



Review

Semiological classification of psychogenic nonepileptic seizures: A systematic review and a new proposal

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ABSTRACT

Purpose: A semiological classification of psychogenic nonepileptic seizures (PNES) may help address proper diagnosis and management and also provide standardization for future studies. The aim of the current paper was to systematically review the literature on the proposed classification systems for the semiology of PNES and to provide a new proposal based on the best available evidence.

Methods: I searched the electronic database PubMed on May 16, 2019 for articles that included the following search terms: “psychogenic” AND “semiology” or “classification” and also “nonepileptic” AND “semiology” or “classification” and also “pseudoseizure” AND “semiology” or “classification” since 1940. I applied the same methodology using the electronic database Scopus, though I limited the search to the title, abstract, and keywords.

Results: I could identify 15 classification systems through this search strategy using the electronic database PubMed. Searching the Scopus did not yield any additional relevant papers.

Conclusion: I proposed a new semiological classification system for PNES based on this systematic review. This includes three major classes of motor seizures, nonmotor seizures, and mixed semiology. A universally accepted and appropriate semiological classification system for PNES may lead to better standardization of future studies and may also help in better understanding of the pathophysiological basis of this condition.

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1. Introduction

Psychogenic nonepileptic seizures (PNES) are self-limited events that are characterized by paroxysmal changes in responsiveness, movements, or behavior. They may resemble epileptic seizures but lack a neurobiological origin similar to that in epileptic seizures and are not associated with electrophysiological epileptiform changes; they are often associated with psychological disorders [1]. They are common occurrences at epilepsy clinics and represent a universal human condition [1,2]. It is believed that despite cultural and socioeconomic differences, patients with PNES from different countries share several similarities [3,4].

Ironically, PNES are largely defined in terms of what it is not rather than what it is. Exclusion of other differential diagnoses (e.g., epilepsy) does little to define this common condition. Unfortunately, there is no universally accepted terminology, definition, and classification system for this condition. Obviously, an international consensus about PNES terminology, definition, and classification is very helpful and, as a matter of fact, is very much required, if we are to standardize measures

that can be used in diagnosis, management, and future research in this area.

Through the years, some experts have tried to provide various classification systems for PNES [5–8]. A semiological classification of PNES may help address proper diagnosis and management and also provide standardization for future studies [5]. The aim of the current paper was to systematically review the literature on the proposed classification systems for the semiology of PNES and to provide a new proposal based on the best available evidence.

2. Materials and methods

I searched the electronic database PubMed on May 16, 2019 for articles that included the following search terms: “psychogenic” AND “semiology” or “classification” and also “nonepileptic” AND “semiology” or “classification” and also “pseudoseizure” AND “semiology” or “classification” since 1940, to obtain the best relevant scientific evidence on the topic. I limited the search to the title and abstract. I selected the relevant articles after reviewing their titles, abstracts, and full texts (Fig. 1). I also included some of the references of the selected articles if they were relevant. Duplicates and non-English articles were excluded. Redundant classification proposals were considered just once. I applied the same methodology

