Seizure: European Journal of Epilepsy 71 (2019) 279–285

Contents lists available at ScienceDirect

Seizure: European Journal of Epilepsy

journal homepage: www.elsevier.com/locate/seizure

Review

Patient and family perspectives of paediatric psychogenic non-epileptic seizures: A systematic review

Amanda Dunne[a,b,*], Rory Carolan[a], Lorraine Swords[b], Gillian Fortune[a]

[a] Children's Health Ireland (CHI) at Crumlin, Cooley Road, Crumlin, Dublin, D12 V004, Ireland
[b] School of Psychology, Trinity College Dublin, the University of Dublin, Dublin 2, Ireland

ARTICLE INFO

Keywords:
Psychogenic nonepileptic seizures
Nonepileptic attack disorder
Nonepileptic events
Subjective experience
Family perspectives
Systematic review

ABSTRACT

Exploring the perspectives of those affected by psychogenic non-epileptic seizures (PNES) may be essential in learning more about the nature of this condition. The aim of this systematic review is to synthesise the evidence regarding the perspectives of children and adolescents with PNES, and the perspectives of their parents, caregivers and families. Studies were included if they (1) explored PNES in a paediatric population, (2) explored the perspectives of the child or adolescent with PNES, or the perspectives of their parents, caregivers or families, (3) were original research, and (4) were written in the English language. Eight studies were identified for inclusion following searching of CINAHL Complete, Medline (Ovid), PsycINFO, PubMed and Web of Science databases, along with additional hand searching of reference lists. Quality assessment of articles was conducted using the Critical Appraisal Skills Programme (CASP) qualitative checklist. Seven articles were deemed high quality, and one article was deemed moderate quality. Common threads across studies included: "legitimacy and the importance of understanding", "distress and the social and emotional impact of PNES" and "moving forward". Clinicians must take care in the delivery of the diagnosis; including the use of an appropriate name for this condition, and providing an explanation of PNES that is acceptable to the patient, as well as ensuring that follow-up support is provided. Further reviews are required that utilise more well-established quality appraisal scoring systems and with the inclusion of grey literature, which refers to evidence not published by commercial academic publishers.

1. Introduction

Psychogenic non-epileptic seizures (PNES) are noticeable changes in an individual's consciousness or behaviour that share a similar appearance to epileptic seizures, with the key difference being that PNES are not accompanied by electrophysiological changes [1]. Although as many as 15 different names have been documented in describing this condition [2], PNES is the term that will be employed throughout this systematic review. Reuber [3] posits that these seizures are best understood through the lens of a biopsychosocial, etiologic model, whereby interacting predisposing, precipitating, perpetuating and triggering factors are seen to play a role in the subsequent presentation of PNES. An incidence of 1.5 per 100,000 each year has been reported in relation to rates of referrals to neurological settings for diagnosis for adults with PNES [4], with these rates reportedly being lower in children [1]. One study found that PNES in paediatric patients was associated with a positive outcome in 76.5% of cases [5].

As the name suggests, PNES are seen as being psychological in nature [2]. In spite of agreement surrounding the psychological basis of PNES, Brown and Reuber [6] note that there remains little consensus regarding the exact mechanisms underlying this illness; highlighting the need for further research to be undertaken. Nevertheless, a review of 24 studies conducted by Reilly et al. [1] found in paediatric populations that physical and sexual abuse, interpersonal and familial conflicts, and school-related difficulties such as bullying and poor academic performance appeared to be the most frequently reported precipitating factors to the onset of PNES. It has been reported that it may take up to seven years to reach a diagnosis of PNES [7]. Prior to reaching the appropriate diagnosis, PNES may be incorrectly diagnosed as epilepsy and presenting individuals subjected to unnecessary interventions and potentially damaging treatment with antiepileptic medication as a result [1,8]. In light of the lengthy process involved in reaching a diagnosis of PNES, and also the distressing precipitating factors associated with seizure onset, exploring the perspectives of children and young people with PNES may be essential in learning more about the nature of this condition so that diagnosis and subsequent treatment are more

* Corresponding author at: Psychology Department, Children's Health Ireland (CHI) at Crumlin, Cooley Road, Crumlin, Dublin, D12 V004, Ireland.
E-mail address: dunnea17@tcd.ie (A. Dunne).

https://doi.org/10.1016/j.seizure.2019.08.014
Received 11 July 2019; Received in revised form 24 August 2019; Accepted 28 August 2019

stream-lined and targeted to their needs.

Evidence from the literature highlights that research regarding paediatric PNES is generally lacking in comparison to literature conducted with adults with this illness [2]. A number of studies can be found exploring perspectives of PNES in adult populations. Stone et al. [9] examined 102 adult general neurology patients' thoughts on the appropriate name for PNES, with 'symptoms all in the mind' and 'hysterical seizures' being deemed highly offensive, while 'functional seizures' and 'stress-related seizures' were found to be significantly less offensive. The naming of PNES may be particularly important, as evidence from 84 adult patients in the study conducted by Carton et al. [10] indicated that a negative reaction to the diagnosis may be a predictor of poor prognosis.

