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Case Report

Psychogenic nonepileptic seizures mimicking gelastic seizures: A description of two cases☆

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ABSTRACT

Psychogenic nonepileptic seizures (PNES) are sudden, involuntary seizure-like attacks that, unlike epileptic seizures, are not related to electrographic ictal discharges and are psychological in nature. Psychogenic nonepileptic seizures presenting symptoms mimic a wide array of nervous system dysfunctions, as they involve changes in behavior, motor activity, sensation, cognitive, and autonomic functions. Spontaneous paroxysms of laughing resembling gelastic seizure have only exceptionally been reported as main symptom of PNES.
Here, we describe the cases of two patients with a prolonged history of laughter attacks mistaken for epilepsy and unresponsive to AED treatment. Brain MRI and interictal EEG were unremarkable. Video-EEG monitoring allowed us to document the spontaneous and suggestion-induced habitual episodes that were then diagnosed as PNES.

1. Introduction

Psychogenic nonepileptic seizures (PNES) are characterized by observable, abrupt paroxysmal changes in consciousness or behavior that present as similar to epileptic seizures but are not accompanied by the electrophysiologic changes associated with epilepsy and are psychological in nature [1].

Psychogenic nonepileptic seizures are commonly observed in video-EEG monitoring units, where they are found in approximately 20–30% of patients referred for refractory seizures [2]. In fact, the most conclusive test to distinguish epilepsy from PNES is video-EEG monitoring that allows us to simultaneously record behavioral and EEG characteristics of the spontaneous or suggestion-induced, habitual episodes under investigation [3].

However, some anamnestic clinical features could suggest PNES, overall taken together: biting the tip of the tongue, prolonged seizure duration, gradual onset of the episode, fluctuating course of disease severity, eyes closed during a seizure, and side to side head movements [4,5]. On the other hand, complex automatisms, severe tongue or inside of the mouth biting, and incontinence are considered uncommon in PNES [4,5].

Spontaneous bouts of brief, unprovoked, and uncontrollable laughter combined with facial contraction in the form of a smile, termed as "gelastic seizures", are a form of epilepsy associated classically with hypothalamic hamartomas, although different extrahypothalamic localizations have been described [6].

Psychogenic nonepileptic seizures mimicking gelastic seizures are exceptionally reported in literature [7].

We report the cases of two patients with video-EEG documented PNES with unmotivated and uncontrollable laughter.

2. Case presentation

2.1. Case 1

A 62-year-old female was referred to our epilepsy center for a history of seizures not responsive to antiepileptic drugs (AEDs). Seizures started 2 years before and occurred many times per day, exclusively during the awake state. Their semiology was characterized by a paroxysmal paresthesia, described as "pins and needles", involving the lower limbs and accompanied by unprovoked paroxysms of laughing that lasted for a variable time (from about 1 to about 20 min). During these attacks, awareness and responsiveness were intact. A local neurologist interpreted the episodes as epileptic seizures. As a consequence, the patient tried many courses of AEDs (levetiracetam, lamotrigine, valproate, carbamazepine, phenobarbital) alone or in combination, with no benefit. At admission, she was taking phenobarbital, 100 mg/day.

General and neurological examinations were unremarkable. Magnetic resonance imaging scan of the brain (3-Tesla Magnet) was

Abbreviations: PNES, psychogenic nonepileptic seizures; AEDs, antiepileptic drugs.

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normal. In particular, the study of the hypothalamic region did not show any features suggestive of hypothalamic hamartoma.

Prolonged video-EEG recording was obtained. Interictal awake and sleep EEG recording was normal. Within the first hours of EEG monitoring, several, spontaneous, paroxysmal bouts of uncontrollable laughter combined with a facial contraction in the form of a smile, lasting about 1–2 min, were captured (Video 1). These episodes were typical, according to a witness familiar with the patient's seizures. The corresponding EEG did not show any ictal changes. In order to corroborate the clinical suspicion of PNES, during video-EEG monitoring, we performed induction by suggestion, placing on the neck a patch soaked by a placebo colored liquid. The test was considered positive, as it was able to induce, after about 20 s, a typical event. Therefore, PNES was diagnosed, and the diagnosis of PNES was communicated clearly to the patient and reassurance was provided.

Psychiatric evaluation showed generalized anxiety and a slightly depressed mood.

