Journal of Epilepsy and Clinical Neurophysiology

J Epilepsy Clin Neurophysiol 2010;16(1):23-25

Video-EEG in the pursuit of documented coexistence of epileptic and psychogenic nonepileptic seizures: How long is too long? – A case report

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ABSTRACT

Continuous Vídeo-EEG monitoring remains the gold-standard tool to confirm or disregard the diagnosis of epilepsy in selected cases in which a differential diagnosis is required and not clearly established in the basis of outpatient procedures. However, it may be a tiresome and stressful experience for patients and it is certainly an expensive test. Thus, we wonder how far (considering both financial and emotional costs) should we pursue the goal of documenting all suspicious events. An illustrative case is presented.

Keywords: Nonepileptic seizures, video-EEG.

RESUMO

Vídeo-EEG em busca da real necessidade da documentação em crises epilépticas e não-epilépticas psicogênicas: quanto tempo é demasiado longo? – Relato de caso

Monitorização continua com Video-EEG permanece como método de eleição no diagnóstico de epilepsia, em casos selecionados onde o diagnóstico diferencial não pode ser perfeitamente definido com base em procedimentos ambulatoriais. Entretanto, a monitorização contínua pode constituir uma experiência cansativa e estressante para os pacientes, além de custo envolvido. Considerando estes custos (emocional e financeiro) é especulada a real necessidade da documentação de todos os eventos suspeitos. Um caso ilustrativo é apresentado.

Unitermos: Crises não epilépticas, vídeo-EEG.

CASE

A 41 y/o female was referred to a tertiary epilepsy center for surgery evaluation. Seizure history started during childhood with a single convulsive episode with no clear etiology. No further seizures were noticed until the age of 38, when she presented with episodes starting with an uncharacteristic feeling ("discomfort") followed by loss of consciousness, accompanied by prolonged staring, marked unresponsiveness, occasional attempts of vocalization, and random movements of both arms lasting up to 2 minutes, after which she would fell confused and disoriented. She did fall during some events, leading to minor bruises, but no significant trauma. Episodes tend

to occur during wakefulness, but a few were reported out of nocturnal sleep. Seizures occur weekly and the longest seizure-free interval was 15 days. Family history was negative for epilepsy. Therapeutic doses of carbamazepine, phenytoin, clonazepan and valproic acid were used, with no significant changes on seizure frequency. The patient is a nurse technician in a major pediatric hospital, but has been on leave of absence several times due to frequent seizures. Additionally she complains of lack of motivation, insomnia, daytime drowsiness and weight gain. Seizures increased following her brother's brutal death, secondary to physical aggression. Neuro-exam was normal.

Diagnostic work-up by the time of referral included routine EEGs showing right anterior temporal spikes and an MRI showing right hippocampal atrophy. Patient was admitted for presurgical protocol. AEDs were tapered

Received Jan. 10, 2010; accepted Feb. 26, 2010.



Figure 1. Nonepileptic Psychogenic Seizure: unresponsiveness, flailing arms, head bouncing and eyes kept closed throughout most of the event.

and discontinued. Interictal EEG showed unilateral right temporal spikes. On Day 3 she had an episode, describe as "typical" by the patient herself and relatives, including unresponsiveness, mumbling and hypotonia lasting 3 minutes. EEG showed normal background activity and no epileptiform patterns, consistent with a psychogenic nonepileptic seizure (PNES). Due to known interictal EEG and MRI abnormalities monitoring was continued in the search of different seizure types. On Day 6 the patient presented a second "typical" episode, with similar clinical and eletrographic features, and also interpreted as a PNES. A third PNES, very similar in its presentation, was recorded on Day 8. Finally, on Day 9, after 216 hours of continuous monitoring she presented with a nocturnal seizure (1 AM), in which she kept her eyes opened, displayed mild orofacial and exuberant right arm automatisms, accompanied by marked unresponsiveness. EEG, showed right temporal rhythmic theta activity, consistent with a complex partial seizure (CPS).

Neuropsychological evaluation showed visual memory impairment. Psychiatric evaluation disclosed a depression disorder, an obsessive-compulsive anxiety disorder and obsessive-compulsive personality. She was put on clobazam and topiramate and discharged on Day 11. At six months follow-up she persisted with monthly spells that more likely represent PNES, generally associated with stressful episodes. A single, rather questionable, nocturnal episode (probably a CPS, by description) was reported by her husband.

DISCUSSION

Seizure identification by clinical description in temporal lobe epilepsy (TLE) tends to be highly accurate (94% accuracy, with a sensibility of 96% and specificity of 50%). Hence, epileptologists are overall good at detecting seizures based on clinical history, but may overcall PNES as epileptic seizures, explaining the low specificity¹. Clinical features on our patient suggested CPS. However, elements frequently seen on PNES were also present. The coexistence (i.e, epilepsy and PNES) is rather common (close to 30%) and represent 20% of the population seen at tertiary epilepsy

centers². Up to 8% of temporal lobectomy (TL) candidates may present with associated PNES³. Thus, the recording of her first PNES came as no surprise. Conversely, the likelihood of MTS patients present with exclusive PNES is possibly low. Out of 207 patients submitted to MRI studies for hearing loss, with no history of seizures, only 2 patients presented with MTS. Their cases were reviewed and both suffered from an epileptic disorder⁴. That explains our determination in documenting her epileptic seizures. After her third PNES, however, a question was posed, as to, for how long should we pursue this goal, knowing that VEEG monitoring can be a tiresome and overall costly procedure. The effort finally paid off on Day 9, with documentation of a legit CPS. Yet, a surgical option was denied, on the basis of her psychiatric comorbidity and the possibility of refractoriness of PNES. Benbadis et al discussed the difficult of disclosing the diagnosis of PNES in patients with MRI proven abnormalities and paroxysmal events, describing 4 cases of MTS patients in whom no epileptic seizures were documented at VEEG monitoring⁵. The outcome of PNES varies according to a number of variables and seizure remission alone may be not a measure of prognosis. In a recent paper only a third of the PNES patients achieved seizure remission and 42% of those remained "unproductive". Conversely, results on epileptic seizures control following TL in MTS patients are consistently encouraging⁷. Results on PNES outcome following TL are scarce.

From the academic and evidence-based standpoint documentation of epileptic and PNES in cases where both hypothesis are suspected remains the gold standard for diagnosis. In our case, however, this determination led to a few puzzling situations: a) how refractory are the patient's true epileptic seizures? b) Is she emotionally stable to undergo epilepsy surgery? c) What is the PNES prognosis on patients with epileptic and PNES in whom there is clear prevalence of the later? d) Should the patient be submitted to additional monitoring, in order to precisely determine the kind of residual seizures she is still presenting, while on her current treatment?

We believe further studies including this selected group of patients should be carried out before answers to these questions can be safely offered to patients and relatives.

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