Extending the findings of Reilly et al. [1] within adult populations, information provided by five adult participants with PNES in the study conducted by Dickinson et al. [11] highlighted histories of stressful life events such as difficulties in relationships, work conflicts, and bereavement. These participants reported feelings of tiredness, overwhelming emotion and stress preceding their seizures, and they also highlighted the frustration and dissatisfaction associated with the lengthy and demanding process of reaching a diagnosis [11]. A major negative impact of PNES has been emphasised in studies with adult participants, including isolation, a loss of normality, and a loss of independence [11,12]. Dissatisfaction with information and support post-diagnosis has also been documented across studies. Adults with PNES have reported difficulties in finding information and abandonment by services post-diagnosis, resulting in limited understanding of the condition [11,12]. In terms of their opinions and preferences regarding treatment outcomes, adults with PNES highlighted a hope for validation due to questioning of the legitimacy of their illness, gaining understanding from others, and finding answers [12].

While the perspectives of adults with PNES are well documented, there remains a need to explore the extent of research dedicated to exploring perspectives of paediatric PNES patients and whether the perspectives from both groups are comparable. The aim of this systematic review is to synthesise the evidence regarding the perspectives of children and adolescents with PNES, and the perspectives of their parents, caregivers and families.

2. Methods

The current systematic review was conducted utilising the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [13].

2.1. Searching

The wide variation in terminology utilised in the naming of PNES across the literature resulted in the necessity to conduct an inclusive and comprehensive search strategy. To capture all relevant studies, the title and abstract fields, or title and topic fields in the case of Web of Science, were searched in the following databases, from the earliest accessible dates at our institution, to the point of searching:

- CINAHL Complete (1982 to June, week 4, 2018)
- Medline (Ovid) (1946 to June, week 4, 2018)
- PsycINFO (1990 to June, week 4, 2018)
- PubMed (1965 to June, week 4, 2018)
- Web of Science (1990 to June, week 4, 2018)

The search was completed in June 2018.

The following terms were utilised in searching all five databases:

(parent OR parents OR parental OR family OR families OR child OR children OR adolescent OR adolescents OR teenager OR teenagers OR caregiver OR caregivers) AND (perspective OR perspectives OR experience OR lived experience OR lived experiences OR view OR views OR outlook OR attitude OR attitudes OR impact) AND (pseudo-seizure OR pseudoseizure OR nonepileptic attack disorder OR non-epileptic attack disorder OR NEAD OR nonepileptic seizure OR non-epileptic seizure OR nonepileptic event OR non-epileptic event OR NEE OR stress-related seizure OR stress related seizure OR functional seizure OR psychogenic seizure OR psychoseizure OR pseudo-epileptic attack OR pseudo epileptic attack OR pseudo-epileptic seizure OR pseudo epileptic seizure OR hysterical seizure OR psychogenic attack OR hysterical attack OR pseudo-epilepsy OR pseudo epilepsy OR hysteroepilepsy OR hysterical epilepsy OR psychogenic nonepileptic seizure OR psychogenic non-epileptic seizure OR PNES OR non-epileptic attack OR nonepileptic attack OR dissociative seizure OR dissociative convulsion OR hysterical convulsion OR conversion seizure)

For each database the searches were limited to title or abstract/topic search, English language, and childhood and adolescence age groups. The age group limiters differed slightly in each database. For example, in the Medline (Ovid) database, the "all child (0–18 years)" limiter was selected, while in the CINAHL Complete database, the "Adolescent: 13–18 years", "All Infant", and "All Child" limiters were selected.

2.2. Selection

Inclusion and exclusion criteria were developed prior to conducting the searches to meet the aims of this systematic review.

Inclusion Criteria:

- Explored PNES in a paediatric population
- Explored the perspectives of the child or adolescent with PNES, or the perspectives of their parents, caregivers or families
- Original research
- Written in English language

Exclusion Criteria:

- Studies exploring non-epileptic seizures that are of organic origin or non-epileptic seizures where a psychogenic origin was not clearly specified
- Reference to the perspectives of the child or adolescent with PNES, or the perspectives of their parents, caregivers or families was too limited to warrant inclusion within the current systematic review
- Did not explore PNES in a paediatric population
- Did not explore the perspectives of the child or adolescent with PNES, or the perspectives of their parents, caregivers or families
- Book reviews, conference abstracts, journal letters and journal notes
- Written in non-English language

The total number of records identified through database searching was 1090. Two additional records were identified through checking reference lists. Hand searching of records for duplicates was conducted, with a total of 974 records remaining following the removal of 118 duplicates. The titles and abstracts of the remaining 974 records were reviewed for relevance, with records clearly violating inclusion criteria being excluded at this stage. 32 records were retained for full-text review following title and abstract screening. The majority of papers were excluded during the full-text review for exploring PNES in adult populations (n = 18). Other reasons for exclusion during this stage included not exploring the perspectives of the child or adolescent with PNES, or the perspectives of their parents (n = 2), not being original research (n = 1), not clearly specifying if the non-epileptic seizures being studied were of psychogenic origin (n = 1), the focus on the perspectives of the child or adolescent with PNES, or the perspectives of their parents, was too limited to warrant inclusion (n = 1), and a lack of differentiation between results pertaining to children and adults with PNES in mixed samples (n = 1). A total of eight studies were included in the systematic review (Fig. 1).