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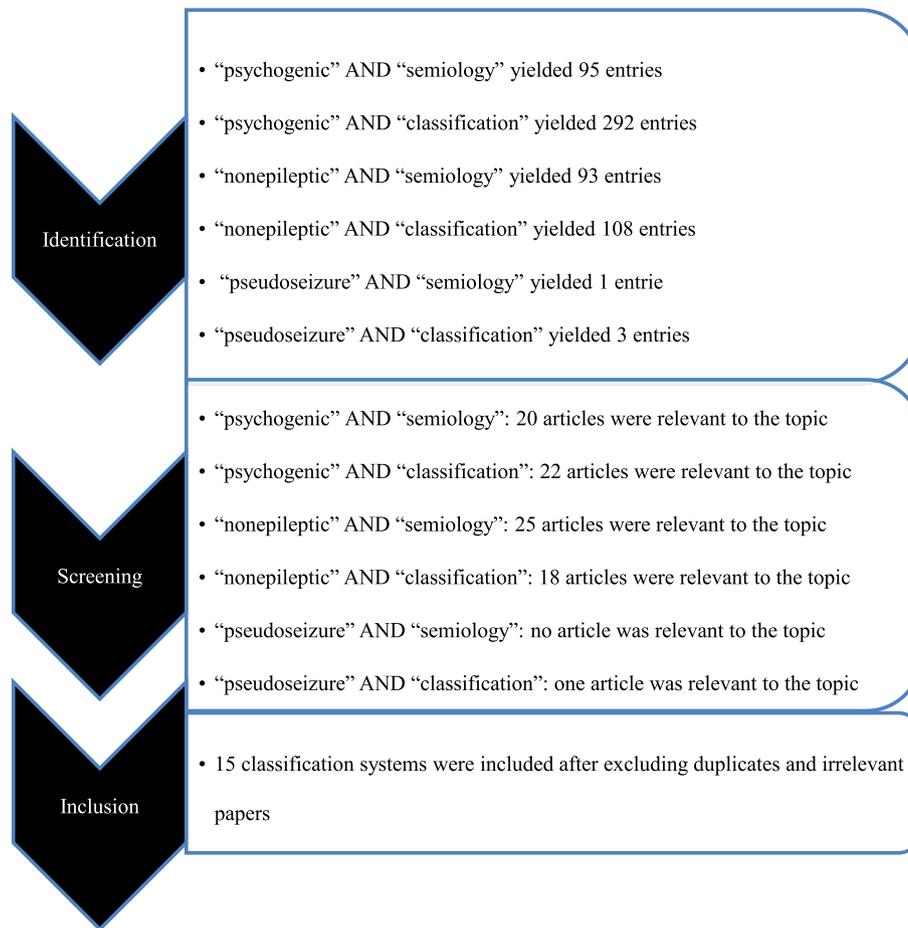


Fig. 1. The search and inclusion process; the electronic database PubMed on May 16, 2019.

using the electronic database Scopus, though I limited the search to the title, abstract, and keywords.

3. Results

I could identify 15 classification systems through the above search strategy using the electronic database PubMed (references [5–29]). Multiple manuscripts by the same author(s) were considered just once if they used the same classification system. Searching the Scopus database did not yield any additional relevant papers. Table 1 shows these articles, their proposed semiological classification systems, the significant associations that were identified with each classification system, and their major limitation(s).

4. Discussion

In this review, I observed that during the past four decades, many experts in the field have tried to propose different classification systems for PNES; this highlights the significance of the issue for the scientific community. However, there is still no universally accepted semiological classification system for PNES. While some of the proposed classification systems were very simple (e.g., motor seizures vs. nonmotor seizures) [9,11], others were more in detail and at times sophisticated [6, 12].

One common ground among all of the proposed classification systems was a try to make a distinction between motor vs. nonmotor seizures. Based on the experience and also review of the literature, this is easily doable for all healthcare professionals to recognize a motor seizure from a nonmotor phenomenon. In addition, this may have

important clinical implications with regard to the recognition of coexisting neurocognitive and psychiatric problems [9,11,14] and even outcome of the patients with PNES [15] (see below).

At the second level, it seems helpful to differentiate between subgroups of each class (motor and nonmotor classes). In motor seizures, it superficially makes sense to differentiate between those who have major motor phenomena (e.g., clonic and hypermotor movements of the upper and/or lower extremities, and pelvic thrusting) in comparison with those who have minor motor features (e.g., trembling of the upper and/or lower extremities). However, none of the studies that tried to do so noticed any benefits in such an attempt (Table 1). In addition, any attempt to make such a distinction may cause confusion and disagreement between different observers. This should be investigated more in future studies. In nonmotor seizures, it makes sense to subclassify the seizures into akinetic (catatonic or dialeptic) seizures and auras (subjective seizures). It has been reported that patients with akinetic seizures may have a better outcome and fewer coexisting neuropsychiatric problems compared with those who have motor seizures [14,15, 20]; however, some other studies had different observations [30,31]. In addition, there is evidence that many patients with PNES may have auras (with or without progression to other seizure types) [32]. Recognition of auras in patients with PNES could have important clinical implications. For instance, the presence of an aura might influence treatment approaches by training patients to recognize warning symptoms and employ cognitive or behavioral strategies aimed at averting a seizure. This hypothetical approach should be tested in future studies [32].

