■ Case vignette

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Epileptic Disorders

Dacrystic seizures in MRI-negative patients

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Dacrystic seizures (DS) are characterized by bursts of crying with stereotyped lacrimation, grimacing, sobbing sounds and/or sad facial expression. Multicentric studies have reported prevalence at between 0.1-0.13% [1, 2]. DS are frequently associated with hypothalamic hamartomas (HH) mainly when coexisting with gelastic seizures (GS), however, other types of lesions involving temporal, frontal or insular lobes have been described [3, 4].

We describe a patient with crying as a manifestation of his seizure, who had repeated negative magnetic resonance imaging (MRI) since his epilepsy started.

The patient was a 34-year-old, right-handed man without risk factors for epilepsy and normal physical examination, who at the age of 12 started with involuntary laughing events during his sleep, lasting for 10 seconds. The events disappeared after valproic acid was started. Two years after being seizure-free, valproic acid was suspended without recurrence of these events. At 30 years of age, he started with stereotypical episodes of a sudden sensation of tensing of his whole body, frequently accompanied by a rising epigastric sensation lasting for 10 seconds, which did not impair his awareness. Overtime, the frequency increased to daily without response to valproic acid. He was admitted to the Epilepsy Monitoring Unit and six seizures were captured. The events started with a sudden onset of crying, some of which were associated with facial grimacing and sobbing, lasting for 16-43 seconds. Ictal automatisms and post-ictal nose wiping with his right hand were seen. His awareness and language were not altered (*video 1*). The electroencephalogram (EEG) showed changes that lasted for 4-12 seconds after crying started and which were characterized by rhythmic theta activity over the right fronto-temporal region (F8-F4-Fp2) (*figure 1*). The late presentation of EEG changes may reflect a deep epileptogenic focus.

The hypothalamus has been proposed as the epileptogenic zone based on studies in patients in whom epileptic discharges arose and remained within the HH during DS [5]. However, patients with DS became seizure-free after lesionectomies in the temporal or frontal regions, therefore those regions seem to be involved [2, 4, 6]. In addition, patients without lesions can also have DS. We were able to identify 10 cases in the literature (*supplementary table 1*) [1, 3, 4, 6-8]. DS were usually reported with other clinical signs and other types of seizure in the same patient. Our patient presented GS only at the beginning of his illness. During his DS, automatisms were seen.

The ictal EEG abnormalities in MRI-negative patients are not typical and different patterns have been described. The localization of these abnormalities was more commonly seen in the frontal and temporal lobes, however, when the information was given in detail, these alterations occurred after the clinical manifestation had started. In a patient in whom there was intracranial implantation of subdural electrodes, DS occurred when the epileptiform activity involved the cingulate gyrus [4, 6]. Furthermore, direct electrocortical stimulation from the anterior insula, posterior orbito-frontal cortex and subcortical structures have provoked crying [9], suggesting implications in the process of crying. In fact, the neurobiology of normal crying is complex, involving a volitional system in the frontoparietal