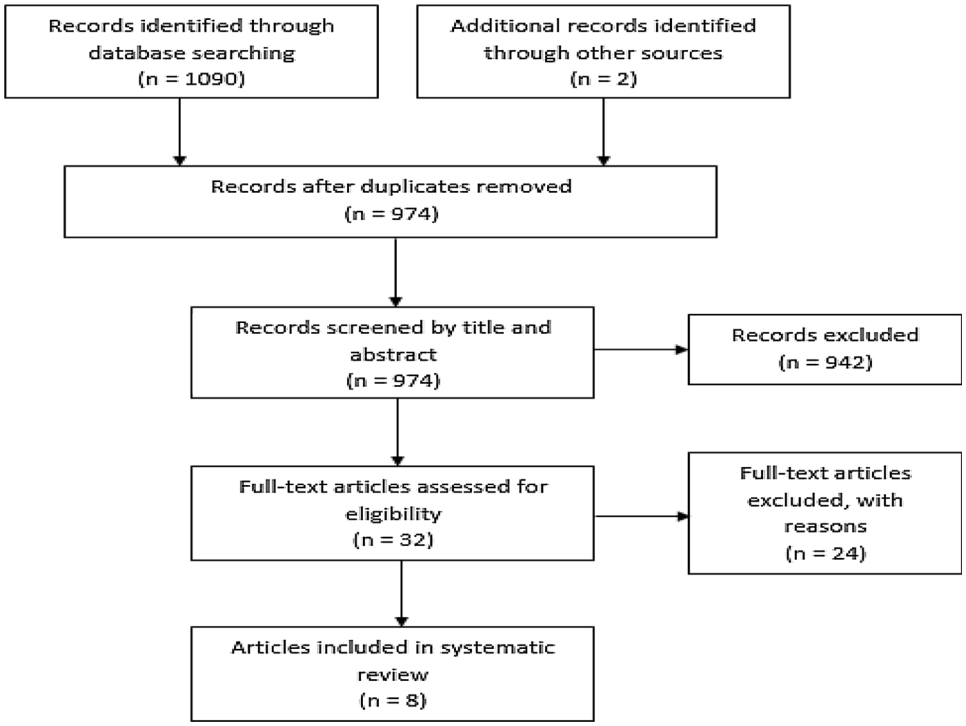

Fig. 1. Flow Diagram Summarising Search and Selection Process.

2.3. Quality assessment

Each paper included in the systematic review was assessed using criteria outlined in the Critical Appraisal Skills Programme (CASP) qualitative checklist [14] as recommended by Hannes [15]. This checklist was adapted slightly for use within this systematic review, and a categorical system of low, moderate and high quality was also implemented as an estimated guide to the quality of studies. For the articles that employed qualitative methods, the CASP qualitative checklist was utilised with a slight alteration to the wording of Question 10 ("How valuable is the research?" was changed to "Is the research valuable?" to facilitate the application of a categorical scoring system). Each question within this checklist was scored using "Yes" (3), "Can't Tell" (2) and "No" (1) responses. Once the responses to the 10 questions were summed, an overall score of 1–10 indicted a low quality study, 11–20 indicated a moderate quality study, and 21–30 indicated a high quality study.

It was found that a number of articles within the systematic review did not fit neatly into any of the available eight CASP checklists. For this reason, the CASP qualitative checklist was again adapted for use with these quantitative articles. Alterations included replacing qualitative terminology with quantitative terminology. For example, while Question 2 originally read as: "Is a qualitative methodology appropriate?", this was changed to "Is a quantitative methodology appropriate?". A final change involved the omission of Question 6: "Has the relationship between researcher and participants been adequately considered?" as it was deemed less applicable for quantitative research. The same categorical scoring system as mentioned above was utilised. However, the inclusion of only nine questions within this adapted checklist resulted in an overall score of 1–9 indicating a low quality study, 10–18 indicating a moderate quality study, and 19–27 indicating a high quality study.

2.4. Inter-rater reliability

The initial searches were independently conducted by two researchers and subsequently compared to ensure that the same results were yielded. Full-text review of articles to determine eligibility was also independently conducted by two researchers. If any inter-rater discrepancies were identified, an a priori procedure would be followed whereby both researchers reassessed the article for inclusion and if any disagreement remained, a third researcher was also asked to rate the article. The decision regarding whether the article was eligible for inclusion in the systematic review or not would then be determined based on the majority decision. The quality assessment of included articles also was separately conducted by two researchers and any discrepancies subsequently discussed until a consensus was reached.

