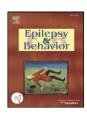
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Rating scale for psychogenic nonepileptic seizures: Scale development and clinimetric testing

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ABSTRACT

Our aim was to develop a clinimetric scale evaluating motor phenomena, associated features, and severity of psychogenic nonepileptic seizures (PNES). Sixty video/EEG-recorded PNES induced by suggestion maneuvers were evaluated. We examined the relationship between results from this scale and results from the Clinical Global Impression (CGI) scale to validate this technique. Interrater reliabilities of the PNES scale for three raters were analyzed using the AC1 statistic, Kendall's coefficient of concordance (KCC), and intraclass correlation coefficients (ICCs). The relationship between the CGI and PNES scales was evaluated with Spearman correlations. The AC1 statistic demonstrated good interrater reliability for each phenomenon analyzed (tremor/oscillation, tonic; clonic/jerking, hypermotor/agitation, atonic/akinetic, automatisms, associated features). KCC and the ICC showed moderate interrater agreement for phenomenology, associated phenomena, and total PNES scores. Spearman's correlation of mean CGI score with mean total PNES score was 0.69 (*P*<0.001). The scale described here accurately evaluates the phenomenology of PNES and could be used to assess and compare subgroups of patients with PNES.

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1. Introduction

Psychogenic nonepileptic seizures (PNES) are characterized by paroxysmal alterations in behavior, consciousness, sensation, and perception that resemble epileptic events but are not associated with abnormal electroencephalographic activity [1]. The estimated prevalence of PNES in the general population is 2 to 33 per 100,000. PNES account for 5 to 10% of neurology outpatients [2,3] and are commonly observed in tertiary referral centers, at frequencies of 5 to 20% among neurology outpatients and 10 to 40% among inpatient admissions [4]. PNES are attributed to a psychological cause and may be associated with different underlying psychopathological conditions such as dissociative disorder, somatoform disorder, and other general psychopathological disorders, with depression, anxiety, and obsession as dominating symptoms [5]. The possible influence of ethnicity, sociocultural status, educational level, and comorbidities (psychiatric disorders or epilepsy) on the objective manifestations of PNES is poorly understood.

Complex movements affecting different body regions characterize PNES. They can mimic the full spectrum of epileptic seizures [6] and may occur in both patients with epilepsy and those without epilepsy. Accurately differentiating PNES from epileptic seizures can be very difficult [7]. Although interrater reliability for the diagnosis of PNES by ictal video/EEG monitoring is only moderate [8], suggestion maneuvers have high sensitivity and specificity for the diagnosis of PNES [9]. At present, no scales are available to evaluate PNES phenomenology. A clinimetric scale would be very useful to study subgroups of patients with PNES with different ethnicity, sociocultural status, psychiatric disturbances, or comorbidities. A validated scale assessing psychogenic motor disorders was recently reported [10]. We drew inspiration from this scale and adapted it for the evaluation of PNES. Our aim was to develop and test the properties of a clinimetric scale to accurately describe phenomenology, anatomic distribution, severity, and duration of PNES.

2. Methods

Sixty-eight consecutive patients were diagnosed with PNES between 1998 and 2008 at twin University Epilepsy Centers located in Reggio Calabria and Catanzaro, Calabria, Southern Italy, sharing protocol, study design, and research objectives. Eight patients had

