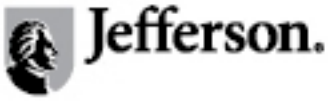

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Ictal crying

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Brief communication

Title: Ictal crying

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Abstract

Purpose: To describe a series of patients with ictal crying to estimate its occurrence and characterize the clinical features and the underlying etiology.

Methods: We retrospectively reviewed all the long-term video-EEG reports from Jefferson Comprehensive Epilepsy Center over a 12-year period (2004-2015) for the occurrence of the terms “cry” or “sob” or “weep” in the text body. All the extracted reports were reviewed and patients with at least one documented ictal crying at the epilepsy monitoring unit (EMU) were included in the study.

Results: During the study period, 5133 patients were investigated at our EMU. Thirty-two patients (0.6%) had at least one documented seizure accompanied by crying. Twenty-seven patients (26 women and one man) had psychogenic nonepileptic seizures (PNES) and five patients (0.1%) had epilepsy. Among patients with epileptic ictal crying, four patients had focal epilepsy (two had definite and two had probable frontal lobe epilepsy), while one patient had Lennox-Gastaut syndrome.

Conclusion: Ictal crying is a rare finding among patients evaluated at the EMUs. The most common underlying etiology for ictal crying is PNES. However, ictal crying is not a specific sign for PNES. Epileptic ictal crying is often a rare type of partial seizure in patients with focal epilepsy. Dacrystic seizures do not provide a reasonable clinical value in predicting localization of the epileptogenic zone.

Introduction

Epileptic seizures with a sudden burst of crying with no apparent cause are called dacrystic seizures. The term was first proposed by Offen et al. in 1976 [1]. Dacrystic seizures are rare and the literature on this seizure type is scarce [2]. Semiologic features include stereotyped lacrimation, grimacing, sobbing, sad facial expression, and/or a subjective feeling of sadness [2]. The possible symptomatogenic zone for dacrystic seizures is located in the hypothalamus. The mechanism of symptoms can be either direct irritation or a release phenomenon because of the loss of inhibition by regulatory cortex [2, 3]. In this study, we describe a series of patients with ictal crying to estimate its occurrence and characterize the clinical features and the underlying etiology.

Methods

We retrospectively reviewed all the long-term video-EEG reports from Jefferson Comprehensive Epilepsy Center over a 12-year period (2004-2015) for the occurrence of the terms “cry” or “sob” or “weep” in the text body. All the extracted reports were reviewed and patients with at least one documented ictal crying at the epilepsy monitoring unit (EMU) were included in the study. For each included patient, the data was reviewed from the electronic medical records, EMU report, and neuroimaging records. The videos of ictal crying were reviewed when they were available. This study was conducted with the approval by Thomas Jefferson University Institutional Review Board.

Results

During the study period, 5133 patients were investigated at our EMU. Thirty-two patients (0.6%) had at least one documented seizure accompanied by crying. Twenty-seven patients (26 women and one man) had psychogenic nonepileptic seizures (PNES) and five patients (0.1%) had epilepsy. Table 1 shows the clinical characteristics of the patients with epileptic ictal crying. The videos were available for three patients with epilepsy. Duration of crying in patients with epilepsy was 5-42 seconds (Table 1). Even in patients in whom the video was not available, the seizure itself lasted 10-20 seconds. We also reviewed videos for six patients with PNES. Ictal crying in patients with PNES lasted 69-339 seconds. This was intermittent in two patients, with periods of body shakings without crying in between episodes of crying. Seizures in these patients with PNES lasted 69-855 seconds.

Discussion

Ictal crying is a rare finding among patients evaluated at the EMUs. The frequency of ictal crying at our center was 0.6% and the frequency of epileptic ictal crying was 0.1%. In a previous multicenter study of the long-term video-EEG monitoring units [2], dacrystic seizures (epileptic ictal crying) occurred in 0.13% of all the patients admitted to the centers, which is concordant with our finding. The most common underlying etiology for ictal crying in our study was PNES. In a previous review article [3], the authors concluded that ictal crying can be used to differentiate PNES from epilepsy. They concluded that this sign has a sensitivity of 4% to 37% and a specificity of 100% to confirm the diagnosis of PNES, based on four controlled studies. However, in the current study we observed that ictal crying is not a specific or pathognomonic sign for PNES. This statement is also verifiable by the presence of other publications about

dacrystic seizure in the literature [4-9]. However, presence of ictal crying should seriously provoke the suspicion for PNES, as this sign is far more common among these patients. Ictal crying in patients with PNES is often longer in duration compared with patients with epilepsy and may be intermittent. Admission at a specialized center with expertise in making the differential diagnosis of seizures and performing video-EEG monitoring with ictal recording may settle the diagnosis in the affected patients.