2.5. Data Extraction

Data extraction was conducted by the first author once consensus was reached regarding the final sample of studies to be included within the systematic review. Information recorded from each of the relevant identified studies included primary author, publication year, PNES sample characteristics (sample size, sex, mean age, and age range), parent/caregiver/family characteristics (sample size, sex, mean age, and age range), qualitative or quantitative methodology, and key findings highlighting the perspectives of children and adolescents with PNES, and the perspectives of their parents, caregivers and families.

3. Results

3.1. Study characteristics

An outline of general characteristics from the eight included studies can be found below in Table 1. These characteristics include: primary author and publication year, sample features, methodology, and key findings.

3.2. Key findings

3.2.1. Legitimacy and the importance of understanding

The concept of legitimacy and its relationship with PNES was a recurring theme in three studies [16–18]. Within the study conducted

Table 1
Characteristics of Included Studies.

Primary Author Publication Year	PNES Sample Characteristics			Parent/Caregiver/Family Characteristics			Qualitative or Quantitative Methodology	Key Findings
	N	Sex	Mean Age (Range)	N	Sex	Mean Age (Range)		
Irwin, 2000	35	24 F, 11M	NR (6-18)	NA	NA	NA	Quantitative	8 had traumatic histories of violence and physical and sexual abuse; 4 were subjected to significant domestic tension; 10 were avoiding school due to bullying, poor performance or falling behind in schoolwork; 3 had dysfunctional relationships with their mothers.
Karterud, 2016	11	11F	NR (14-24)	NA	NA	NA	Qualitative	4 key themes: (1) Delegitimising experiences (2) Concealing the diagnosis (3) Support and understanding from close relationships (4) Perceiving PNES as a legitimate disorder
Karterud, 2015	11	11F	NR (14-24)	NA	NA	NA	Qualitative	3 key themes: (1) Threatened self-image (2) Being believed and belief in oneself (3) Getting an explanation that makes sense
Lancman, 1994	18	NR	NR (NR)	NR	NR	NR (NR)	Qualitative	School performance, independence and attendance in school were apparently more impaired than family and social relationships when the seizures were present as compared to seizure-free periods.
McWilliams, 2016	10	4F, 6 M	15.5 (11-19)	29	21 F, 6 M, 2NR	NR (NR)	Qualitative	6 key themes: (1) Upset and afraid (2) Missing out (3) Feeling misunderstood (4) Confusion and uncertainty (5) Less than epilepsy (6) Making sense and moving on
Morgan, 2013	NA	NA	NA	146	NR	NR (NR)	Quantitative	The most offensive term overall was “all in his/her head”. This term along with “hysterical seizures” and “psychogenic seizures” made up Group 1 of the most offensive terms. The least offensive term overall was “nonepileptic events”. This term along with “nonepileptic attack disorder” and “functional seizures” made up Group 2 of the least offensive terms. The moderately offensive terms in Group 3 were “stress-related seizures”, “paroxysmal seizures” and “pseudo-seizures”.
Say, 2014	34	23 F, 11M	14.26 (11-18)	NA	NA	NA	Quantitative	Adolescents with PNES experienced significantly higher rates of parental conflicts and difficulties in relationships with peers and siblings in comparison to adolescents with epilepsy and healthy controls. School underachievement and a history of traumatising/stressful events, physical and sexual abuse were also significantly more common in adolescents with PNES.
Yi, 2014	25	14 F, 11M	13.82 (8-19)	NA	NA	NA	Quantitative	PNES onset was frequently precipitated by stressful situations, such as interpersonal and familial distress. 10 participants experienced familial distress and 6 participants experienced social distress; including bullying, assault, adjustment failure and academic failure.

Notes: N = sample size; F = female; M = male; NR = not reported; NA = non-applicable.

by McWilliams et al. [18], a number of participants reported having negative experiences with acquaintances, teachers and clinicians whereby the suggestion was made that their seizures were fabricated. Comparisons between PNES and other somatic illnesses were drawn across two studies [17,18]. Participants expressed feeling that PNES may be regarded as a less legitimate illness than others deemed more somatic in nature [17], and both young people with PNES and their families reported that individuals with epilepsy gained much greater social and educational support [18]. Young people in the study conducted by Karterud et al. [16] noted that snide comments made by teachers highlighted their belief that the PNES were under their control, and they also reported feeling delegitimised by work colleagues and employers who stated that it was up to themselves to recover [16].

