Seizure: European Journal of Epilepsy 64 (2019) 20–22

Contents lists available at ScienceDirect

Seizure: European Journal of Epilepsy

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

Clinical letter

Temporally linked occurrences of epileptic and psychogenic nonepileptic seizures – Coincidental or pathogenically related?

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ARTICLE INFO

Keywords:
Psychogenic nonepileptic seizures
Epileptic seizures
Sequential
Simultaneous

1. Introduction

Patients with coexisting epileptic seizures (ES) and psychogenic nonepileptic seizures (PNES) pose a diagnostic challenge. In such cases, ES and PNES usually occur independently across separate times and demonstrate distinguishable semiologies. More enigmatic would be rarer clinical scenarios when ES and PNES occur closely together temporally, whereby the end of one seizure type is not easily demarcated from the start of the other. We present two cases with sequential occurrences of ES and PNES within close temporal proximity – one with ES followed by PNES; and, one with PNES followed by ES.

2. Case 1

A 57-year-old man has known co-existing ES and PNES documented during a prior video-EEG (VEEG) study. His ES were earlier characterized by an initial diffuse paresthesia, followed by "out-of-body" experience, visual distortion, as well as altered awareness associated with incomplete amnesia. Concurrent EEG coincided with ictal rhythmic theta activity emanating from the right temporal region. His PNES were also documented on VEEG, manifesting an initial diffuse paresthesia, followed by side-to-side head shaking, complex hand movements, delayed but intact following of commands, and non-epileptiform EEG correlate.

Across the 6-year span since his earlier VEEG evaluation, patient endorsed remission of his PNES while his ES frequency continued to range between weekly to monthly occurrences. Notably over this same time span, he developed an apparent new event type which demonstrated his habitual heralding paresthesia but was followed by abrupt loss of axial tone (leading to head drop or falling forward), behavioral arrest, and complete unresponsiveness/amnesia. Subsequent VEEG documentation of this new seizure type revealed ictal rhythmic 7–8 Hz activity emerging from and remaining restricted to the left temporal region. All captured ictal discharges were brief (lasting less than 20 s) coinciding with patient's paresthesia sensation, and were almost immediately followed by return of patient's baseline EEG background activity. Clinically, however, patient maintained partial loss of axial tone, unresponsiveness, eye closure, and amnesia for at least 2 to 4 min despite near-immediate recovery of a reactive posterior dominant rhythm (PDR) on the EEG post-ictally (i.e., neurophysiological evidence of wakefulness). These disproportionately prolonged periods of incapacitation (despite an awake EEG) was interpreted to be non-physiological, reflecting functional elaboration to internal stimuli in the form of subtle electrographic seizures; or PNES immediately following ES (Table 1).

3. Case 2

For 19 years, a 63-year-old man with military combat-related posttraumatic stress disorder (PTSD) has experienced stereotyped behaviors that were complex, prolonged, and strictly sleep-related. Per family, episodes begin with warfare-related nightmares, somniloquy, emotional overlay (hands holding his head distressfully) during which the patient cannot appropriately follow commands. After several minutes in this state, strong diffuse body shaking ensues.

During VEEG monitoring, patient underwent provocative induction involving placebo saline infusion, followed by photic stimulation and then hyperventilation maneuvers. About 3 min into the induction

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https://doi.org/10.1016/j.seizure.2018.11.014
Received 15 October 2018; Received in revised form 10 November 2018; Accepted 24 November 2018
1059-1311/ Published by Elsevier Ltd on behalf of British Epilepsy Association.

Table 1
Demographic characteristics, psychiatric diagnoses, and seizure histories of Case 1 and Case 2.

	Case 1 (ES → PNES)	Case 2 (PNES → ES)
Demographic characteristics		
Age (years)	57	63
Gender	male	male
Marital Status	married	married
Years of education	12	12
Employment	unemployed	retired
Disability-related benefits	yes – US military service connected benefit related to PTSD	yes – US military service connected benefit related to PTSD and MDD
Medical diagnoses		
	(1). Prior strokes (lacunar infarcts) (2). Coronary artery disease (3). Mild cognitive Impairment (4). Hypertension (5). Benign prostate hypertrophy	(1). Prostate cancer (2). Lung mass (undergoing further evaluations) (3). Hypertension (4). Osteoarthritis
Psychiatric diagnoses		
	(1). PTSD (2). Generalized anxiety disorder	(1). PTSD (2). MDD (3). Unspecified disruptive, impulse-control, and conduct disorder
Psychiatric treatments		
Number of psychotropic drugs	3	3
Prior or current psychotherapy	yes	yes
Seizure history		
Duration of seizure history (years) *	12	19
Number of anti-seizure medications	2	1
Baseline ES frequency**	weekly to monthly	monthly
Baseline PNES frequency**	remitted	unknown whether or not PNES occur independent of ES
Inter-ictal EEG	independent l eft and right temporal spikes	right temporal sharp waves.
Brain MRI	unremarkable	unremarkable