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both epileptic seizures and PNES and were excluded from the study. The remaining 60 patients with PNES included 46 women and 14 men in the age range 17 to 85 years (38.9 \pm 18.7). The diagnosis of PNES in these 60 patients was confirmed by positive responses to suggestion maneuvers. Thirty of these 60 patients were on antidepressants and benzodiazepines, 10 were taking only antidepressants, 5 only benzodiazepines, and the remaining 15 were not receiving treatment. No patient was taking antiepileptic drugs. Video/EEG recordings demonstrated the absence of ictal epileptiform activity or postictal slowing. The interictal EEG showed no epileptiform activity. One PNES was recorded for each patient. Sixty PNES were induced by different suggestion maneuvers: 58 by placing a colored patch on the neck patch [9], 1 by intermittent light stimulation [6], and 1 by infusion of intravenous saline [11]. A suggestion maneuver (placing a patch on the neck or pressing the arms or trunk) was able to inhibit 58 PNES at 1 to 4 minutes after onset. Two PNES ceased spontaneously. In all patients, the provoked seizures resembled a typical event. A witness familiar with the patients confirmed the similarity between induced and spontaneous seizures, by matching the induced episode with the patient's description. Patients were recorded according to a standardized video/EEG protocol (full-body view, closeup of face or head, hands, legs or trunk over the whole seizure course). These 60 videos served as source material for ratings. All patient provided written informed consent. The local ethical committees approved the study. The PNES scale rated six motor phenomena (tremor/oscillation, tonic, clonic/jerking, hypermotor/agitation, atonic/akinetic, automatisms), 13 body regions (upper face, lips/perioral, jaw, neck, head, left shoulder, right shoulder, left upper extremity, right upper extremity, left lower extremity, right lower extremity, pelvis, trunk), and 5 associated features (sphincteric incontinence, tongue biting, drooling, eye closure, hyperventilation, lament or crying). The following motor phenomena were analyzed: (1) shivering, tremor, or oscillation of one or more body regions (tremor/oscillation); (2) sustained muscle contractions or eye upward deviation (tonic); (3) jerky rhythmic or arrhythmic movements (clonic/jerking); (4) complex motor phenomena or large movements such as thrashing, flailing, writhing, punching, or kicking (hypermotor/agitation); (5) falling, staring, or inability to move (atonic/akinetic); (6) oral, gestural, and other apparently purposeful movements (automatisms). Responsiveness was evaluated during atonic/akinetic fits only. In part 1 of the scale (Table 1), each phenomenon was first rated as present (1) or absent (0). If present, the phenomenon was given a severity grade and duration factor ranging from 0 (lowest) to 4 (highest) (Table 2). Severity scores were given to both negative (atonic/akinetic) and positive (tremor, clonic/jerking, etc.) motor phenomena. The duration factor rated how long the phenomenon lasted during the observation

period and thus impacts the total points assigned to each phenom-

Table 2PNES scale: Severity, status, and duration factors.

Severity of phenomena	Status of function and associated features	Duration factor	
0, none 1, minimal 2, mild 3, moderate 4, severe	0, absent 1, present	0, none 1, <25% of the time 2, 25–50% of the time 3, 50–75% of the time 4, >75% of the time	

enon. Global Severity was assessed for each phenomenon, and was rated on a 0 to 4 scale. Global Severity scores reflected the predominant severity of a movement phenomenon, which was especially important if multiple different scores were assigned across regions. Part 2 of the PNES scale (Table 3) rated the status (presence/ absence) of associated features (sphincteric incontinence, tongue biting, drooling, eye closure, hyperventilation, lament, or crying). Associated features were included because they were not reflected by the localizable phenomena in the first part of the scale, but are routinely assessed in clinical practice and are important functionally. Total scores for phenomena, associated features, and their sum were calculated and documented in part 3 of the scale (Table 4). The Total Phenomenology Score was calculated as the sum of all severity and duration ratings of all phenomena across body regions. The Total Associated Feature Score was the sum of scores for the presence of sphincteric incontinence, tongue biting, drooling, eye closing, hyperventilation, lament, or crying. The Total Psychogenic Nonepileptic Seizure Score was the sum of the Total Phenomenology Score and the Total Associated Features Score. To test validity, we examined the relationship between the PNES scale and a nonspecific scale, the Clinical Global Impression (CGI) scale [12] (Section 1: Severity of Illness, Attachment 1). Three epileptologists (C.V., H.S.M., L.M.A.) independently rated 60 videotapes using both the PNES and CGI scales. The interrater reliabilities of the PNES scale for each rater were analyzed using AC1 statistic, Kendall's coefficient of concordance (KCC), and intraclass correlation coefficients (ICCs). The comparison of the CGI and PNES scales was evaluated with Spearman correlations that included the data from two observers.

3. Results

The most frequently observed motor phenomena were tremor/oscillation (40% of patients) and atonic/akinetic (37%). Responsiveness was compromised in most patients with atonic/akinetic fits. Clonic/jerking were observed in 28%, tonic in 22%, hypermotor/agitation in 8%, and automatisms in 8% of patients. The most frequently involved

Table 1PNES scale Part 1: Motor phenomena.

	Tremor/ oscillation	Tonic	Clonic/ jerking	Hypermotor/ agitation	Atonic/ akinetic	Automatisms
Upper face						
Lips/perioral						
Jaw						
Neck						
Head						
Left shoulder						
Right shoulder						
Left upper extremity						
Right upper extremity						
Left lower extremity						
Right lower extremity						
Pelvis						
Trunk (no pelvis)						
Global severity						
Duration factor						

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