Recognition of the importance of PNES to be regarded as a legitimate illness is encompassed within the overarching need of young people to understand their own illness and to be understood by others. Eight of 11 participants within the study conducted by Karterud et al. [17] resisted their PNES diagnosis because they regarded themselves as being mentally healthy, whereas they perceived a diagnosis of PNES to be associated with mental illness. Young people almost automatically expected to not be believed or taken seriously by healthcare professionals, and some also displayed difficulty in understanding whether their seizures really were beyond their conscious control [17]. Some reported feeling better understood by their friends and peers in comparison to their employers and teachers, and they also noted that the amount of information about their diagnosis they shared with others depended on what they felt the other person was able to accept [16]. Young people tended to be most open with people closest to them, and one motivation for this appeared to stem from their desire to be understood and believed [16]. Young people with PNES and their families highlighted that the majority of people they met displayed a fundamental lack of understanding of this illness, and they described how healthcare professionals in non-specialist settings appeared unsure as how to investigate and manage PNES [18]. Indeed, families of young people with PNES themselves appeared uncertain about the nature of this illness [18].

3.2.2. Distress and the social and emotional impact of PNES

Across a number of studies, young people with PNES were reported as having histories of personal and familial distress. Lancman, Asconapé, Graves and Gibson (1994) found that five participants had a history of sexual abuse, three participants had a history of drug abuse, and 14 had a history of conflict within their familial relationships. In comparison to adolescents with epilepsy and healthy controls, a history of traumatising/stressful events, sexual and physical abuse was found to be significantly more common in adolescents with PNES [19]. Yi et al. [20] reported that histories of interpersonal or familial distress were seen as precipitants of PNES in their sample. Such traumatic histories were also classified as potential precipitants to PNES onset in the study conducted by Irwin, Edwards and Robinson (2000), with experiences of domestic tension, bullying, dysfunctional maternal relationships, violence, and physical and sexual abuse being reported.

PNES were found to have a significant impact on the lives of young people across multiple domains. The young people and their families both expressed fear about the impact of this diagnosis on their current and future lives; with young people feeling guilty about the strain that their illness could be placing on their families, and family members feeling frustrated that they could not be of more assistance when a seizure occurred [18]. Young people found it frightening to be given a diagnosis that did not fit with their self-image of being mentally healthy, and felt alienated when healthcare professionals used words highlighting its psychogenic nature [17].

As well as having an emotional impact, PNES also were found to have a profound impact on social, behavioural, and academic functioning. Families in the study conducted by McWilliams et al. [18] reported a loss of employment and avoidance of daily outings and holidays as a result of the diagnosis. Young people described a loss of independence and friendships, difficulties in relationships with parents, siblings and teachers, underachievement in school, and impaired school attendance across studies [18–21]. Some young people resorted to hiding their diagnosis as a form of protection against stigmatisation; with this desire to conceal resulting in withdrawal from attendance at school and work, not socialising with friends and avoidance of public places at the extreme end [16].

3.2.3. Moving forward

Many studies discussed factors which may be important in facilitating the young person in moving forward with their illness. Particular attention was given to the issue of appropriately naming the condition in two studies [18,22]. Most families preferred names which highlighted its relationship with epilepsy such as non-epileptic seizures, while terms associated with fabrication, such as pseudoseizures, were reported as being unhelpful [18]. Young people in this study appeared more indifferent to the naming of their illness, but did note that a name highlighting that it is not epilepsy would be helpful particularly in school settings to clarify that the management is not the same [18]. Parents in the study conducted by Morgan et al. [22] unanimously agreed that “all in his/her head” was the most offensive term, followed by hysterical seizures and psychogenic seizures. The least offensive term overall was non-epileptic events, followed by non-epileptic attack disorder and functional seizures. In slight contrast to the findings by McWilliams et al. [18], the term pseudoseizures was placed into the moderately offensive group instead of being deemed most offensive, along with stress-related seizures and paroxysmal seizures [22].

Some young people felt that boredom, emotional stress and fatigue may trigger the onset of a seizure, with prevention techniques including attempts to relax or removal from a stressful situation [18]. Families tried to show affection or employ distraction techniques to prevent or cease seizures [18]. Assurance was provided when those closest to the young people displayed an understanding of the illness, which in turn facilitated social participation [16]. Young people also found it easier to socialise if their own understanding of PNES was acceptable to them [16]. An explanation of PNES as being associated with unconscious processes was found to aid young people in overcoming doubts about their own role in their seizures [17]. The provision of a multi-factorial, bio-medical explanation from a holistic perspective that highlighted how stress may cause bodily reactions seemed to aid their own understanding of their illness, while also acting as a possible prevention for being labelled by others as mentally ill [17]. Overall, young people felt that being taken seriously and understood was most helpful in moving forward [17].

3.2.4. Results of quality assessment

As mentioned above, a categorical system of low, moderate and high quality was implemented alongside criteria within the CASP qualitative checklist [14] to provide an estimate of the quality of included studies. Overall, seven studies were deemed to be of high quality under this criteria [16–20,22,23], and one study was deemed to be of moderate quality [21].

Across all studies, maximum scores were achieved for the provision of a clear statement of their aims, choosing an appropriate methodology, and implementing an appropriate recruitment strategy. The next highest scores across studies were obtained in relation to the rigorousness of data analysis and a clear statement of findings; with seven studies achieving the maximum score of 3. The only exception to this was the study conducted by Lancman et al. [21], due to insufficient discussion of the data analysis process, and limited discussion of participants’ perspectives surrounding social outcome.