Furthermore, based on the experience and also review of the literature, it makes sense to have a group designated for seizures with mixed

Table 1
Proposed semiological classification systems for psychogenic nonepileptic seizures.

Author/year	Classification system	Description	Significance	Pitfalls
Groppe/2000	Motor seizures; Minor motor seizures; Atonic seizures	Motor seizures: clonic and hypermotor movements of the upper and/or lower extremities, pelvic thrusting, head movements, and tonic posturing of the head; Minor motor seizures: trembling of the upper and/or lower extremities; Atonic seizures: falling as the only symptom		Small sample size of 27 patients
Selwa/2000	Catatonic; Thrashing; Automatisms; Tremor; Intermittent long spells; Subjective	Catatonic: long periods of motionless unresponsiveness, often accompanied by waxy flexibility; Thrashing: relatively brief, four extremities, asynchronous, often violent movements, with or without ictal cries; Automatisms: small-amplitude, near-purposeful movements of the face or upper extremities with amnesia for ictal events; Tremor: rhythmic synchronous bilateral low-amplitude movements; Intermittent long spells: that included unusual behaviors such as rocking, screaming, and sometimes thrashing with interspersed sudden motionless unresponsiveness; Subjective: reports of sensory or emotional experiences, generally with retained consciousness and behavior, but amnesia for the events.	There was a higher incidence of baseline EEG abnormalities in the thrashing group (31%) than in the catatonic group (0%). There was a higher incidence of complete remission of spells in the catatonic group (53%) than in the thrashing group (21%).	Small sample size of 57 patients
Griffith/2007	Catatonic; Minor motor; Major motor; Subjective	Catatonic (long periods of motionless unresponsiveness, often accompanied by waxy flexibility); Minor motor (low-amplitude near-purposeful movements of the face or upper extremities, low-amplitude rhythmic/synchronous bilateral movements); Major motor (brief four-extremity asynchronous [often violent] movements, long spells that include unusual behaviors [e.g., rocking, screaming, thrashing] interspersed with sudden motionless unresponsiveness); Subjective (reports of sensory or emotional experiences, generally with retained consciousness and behavior)	1. Catatonic subtype was associated with significantly lower scores on Minnesota Multiphasic Personality Inventory 2 (MMPI-2). 2. Major motor activity was the main feature in adults, and minor motor activity was more prevalent among children (Alessi [29]).	Small sample size of 62 patients
Griffith/2008	Catatonic; Minor motor; Major motor	Catatonic (long periods of motionless unresponsiveness, often accompanied by waxy flexibility); Minor motor (low-amplitude near-purposeful movements of the face or upper extremities, low-amplitude rhythmic/synchronous bilateral movements); Major motor (brief four-extremity asynchronous [often violent] movements, long spells that include unusual behaviors [e.g., rocking, screaming, thrashing] interspersed with sudden motionless unresponsiveness)	Catatonic subtype was associated with fewer depressive symptoms and better verbal memory.	Small sample size of 47 patients
Chen/2010	The hypermotor group; The hypomotor group	The hypermotor group: characterized by violent and disorganized movements of the extremities, pelvic thrusting, head movements, or posturing mannerisms; The hypomotor group: characterized by altered responsiveness associated with either mild motor or trembling-like activities, unifocal negative (loss of function) symptoms, or diffuse absence of any motor phenomena		Small sample size of 68 patients
An/2010	Minor motor seizures; Major motor seizures; Unresponsive seizures	Minor motor seizures: trembling of the extremities; Major motor seizures: hypermotor movements of the extremities, hyperventilation, vocalization, head movements, body rigidity, pelvic thrusting, atonia, gaze, and jaw clenching; Unresponsive seizures: eye closure, unresponsiveness, and absence of any prominent movements		Small sample size of 64 patients
Seneviratne/2010	Rhythmic motor; Hypermotor; Complex motor;	Rhythmic motor: rhythmic tremor or rigor-like movements; Hypermotor: violent movements;		Video-EEG studies were visually analyzed and