Among our patients with epileptic ictal crying (i.e., dacrystic seizure), four patients had focal epilepsy (two had definite and two had probable frontal lobe epilepsy), while one patient had a generalized epilepsy syndrome (i.e., Lennox-Gastaut syndrome). Three of our patients had crying during their tonic seizures and two patients cried during complex partial seizures. Crying during tonic seizures is not necessarily an ictal phenomenon and could be a reaction to the pain inflicted by the severe muscle spasm, as one of our patients described (patient #2), or an emotional response to the seizure itself. We can conclude that dacrystic seizure is usually a rare type of partial seizure in patients with focal epilepsy possibly arising from frontal lobes. This is in contrast to most previous reports that temporal lobes were responsible for the seizure onset in patients with epileptic ictal crying [1, 2, 4-7].

In previous studies [2], dacrystic seizures in combination with gelastic seizures tended to occur in younger patients and were often associated with hypothalamic hamartoma. In a previous study [2], the authors investigated 15 patients with dacrystic and gelastic seizures (from their series and also from the literature); 14 patients (i.e., 93.3%) had a hypothalamic hamartoma. One case report in the literature described gelastic-dacrystic seizures in association with perinatal asphyxia and bilateral gliotic lesions in the parietotemporal areas [8]. In our study, one patient had dacrystic and gelastic manifestations during her seizure, but she was a 54-year-old adult with a normal

MRI. Another patient also had crying and giggle during her seizures, but an MRI was not available. In the group of 15 patients with dacrystic seizures without gelastic seizures in the same previous study [2], the authors found a cortical lesion in 12 patients, no underlying lesion in two patients, and white matter lesions in one patient. Cortical lesions were located in the anterior temporal lobe in eight patients (six patients had mesial temporal sclerosis and two had a tumor). In our study we could identify one definite imaging brain lesion, which was a frontal focal cortical dysplasia. Pathological crying has been described previously with lesions in the frontal cortex [10]. Pathological crying could be due to disrupted pathways that arise in the motor areas of the cerebral cortex that descend to the brainstem and inhibit a putative center for crying [4]. However, dacrystic seizures in patients with hypothalamic lesions could be secondary to the activation of excitable neurons in the autonomic hypothalamic centers [2]. In contrast to the previous study [2], we did not observe mesial temporal sclerosis among our patients. As a result, dacrystic seizures do not seem to provide a reasonable clinical value in predicting localization of the epileptogenic zone.

The major limitations of this study are its retrospective design and the small sample size. Besides, there is a potential for sampling bias due to the limited search terms used for identifying ictal crying. We did not have access to many of the videos to verify the findings ourselves, but all the videos were reviewed by the board certified epileptologists working at this institution.

Conclusion

We add to the literature on ictal crying by providing additional information on its differential diagnosis, frequency, semiology, and etiology. Ictal crying is a rare finding among patients evaluated at the EMUs. The most common underlying etiology for ictal crying is PNES.

However, ictal crying is not a specific or pathognomonic sign for PNES. Epileptic ictal crying is often a rare type of partial seizure in patients with focal epilepsy. Dacrystic seizures do not provide a reasonable clinical value in predicting localization of the epileptogenic zone.

Conflict of interest

Ali A. Asadi-Pooya, M.D. and Dale Wyeth report no disclosures.

Michael R. Sperling, M.D., Consulting: UCB Pharma; Research: contracts with Thomas Jefferson University, Eisai, UCB Pharma, Sunovion, SK Life Sciences, Marinus, Lundbeck, Medtronics, Visualase, Accorda, Upsher-Smith, Brain Sentinel.

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