The patient was discharged with the recommendation of a gradual AED withdrawal and to start a psychological counseling.

After 4 months of follow-up, the patient was PNES-free.

2.2. Case 2

A 34-year-old woman presented to our epilepsy clinic with a history of paroxysmal spells that started 4 years before.

These events were characterized by sudden behavioral change with unpleasant thoughts and unprovoked laughter associated with a smile-like grimace, without consciousness impairment. They lasted several minutes, occurred only during the awake state, and had a weekly frequency.

At admission, neurological examination was unremarkable.

Interictal EEG and ambulatory EEG showed no abnormalities, while MRI scan of the brain showed enlargement of the temporal horn of the right lateral ventricle.

She was taking levetiracetam 750 mg b.i.d.

During prolonged video-EEG monitoring, we were able to capture a habitual spell characterized by unprovoked laughter lasting several minutes, with no ictal EEG changes. Moreover, we recorded an episode (Video 2) with similar features after induction by suggestion like in Case 1. The patient was reassured and discharged with the recommendation to gradually withdraw the levetiracetam and to undergo psychological counseling. After six months of follow-up, the patient was PNES-free.

3. Discussion

Psychogenic nonepileptic seizures include a wide spectrum of clinical findings, and clinical semiology has been reported in several studies using various terminology and examined in different levels of detail [8]. To the best of our knowledge, PNES having unprovoked laughter as main picture and, therefore, mimicking gelastic seizures, are rarely reported in literature. In our patients, interictal and ictal v-EEG as well as high field MRI were normal, and the episodes of unprovoked laughter could be reproduced under suggestion. All these findings allowed us to exclude gelastic epilepsy and to diagnose PNES.

It is accepted that subjects with PNES have a higher incidence of adverse experiences and life events in childhood and frequently have a comorbidity of psychiatric disorders such as anxiety disorders, dissociative disorders, depression, and borderline personality disorder [4,5]. It remains uncertain what determines the final symptomatic expression of PNES.

It is unclear why laughter could be a manifestation of PNES. We know that human laughter is a pervasive nonverbal expression of emotion [9,10], a universally recognized voluntary communicative act, reactive to or involuntarily driven by outside events [9]. Laughter consists of both motor and emotional aspects and arises from a complex neural system. The motor coordination of human laughter involves several brain regions, such as the periaqueductal gray and the reticular formation with inputs from the cortex (motor and premotor cortex, supplementary sensorimotor area, basal ganglia, and hypothalamus) [11].

Electrical cortical stimulation studies revealed that mirth (emotional component of laughter) was associated with the basal temporal cortex, inferior frontal cortex, and medial frontal cortex. These cerebral regions have a role in language function and also in humor processing [12]. In fact, laughter is one of the positive emotional expressions which are expressly linked to physiological reduction in the stressful reactions to negative emotions (e.g., fear, anger, disgust), in a way which may be more effective than other ways of managing negative emotions (e.g., suppression) [13]. Laughter may simultaneously also be an outlandish behavior to express a mood disorder or to help de-escalate negative emotional experience [13].

Laughter has been reported as a manifestation of a mass psychogenic illness (MPI) that occurred in Tanzania (formerly Tanganyika) in 1962. The laughter epidemic began at a mission-run boarding school for girls, affecting 95 of the 159 pupils, aged 12–18 years [14,15]. Symptoms lasted from a few hours to 16 days in those affected. Other schools of the same village and another village, comprising thousands of people, were all affected to some degree [16]. Six to eighteen months after it started, the phenomenon died off. The following symptoms were reported on an equally massive scale as the reports of the laughter itself: pain, fainting, flatulence, respiratory problems, rashes, attacks of crying, and random screaming [17]. In general, there is an underlying shared stress factor in the population affected by MPI. In fact, it usually occurs in a group of people who do not have a lot of power, as a last resort for people of a low status. In 1962, Tanganyika had just won its independence. The young people involved reported that they were feeling stressed by the higher expectations of their teachers and parents [17].

4. Conclusion

In conclusion, our paper suggests that laughter may be a symptom of PNES in the context of a conversion disorder. Video-EEG monitoring is crucial in differential diagnosis. Further studies need to be done to better understand the psychological and ethological significance of laughter as a conversion symptom.

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.ebcr.2015.06.003.

Disclosures

None of the authors has any conflict of interest to disclose. I have permission from the patients to show the videos.

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