103]	34	NR	27%	35%	29%	NR	3%	NR	NR	NR	9%	• Nicotine use disorder 15%
Verrotti et al 2009[81]	DSM-IV[114]	36	42%	19%	8%	NR	NR	NR	NR	NR	NR	NR	• Generalized anxiety disorder 8%

	ICD-10[6]												• Panic disorder 11%
Patel et al 2007[84]	Pre-existing diagnosis of diagnosis made at time of diagnosis by psychiatrist	59	41%	25%	7%	NR	NR	NR	NR	NR	NR	NR	NR
Vincentiis et al 2006[85]	DSM-IV[114] ICD-[106] KIDDIE-SADS[115]	21	NR	62%		NR	NR	NR	NR	NR	NR	10%	• Pure dissociative disorder 14% • Conduct disorder 9.5%
Bhatia & Sapra 2005[87]	ICD-10[6]	50	NR	24%	32%	NR	NR	NR	NR	NR	NR	NR	• Panic disorder 12%
Pakalnis &Paolicchhi 2003[89]	Interview	22	NR	41%	41%	5%	NR	NR	5%	NR	5%	5%	
Wyllie et al 1999[94]	DSM-IV[114]	34	NR	29% Major depression and dysthmic disorder	29% separation anxiety and overanxious disorder	15%	NR	9%	6%	NR	3%	3%	• Panic disorder 9% • Impulse control disorder 3% • Adjustment disorder with mixed emotional features 6%

*includes Tourette's syndrome and conduct disorder, ^Based on prevalent disorder and not incident disorders , ASDI=Asperger syndrome Diganostic Interview, ADOS= Autism Diagnostic Observation Schedule, ADI-R= Autism Diagnostic Interview , DSM-5= The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (***DSM-5***), DSM-IV-TR - Diagnostic and statistical manual of mental disorders, text revision, DSM-IV= Diagnostic and Statistical Manual of Mental Disorders -Fourth Edition , KIDDIE-SADS -Schedule for Affective Disorders and Schizophrenia for School-Age Children—Epidemiological Version (KIDDIE-SADS), KSADS-PL= Diagnostic and Statistical Manual of Mental Disorders, ICD-10 = International Classification of Diseases, Tenth Revision, NR= Not reported , SSRD = somatic symptom and related disorders