(continued on next page)

Table 1 (continued)

Author/year	Classification system	Description	Significance	Pitfalls
	Dialeptic; Nonepileptic auras; Mixed PNES	Complex motor: complex movements such as flexion, extension, abduction, adduction, rotation, with or without clonic-like and myoclonic-like components of varying combinations and anatomic distribution; Dialeptic: unresponsiveness without motor manifestations; Nonepileptic auras: subjective sensations without any external manifestations; Mixed PNES: combinations of above seizure types		classified by a single observer
Hubsch/2011	Dystonic attack with primitive gestural activity; Paucikinetic attack with preserved responsiveness; Pseudosyncope; Hyperkinetic prolonged attack with hyperventilation and auras; Axial dystonic prolonged attack			High rate of comorbid epilepsy (46%)
Szabó/2012 (modified from Seneviratne 2010)	Minor motor; Major motor; Dialeptic; Nonepileptic aura; Mixed PNES	Minor motor: a homogenous motor activity (mainly tremor but also tonic-like movement could be observed), which is localized, mainly synchronized, and more often involves the upper limb accompanied by retained responsiveness; Major motor: complex movements involving several limbs or migrating and appearing as various types, with impaired consciousness; Dialeptic: unresponsiveness without motor manifestations; Nonepileptic auras: subjective sensations without any external manifestations; Mixed PNES: combinations of above seizure types	Dialeptic PNES seems to be more frequent among younger children	Small sample size of 27 children
Dikmen/2013	Pseudoaura; Dialeptic; Motor; Special spells (atonic, astatic, hypotonic)	Pseudoaura; Dialeptic: unresponsive status to external stimulus; Motor: muscle contractions that produce movement; Special: atonic (sudden loss or diminution of muscle tonus), astatic (loss of erect posture), and hypotonic (gradual decrease in muscle tone)		Small sample size of 37 patients
Dhiman/2013 and 2014 (modified from Seneviratne 2010)	Abnormal motor; Affective/emotional behavior phenomena; Dialeptic; Aura; Mixed pattern	Abnormal motor [1. Hypermotor (Movement of the whole body, pelvic thrusting, out-of-phase limb movements, thrashing/grabbing/violent/kicking/punching movements. Whole body rigidity, whole body jerky movements, opisthotonic movements) 2. Partial motor (Head and neck: side to side, flexion/extension movements; limbs: flexion/extension, abduction/ adduction movements, jerking, facio-pharyngo-respiratory: coughing, gagging, hyperventilation)]; Affective/emotional behavior phenomena (Weeping, grimacing, screaming, moaning, grunting); Dialeptic (Coma-like state, no response to external stimuli, fall, flaccidity); Aura (Subjective feelings); Mixed pattern	N/A	1. There was no clear mention about the state of consciousness. 2. Small sample size (56 children in 2014 study and 82 patients in 2013 study)
Wadwekar/2014 (modified from Hubsch 2011)		Class 1 – dystonic attacks with primitive gestural activities. Class 2 – paucikinetic attacks with or without preserved responsiveness. Class 3 – pseudosyncope with or without hyperventilation. Class 4 – hyperkinetic prolonged attacks with hyperventilation, involvement of limbs and/or trunk. Class 5 – axial dystonic attacks. Class 6 – unclassified type.	They could classify 94.4% of their patients.	Small sample size of 54 patients. They did not investigate subjective symptoms (auras).
Gale/2015	Motor; Nonmotor		Patients with more dramatic seizure semiology (i.e., motor subgroup) appear to have lower	A cross-sectional study

Table 1 (continued)