* Duration since time of the incipient seizure event.
** Seizure frequency at the time when temporally linked occurrence of ES and PNES was documented by video-EEG monitoring.
ES, epileptic seizures; PNES, psychogenic nonepileptic seizures; PTSD, posttraumatic stress disorder; MDD, major depressive disorder; MRI, magnetic resonance imaging.

procedure, patient exhibited low amplitude, rapid tremors of both arms and legs. At one point, he appeared visibly distressed and mumbled "I don't want to see this," while his eyes remained closed and he did not followed any verbal commands. Concurrent EEG coincided with a well-sustained PDR characteristic of the patient's alert baseline state. For the subsequent 10 min, the patient appeared to be quietly lying in bed with eyes closed. Notably, his eyes then abruptly opened with head deviating slightly to the right side. His left arm became extended, while his head turned toward the opposite side (left version). Diffuse tonic stiffening, followed by rhythmic clonic movements ensued thereafter. Electrographically, these latter behaviors coincided with an initial brief right hemispheric voltage attenuation, followed by ictal rhythmic 6–8 Hz theta within the right temporo-central region before becoming obscured by copious myogenic artifacts. The suggestible (inducible) nature in conjunction with discordantly intact PDR in setting of clinical unresponsiveness supported a functional etiology to the initial dys-cognitive phase of the event. A secondarily generalized tonic-clonic convulsive electro-clinical seizure followed; or ES shortly following PNES (Table 1).

4. Discussion

The coexistence of ES and PNES in a given patient has been described to manifest in two forms: (a) sequential, and (b) simultaneous [1]. In sequential presentations, ES emerge first, may become well treated, and PNES manifest later. In these cases, PNES may be driven by the emotional benefit of prior illness experiences related to ES [2]. Simultaneous epilepsy and PNES describes patients with both active ES and PNES. Simultaneous presentations of epilepsy and PNES have been described to be less common than sequential presentations [1]. Even rarer are clinical scenarios when ES and PNES occur closely together temporally, with only few case series reported in literature [3].

The temporal proximity of ES and PNES may be coincidental if the event frequencies of both seizure types were to be quite frequent. Regarding our cases, however, the original PNES event type appeared to have remitted in case 1. For case 2, the patient and family endorsed exclusive linkage of PNES evolving to ES, denying (to their knowledge) independent occurrences of either event types. Such timing of PNES in relation to ES strongly suggests a causal inter-relationship.

ES can induce psychological changes predisposing to conversion symptoms. Ictal activation of the temporal lobe may trigger memory contents and/or autonomic symptoms (e.g., epileptic orgasmia) reminiscent of prior sexual abuse, hence precipitating conversion symptoms in the form of PNES [3]. Ictal propagation to the frontal lobe may contribute to behavior disinhibition, hence lowering dissociative thresholds in patients with pre-existing predisposition. Conversely, the prodromal phase and/or ictal aura can emerge hours to seconds before overt clinical manifestation of an ES. Such pre-ictal symptoms, albeit subtle, may provide the "internal" stimuli activating an automatic execution of a learnt mental representation of seizures (the "seizure scaffold") [4]. Notably, the diagnosis of posttraumatic stress disorder (PTSD) was shared by both cases. PTSD may contribute to dissociative tendencies and reflects a "shaping factor" to each patient's seizure scaffold [4].

Proper recognition of lone PNES can be challenging. Compounding this diagnostic challenge is not only the estimation that 10% of patients with PNES also have ES, but that some patients may manifest occurrences of ES and PNES within close temporal proximity [3]. VEEG documentation of such perplexing events is indispensable, as witness' verbal accounts or outpatient video recordings of such events would unlike capture the full spectrum of temporally-related ES followed by PNES, or vice versa.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.seizure.2018.11.014.

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