Six studies achieved the top score for the appropriateness of data collection techniques [16–18,20,22,23], and for the value of their research [16–20,22]. Two studies achieved a score of 2 in relation to their data collection [19,21], due to issues such as the form of data collected

not clearly being specified, or a lack of justification of the methods chosen. One study achieved a score of 2 in relation to the value of their research [21], due to issues such as a lack of discussion in relation to the contribution of their study to previous knowledge. One study achieved a score of 1 for this question due to issues such as a lack of discussion surrounding ways in which their findings could be transferred or utilised in other ways, and a lack of identification of areas of research that may still be necessary following their research [23].

Scores across studies in relation to the appropriateness of the research design, consideration of the relationship between researcher and participants, and the consideration of ethical issues were more varied. Five studies achieved top scores for the appropriateness of their research design [16–19,22], while the three remaining studies achieved a moderate score of 2 [20,21,23]. Consideration of ethical issues was only moderately highlighted in six studies [16–20,22], and appeared to be almost omitted entirely from the write-up of two studies [21,23]. Approval from an ethics committee was not commented upon within both these studies. For the four studies for whom the question in relation to the adequate consideration of the relationship between researcher and participants was deemed relevant, one study scored top marks [17], two studies scored moderately [16,18], and one study scored minimally for their lack of clear consideration of this relationship within their write-up [21].

4. Discussion

The purpose of this systematic review was to synthesise any available evidence regarding the perspectives of children and adolescents with PNES, and the perspectives of their parents, caregivers and families. Common threads were found across the included studies; including the concept of legitimacy and the importance of understanding, a significant history of distress preceding the onset of seizures, and the negative impact of PNES, and reporting of factors that may affect moving forward with the illness.

Evidence from the paediatric literature largely extended on previous findings from the adult literature; highlighting a possible similarity of experiences that extends across different generations with PNES. For example, adult participants in the study conducted by Fairclough et al. [12] expressed a hope for validation from supports, due to experiences of the legitimacy of their illness being questioned; with similar experiences also being highlighted in relation to the legitimacy of paediatric PNES across three studies in this systematic review [16–18]. Significant distress preceding PNES and a largely negative impact was also echoed across both the adult and paediatric literature [11,12,16–21,23]. While a similarity in experiences was noted, the reasons behind such similar experiences varied between adults and children with PNES. One example of this pertains to the loss of independence reported in both adult and paediatric populations [11,18]. A loss of independence in adults with PNES pertained to the loss of their driving licence as a result of their seizures [11], while a loss of independence in young people was a result of not being allowed to socialise with friends, or parents insisting on accompanying them on public transport [18].

Findings from the literature highlight the difficulties associated with a paediatric PNES diagnosis, and the impact they may have on both the young person's and their family's experiences. Young people with PNES across the literature reported having negative experiences with acquaintances, teachers, clinicians, work colleagues and employers, who suggested that their seizures were fabricated, that they were under their control, and that it was up to themselves to recover [16,18]. An automatic expectation to not be believed or taken seriously by healthcare professionals was reported by young people [17]. A belief that healthcare professionals working within non-specialist settings demonstrate a fundamental lack of understanding of paediatric PNES was also reported [18]. Indeed, a study conducted with 61 paediatricians working in the field of neuropaediatrics and social paediatrics in Denmark highlighted disagreement regarding the use of terminology to describe PNES and diagnostic approach, as well as considerable inconsistencies in coding practices and variation in the use of the 'gold standard' video-electroencephalograph (v-EEG) to facilitate diagnosis [24]. Such inconsistencies noted among professionals working in the area of paediatric PNES may intensify the confusion surrounding this diagnosis experienced by young people and their families [24].

Young people with PNES and their families both expressed fear about the impact of this diagnosis on their current and future lives; with young people feeling guilty about the strain that their illness could be placing on their families, and family members feeling frustrated that they could not be of more assistance when a seizure occurred [18]. PNES also were found to have a profound impact on social, behavioural, and academic functioning, to include loss of employment, avoidance of daily outings and holidays, loss of independence and friendships, difficulties in relationships, underachievement in school, and impaired school attendance [18–21]. Further impacting the experiences of young people with PNES and their families is the high rates of psychopathology that have been found to be associated with this illness [25]. Research has shown that in comparison to their siblings, significantly more children with PNES have multiple psychiatric diagnoses, to include depression, anxiety, and post-traumatic stress disorder (PTSD) [25]. Taken together, findings from the research exploring the experiences of young people with PNES and their families further highlight the need for clinical guidelines to be developed to facilitate the diagnosis and appropriate treatment of this condition, so that this population may be better supported during this difficult time.