Author/year	Classification system	Description	Significance	Pitfalls
Magaudda/2016	Hypermotor; Akinetic; Focal motor; Subjective symptoms	Hypermotor: tonic, clonic, or dystonic generalized movements. Akinetic: absence of movement. Focal motor: Seizures with focal motor movements. Subjective symptoms: experiential phenomena reported by the patients.	neurocognitive functioning. Patients with PNES with the least dramatic semiology appear to have fewer neurocognitive abnormalities (Hill & Gale [11]). Increased likelihood of motor seizure semiology when the males with PNES had abnormal scores on the somatic complaints subscale of the Psychological Assessment Inventory (PAI) Seizures were univocally assigned to a given class in 83.6% of seizures. Machine learning approach validated the classification.	Small sample size of 55 patients
Asadi-Pooya/2016 & 2017 & 2018 & 2019 (modified from Magaudda/2016)	Generalized motor; Akinetic; Subjective symptoms; Focal motor	1. Generalized motor: seizures mainly characterized by tonic, clonic, or dystonic generalized movements, tremors, rigor-like movements, whole-body rigidity, pelvic thrusting, pedaling, and/or side-to-side head movements. 2. Akinetic: seizures mainly characterized by unresponsiveness and the absence of movement. Minor distal limb tremors were permitted. 3. Subjective symptoms: seizures mainly characterized by experiential phenomena reported by the patients. Eyelid myoclonia and minor distal limb tremors were permitted. 4. Focal motor: seizures with focal movements.	Cross-cultural and international semiological differences exist. Neither the stereotypy nor the variability of PNES can or should be used as a marker of the disease and to differentiate PNES from epilepsy. Patients with generalized motor seizures demonstrated a trend for later age at onset and, more often, had loss of responsiveness with their seizures compared with patients who had nonmotor seizures.	Video-EEG studies were visually analyzed and classified by a single observer.

semiological features. One may decide to characterize this latter group according to the most prominent feature; but there may be conflicting ideas between different observers as for determining the most prominent feature during a complex seizure. Therefore, if a patient, for example, has an aura, and then falls on the ground and becomes motionless for a few minutes, and finally has a few generalized movements before his/her seizure ends, I would rather classify this as mixed semiology (i.e., aura plus akinetic plus motor). In addition to practical implications, this may also have potential therapeutic implications as described above. For example, if a patient has a mixed semiology consisting of an aura and then an akinetic seizure with drop attacks, the treatment

strategy might be different compared with that in a patient with a sudden akinetic seizure with drops (without auras) (see above). Therefore, it is preferable to classify the former example as mixed semiology (aura plus akinetic) rather than considering the most prominent feature (akinetic seizure in this example). Similarly, it makes sense to identify if the seizure is associated with loss of responsiveness or not. Many patients with PNES may experience loss of responsiveness during their seizures [22,33]. This has important clinical implications as loss of responsiveness is associated with higher likelihood of experiencing ictal injury in patients with PNES [22]. Altered responsiveness during PNES is hypothetically a marker of lower emotional resilience or ability