VIDEO ONLINE

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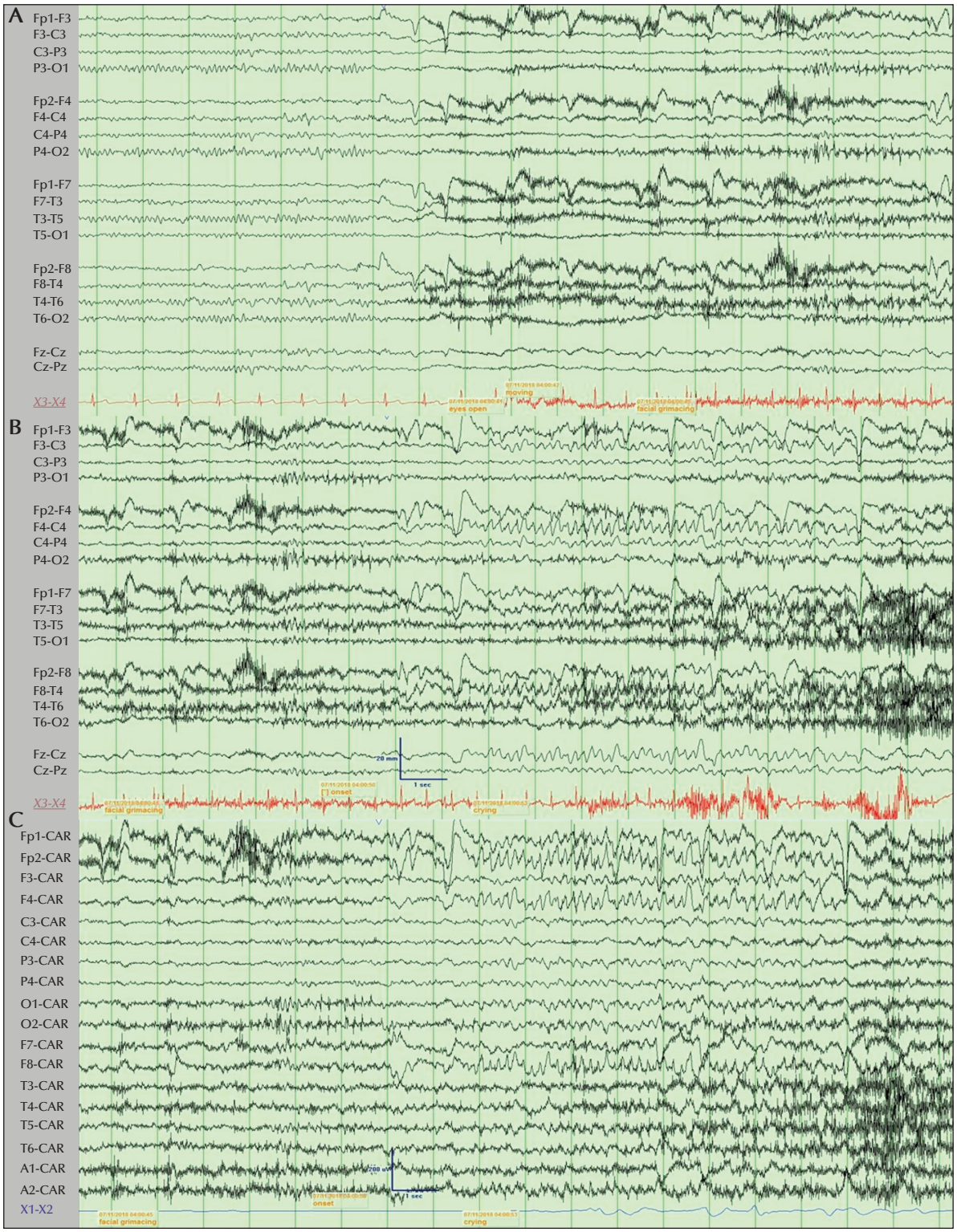

■ **Figure 1.** EEG showing rhythmic theta activity over the right frontotemporal region (F8-F4-Fp2) lasting for 4-12 seconds after the initiation of crying.

region that inhibits (through corticopontine projections) an emotional system between fronto-temporo-insular regions and subcortical structures (hypothalamus-periaqueductal grey-dorsal tegmentum), regulating emotional displays [10]. DS could be the result of direct involvement of the emotional system or loss of inhibition from the volitional system; therefore, it seems that DS do not provide clinical value in predicting the epileptogenic zone [1, 2, 4]. On the other hand, GS has been suggested to be semiologically differentiated from HH, and temporal and frontal regions based on natural laughter without emotion, natural laughter with emotion, and unnatural laughter (greater motor component) without emotion, respectively [11]. In our patient we believe that the epileptogenic zone is in deep structures, either the orbitofrontal or insular cortex. We were able to lateralize the seizure because of other clinical signs and the findings on EEG.

More than 80% of patients with lesional DS have drug-resistant epilepsy (DRE) [2, 4]. However, the rate of DRE in patients with MRI-negative is lower (60%). An explanation for the high rate of DRE could be that most reports are derived from tertiary epilepsy centres. Interestingly, up to 90% of patients with GS without lesions are free of seizures [12]. Our patient was discharged with higher doses of valproic acid and the addition of lamotrigine, which was started gradually. Two years later, the patient continues to be seizure-free. ■

Supplementary material.
Supplementary data and summary slides accompanying the manuscript are available at www.epilepticdisorders.com.

Disclosures.
None of the authors have any conflicts of interest to declare.

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Legend for video sequence

Patient laying down in bed with eyes closed:
00:00:11s: He suddenly opened his eyes and he tried to sit up.
00:00:18s: He started to cry with a sobbing sound and facial grimacing. No tears were seen. Then he said "I am okay, I am okay" when the nurse approached him. After that, he started to make other noises and rub his nose with the pillow.
00:01:38s: Finally, the nurse went back and asked him if he had had a seizure and he confirmed it. His language was not impaired.

Key words for video research on www.epilepticdisorders.com

Phenomenology: dacrystic seizure, crying
Localization: unknown
Syndrome: not applicable
Aetiology: unknown

TEST YOURSELF

(1) Regarding dacrystic seizures (DS), which of the following is correct?

A. Hypothalamic hamartomas are always present in patients with DS, therefore constituting the epileptogenic zone.

B. DS are frequently reported isolated (without other ictal manifestations)

C. DS may be present in MRI-negative patients.

(2) Which of the following structures have been implicated in dacrystic seizures?

A. Hypothalamus
B. Temporal lobe
C. Frontal lobe
D. All above

(3) Explain the probable physiopathology of dacrystic seizures.

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com.