The results of the quality assessment was largely positive, with seven studies being deemed high quality and one study deemed to be of moderate quality. The use of the CASP qualitative checklist [14] has been previously recommended for use in the critical appraisal of research [15]. However, caution should be exercised in interpreting the results of our quality appraisal. Firstly, the CASP qualitative checklist was adapted slightly in the current review to encompass all identified articles, and also to facilitate the implementation of a categorical scoring system. Secondly, the CASP qualitative checklist [14] was initially intended for use as an educational tool; and thus a scoring system has not been developed for use with this checklist. This was overcome in the present study through the scoring of original response options on a range of 1-3. Nevertheless, the integrity of the CASP qualitative checklist remained largely intact within this review; a checklist developed by a team of experts in the field [14], permitting a degree of confidence to be held in the results of the quality assessment. Future reviews may overcome this limitation through the use of more well-established scoring systems for the quality appraisal of articles exploring personal and familial perspectives of paediatric PNES.

Another limitation is the exclusion of grey literature, which was outside the scope of the current systematic review. Grey literature refers to evidence not published by commercial academic publishers, and can include documents such as government reports, conference papers, theses, dissertations, and committee reports [26,27]. The inclusion of grey literature may have provided a more complete picture of the available research evidence [28], through the potential increase in the number of sources exploring personal and familial perspectives of paediatric PNES. The inclusion of grey literature may also reduce the risk of publication bias, and increase the overall comprehensiveness of the review [27]. A small number of articles were identified for inclusion within this review, following the use of strict inclusion and exclusion criteria. However, this may be representative of the limited research overall in the area of paediatric PNES [2]. Future reviews should endeavour to also explore grey literature to gain a more comprehensive view of the available literature.

In spite of these limitations, there are a number of strengths that should also be taken into account. To the best of our knowledge, this is the first paper that attempts to systematically review research exploring the perspectives of children and adolescents with PNES, and the

perspectives of their parents, caregivers and families; addressing a gap in the literature. The common threads identified across the paediatric literature highlight ways in which healthcare professionals and the general public may facilitate a young person with PNES in moving forward positively with their illness. Clinicians must take care in the delivery of the diagnosis, to include the use of an appropriate name for this condition. Use of a name highlighting its relationship with epilepsy may facilitate public understanding that different management protocols are necessitated for each condition [18,22]. Avoidance of terms associated with fabrication may aid in reducing the stigma experienced by young people with this condition and their families, and may also facilitate the young person's acceptance of this condition [18,22]. The provision of an explanation of PNES that is acceptable to the patient is required, to include an explanation that highlights the role of stress and other unconscious processes in causing bodily reactions [17]. Widespread education regarding the nature of paediatric PNES is required, as research has demonstrated that assurance is provided to young people with this condition when those closest to them display an understanding of their illness [16]. The development of clinical guidelines by specialists working within this field is greatly necessitated, so that young people with this condition and their families may receive the provision of appropriate support both during diagnosis and beyond.

Funding sources

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

Declaration of Competing Interest

All authors have no conflicts of interest to declare.

Acknowledgements

Thank you to Children's Health Ireland (CHI) at Crumlin for facilitating this research.