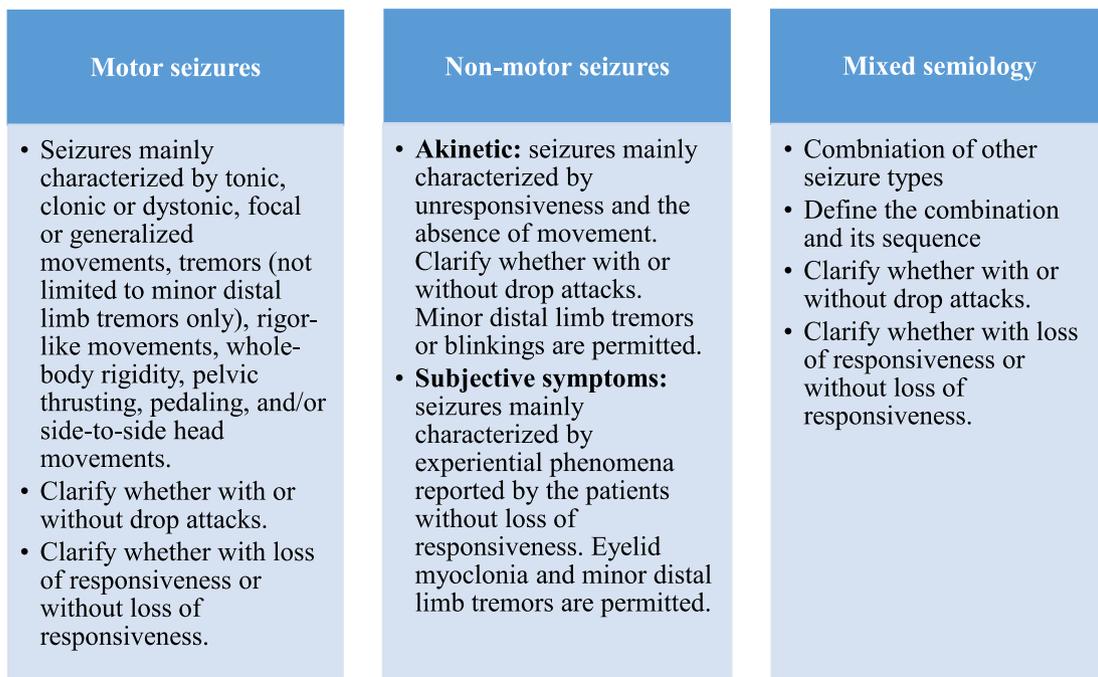


Fig. 2. A proposed semiological classification system for psychogenic nonepileptic seizures.

to tolerate emotions among these patients [33]. Hypothetically, it may be helpful to apply specific and appropriate management strategies for patients with PNES and loss of responsiveness in order to decrease the possibility of any associated consequences, including ictal injury [22]. It has been suggested that “emotion management” may be an important therapeutic target for patients with PNES and loss of responsiveness [33].

Lastly, it might be helpful to clarify whether the patient has drop attack or not with the seizures. It has been shown that patients with PNES may frequently present with dramatic features and, as a result, are at greater risk of receiving wrong diagnosis and unnecessary treatments [24]. One of the potential consequences of drop attacks are ictal injuries; these may include lacerations, fractures, burns, and dental injuries [24]. Therefore, clarifying and emphasizing the existence of drop attacks with the seizures may have important implications in the management process of the patients.

I should emphasize that any reliable semiological classification ought to be done after ictal recording during video-electroencephalography (EEG) monitoring of the patient. It has been shown that seizure witnesses often provide unreliable accounts of seizure semiology [34]. Where and when access to video-EEG monitoring is limited, acquisition of home video recordings of seizures may be helpful and decrease the need to rely on eyewitness reports [34,35].

Fig. 2 shows my proposed semiological classification system. In designing this proposal, I considered my clinical experience and also all the previous proposals, their strengths and their pitfalls. In addition, I considered this practical point that “the larger the number of scale categories, the greater the potential for disagreement; an unweighted kappa will be lower with more categories than with fewer categories” [36,37]. Therefore, I tried to keep the categories of this classification system to the minimum numbers required, according to the above discussions. The most important elements that I considered in designing this classification system were the following: motor vs. nonmotor seizures, akinetic seizures and auras, drop attacks, and responsiveness during the attacks. All these elements may have important clinical and therapeutic implications in the management process of patients with PNES (see above). It is noteworthy to consider that a classification system that considers too many clinical signs, especially if they are not specific to one class, may cause confusion for the users and lead to poor interrater reliability [36]. The study by Duwicquet et al. [36] highlighted that motor manifestation seems to be inadequate to distinguish different classes of PNES. Sometimes and particularly in the case of prolonged PNES, motor semiology may evolve during the same attack from one class to another, causing disagreement among different observers. They also reported that acceptable interrater agreement was observed in subjective seizures and dialeptic (akinetic) PNES [36]. Finally, they noted that consciousness (responsiveness) could be discriminant in PNES classification [36].

It is necessary to test this proposed semiological classification system for practicality (interrater reliability) and reproducibility in different cultures and across borders in future studies. A previous study demonstrated that many of the currently proposed and existing semiological classification systems have only a mild to moderate interrater reliability (κ from 0.44–0.68) for classifying PNES [36]. It is also necessary to investigate its clinical value with respect to therapeutic implications and prognostication. A universally accepted and appropriate semiological classification system for PNES may lead to better standardization of future studies and may also help in better understanding of the pathophysiological basis of this condition.

Declaration of competing interest

Honoraria from Cobel Daruo, Sanofi, and RaymandRad; Royalty: Oxford University Press (Book publication).

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None.

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