References

[1] Reilly C, Menlove L, Fenton V, Das KB. Psychogenic nonepileptic seizures in children: a review. Epilepsia 2013;54(10):1715–24. https://doi.org/10.1111/epi.12336.
[2] Reilly C, McWilliams A, Heyman I. What's in a name? 'Psychogenic' non-epileptic events in children and adolescents. Dev Med Child Neurol 2014;57(1):100–1. https://doi.org/10.1111/dmcn.12605.
[3] Reuber M. The etiology of psychogenic non-epileptic seizures: toward a biopsychosocial model. Neurol Clin 2009;27(4):909–24. https://doi.org/10.1016/j.ncl.2009.06.004.
[4] Sigurdardottir KR, Olafsson E. Incidence of psychogenic seizures in adults: a population-based study in Iceland. Epilepsia 1998;39(7):749–52. https://doi.org/10.1111/j.1528-1157.1998.tb01161.x.
[5] Rawat VS, Dhiman V, Sinha S, Sagar KJV, Thippeswamy H, Chaturvedi SK, et al. Comorbidities and outcome of childhood psychogenic non-epileptic seizures—An observational study. Seizure 2015;25:95–8. https://doi.org/10.1016/j.seizure.2014.09.011.
[6] Brown RJ, Reuber M. Psychological and psychiatric aspects of psychogenic non-epileptic seizures (PNES): a systematic review. Clin Psychol Rev 2016;45:157–82. https://doi.org/10.1016/j.cpr.2016.01.003.
[7] Reuber M, Fernandez G, Bauer J, Helmstaedter C, Elger CE. Diagnostic delay in psychogenic nonepileptic seizures. Neurology 2002;58(3):493–5.
[8] Reuber M, Elger CE. Psychogenic nonepileptic seizures: review and update. Epilepsy Behav 2003;4(3):205–16. https://doi.org/10.1016/S1525-5050(03)00104-5.
[9] Stone J, Campbell K, Sharma N, Carson A, Warlow CP, Sharpe M. What should we call pseudoseizures?: the patient's perspective. Seizure 2003;12(8):568–72. https://doi.org/10.1016/S1059-1311(03)00055-4.
[10] Carton S, Thompson PJ, Duncan JS. Non-epileptic seizures: patients' understanding and reaction to the diagnosis and impact on outcome. Seizure 2003;12(5):287–94. https://doi.org/10.1016/S1059-1311(02)00290-X.
[11] Dickinson P, Looper KJ, Groleau D. Patients diagnosed with nonepileptic seizures: their perspectives and experiences. Epilepsy Behav 2011;20(3):454–61. https://doi.org/10.1016/j.yebeh.2010.12.034.
[12] Fairclough G, Fox J, Mercer G, Reuber M, Brown RJ. Understanding the perceived treatment needs of patients with psychogenic nonepileptic seizures. Epilepsy Behav 2014;31:295–303. https://doi.org/10.1016/j.yebeh.2013.10.025.
[13] Liberati A, Altman DG, Tetzlaff J, Mulrow C, Gøtzsche PC, Ioannidis JPA, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. PLoS Med 2009;6(7):1–28. https://doi.org/10.1371/journal.pmed.1000100.
[14] Critical Appraisal Skills Programme. CASP qualitative checklist Retrieved from 2018https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-Download.pdf.
[15] Hannes K, et al. Critical appraisal of qualitative research. In: Noyes J, Booth A, Hannes K, Harden A, Harris J, Lewin S, editors. Supplementary guidance for inclusion of qualitative research in cochrane systematic reviews of interventions. United Kingdom: Cochrane Collaboration Qualitative Methods Group; 2011. Retrieved from https://methods.cochrane.org/qi/supplemental-handbook-guidance.
[16] Karterud HN, Haavet OR, Risør MB. Social participation in young people with nonepileptic seizures (NES): a qualitative study of managing legitimacy in everyday life. Epilepsy Behav 2016;57:23–8. https://doi.org/10.1016/j.yebeh.2016.01.009.
[17] Karterud HN, Risør MB, Haavet OR. The impact of conveying the diagnosis when using a biopsychosocial approach: a qualitative study among adolescents and young adults with NES (non-epileptic seizures). Seizure 2015;24:107–13. https://doi.org/10.1016/j.seizure.2014.09.006.
[18] 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 Behav 2016;59:128–36. https://doi.org/10.1016/j.yebeh.2016.03.029.
[19] Say GN, Tasdemir HA, Akbas S, Yüce M, Karabekiroglu K. Self-esteem and psychiatric features of Turkish adolescents with psychogenic non-epileptic seizures: a comparative study with epilepsy and healthy control groups. Int J Psychiatry Med 2014;47(1):41–53. https://doi.org/10.2190/PM.47.1.d.
[20] Yi YY, Kim HD, Lee JS, Cheon KA, Kang HC. Psychological problems and clinical outcomes of children with psychogenic non-epileptic seizures. Yonsei Med J 2014;55(6):1556–61. https://doi.org/10.3349/ymj.2014.55.6.1556.
[21] Lancman ME, Asconapé JJ, Graves S, Gibson PA. Psychogenic seizures in children: long-term analysis of 43 cases. J Child Neurol 1994;9(4):404–7.
[22] Morgan LA, Dvorchik I, Williams KL, Jarrar RG, Buchhalter JR. Parental ranking of terms describing nonepileptic events. Pediatr Neurol 2013;48(5):378–82. https://doi.org/10.1016/j.pediatrneurol.2012.12.029.
[23] Irwin K, Edwards M, Robinson R. Psychogenic non-epileptic seizures: management and prognosis. Arch Dis Child 2000;82(6):474–8.
[24] Wichaidit BT, Østergaard JR, Rask CU. Diagnostic practice of psychogenic nonepileptic seizures (PNES) in the pediatric setting. Epilepsia 2015;56(1):58–65. https://doi.org/10.1111/epi.12881.
[25] Plioplys S, Doss J, Siddarth P, Bursch B, Falcone T, Forgey M, et al. A multisite controlled study of risk factors in pediatric psychogenic nonepileptic seizures. Epilepsia 2014;55(11):1739–47. https://doi.org/10.1111/epi.12773.
[26] Haddaway NR, Collins AM, Coughlin D, Kirk S. The role of Google Scholar in evidence reviews and its applicability to grey literature searching. PLoS One 2015;10(9):1–17. https://doi.org/10.1371/journal.pone.0138237.
[27] Paez A. Grey literature: an important resource in systematic reviews. J Evid Based Med 2017;10(3):233–40. https://doi.org/10.1111/jebm.12265.
[28] Mahood Q, Van Eerd D, Irvin E. Searching for grey literature for systematic reviews: challenges and benefits. Res Synth Methods 2014;5(3):221–34. https://doi.org/10.1002/jrsm.1106.