

Case Report

Crying seizures without tears and amygdaloid lesions: a case report

Xin Xu, Xin-Guang Yu, Zhi-Qi Mao, Zhi-Qiang Cui, Long-Sheng Pan, Shu-Li Liang, Zhi-Pei Ling

Department of Neurosurgery, Chinese PLA General Hospital, Beijing, China

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Abstract: Backgrounds: Crying seizures is a rare type of seizure. Some scholars believe that crying seizures are mainly associated with lesions in the anterior temporal lobe, typically the non-dominant hemisphere, which has close connections with negative emotions. However, the role of amygdaloid lesions in crying seizures is not fully understood. Methods: By a patient with crying seizures and lesions in the amygdaloid nucleus who displayed an outburst of crying without tears and synchronous discharges on EEG, to understand crying seizures associated with amygdaloid nucleus lesions. Conclusion: The distorting and fearful face displayed during the episodes of crying seizures may be associated with functional disorders of the amygdaloid nucleus. The patient described in this study can shed new light on our understanding about the role of the amygdaloid nucleus in crying seizures and the feeling of fear.

Keywords: Crying seizures, amygdaloid lesions

Introduction

Crying seizures is a rare type of seizure [1] that involves a sudden burst of energy, usually in the form of crying. According to statistics derived from 5 third-grade epilepsy centers in the US and Germany, the incidence of crying seizures is 0.06%-0.53% among all seizures, with an average of 0.13%. Some scholars [1-8] believe that crying seizures are mainly associated with lesions in the anterior temporal lobe, typically the non-dominant hemisphere, which has close connections with negative emotions [1-5]. Blumberg and his colleagues [8] conducted an investigation involving the etiology of crying seizures and found that the seizures accompanied with outbursts of crying and laughing stemmed from hypothalamic hamartomas; seizures with crying, but no laughing, originated from the cortex (anterior temporal lobe). Blumberg and his colleagues [8] reported four crying seizures patients who exhibited crying and no laughter, and were all affected in the left temporal region. Three of the patients had left hippocampal sclerosis and one had a glioma in the left temporal lobe [8]. The role of amygdaloid lesions in crying seizures is not fully

understood. This study describes a patient with crying seizures and lesions in the amygdaloid nucleus who displayed an outburst of crying without tears.

Case presentation

The patient surnamed Liu was 51 years of age and born in Anhui Province, China. Her mother was healthy during pregnancy and the patient was healthy at the time of birth. She had from paroxysmal headaches since 2000 (46 years of age), which were aggravated by anger or emotional changes. The patient displayed her first outburst of crying in September 2014 (50 years of age), followed by a higher frequency of crying 1 month later. She received treatment for a mental disorder until an unexpected syncope episode in the afternoon of 8 December 2015, with loss of consciousness and body stiffness. The symptoms abated after a few minutes, but paroxysms of involuntary and uncontrollable crying ensued. Each episode lasted for approximately 30 sec and the time interval between 2 episodes was approximately 1 min. During the episodes, her head turned to the right and the right side of the body went stiff

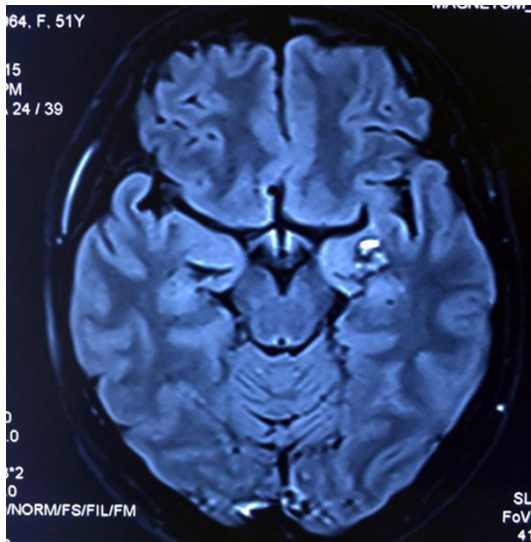


Figure 1. Cavernous hemangioma near the left amygdaloid nucleus by MRI.

with twitching. The symptoms resolved several minutes later. The patient showed several paroxysms of involuntary and uncontrollable crying with twitching the next day (> 9 times) and psychiatric symptoms, such as biting and beating people. The patient could not recall any of the above events, which occurred repeatedly and intermittently over the next 3 months. An evaluation was performed before epileptic surgery after admission to our hospital on 15 January 2016.

Imaging examination

A MRI indicated a cavernous hemangioma near the left amygdaloid nucleus (**Figure 1**).

Video EEG monitoring

Low-to-moderate amplitude α waves occurring 9-10 times per sec were observed in the rear region bilaterally when the patient was awake with her eyes closed; this rhythm was inhibited with the eyes open. Stages I-IV sleep were observed, and in stage II spindle waves occurring 12-14 times per sec in basic symmetry were noted in the frontal lobes bilaterally and in the central region. More mixed slow waves occurring 2-7 times per sec were seen in the left temporal lobe. Eye movements occurred at a higher frequency when awake (as high as 4 times per sec).

Intermittent stage

1. Moderate-to-high amplitude mixed slow waves occurring 2-7 times per sec were seen per-

sistently in the left temporal leads (MI-F7-T3). 2. Moderate-to-high amplitude spike waves-slow waves were seen in the left temporal leads (MI-F7-T3) propagating to the left frontal region (Fp1-F3; **Figure 2**).

Acute stage

During the 3-day monitoring, several episodes of seizures occurred, which manifested as sudden outbursts of crying with facial twitching. The patient, with eyes open, made no response to calls, but showed behaviors of fear or avoidance under stimuli. The crying was extremely loud, but without tears and started and ended suddenly. Each crying lasted for 20-30 sec, occurring once on the first day, twice on the next day, and at higher frequencies starting from the morning of the third day (once every 20 sec, with an interval of 20 sec). The paroxysms lasted for approximately 30 min and the patient would occasionally shout (**Figure 3A-D**). The symptoms resolved after the intramuscular injection of a stabilizer. The EEG revealed muscle artifacts at the beginning of or during the episodes. After the end of the episodes, a slow wave rhythm was found in the left temporal region (**Figure 4**). An electrocardiogram (ECG) showed accelerated heart beats after the episode. The heart rate was 60 and 72 beats/min during sleep and while awake, respectively. At the beginning of the episode, the heart rate was 120 beats/min, which was increased by 60% compared to the intermittent stage.

Treatment

The patient was diagnosed with secondary seizures in the left temporal lobe based on the position of the lesion and abnormal epileptiform discharges on EEG. EEG monitoring was performed in the cortex and deep brain during surgery. The patient underwent resection of the left hippocampal head, amygdaloid nucleus lesion, and anterior temporal lobectomy. The patient was pathologically-confirmed to have a cavernous hemangioma and 300 mg of oxcarbazepine was administered 3 times a day. No crying seizures occurred after surgery.

Discussion

Relationship between crying seizures without tears and brain lesions

In existing reports, crying seizures are most commonly seen in patients with brain tumors,

Crying seizures without tears

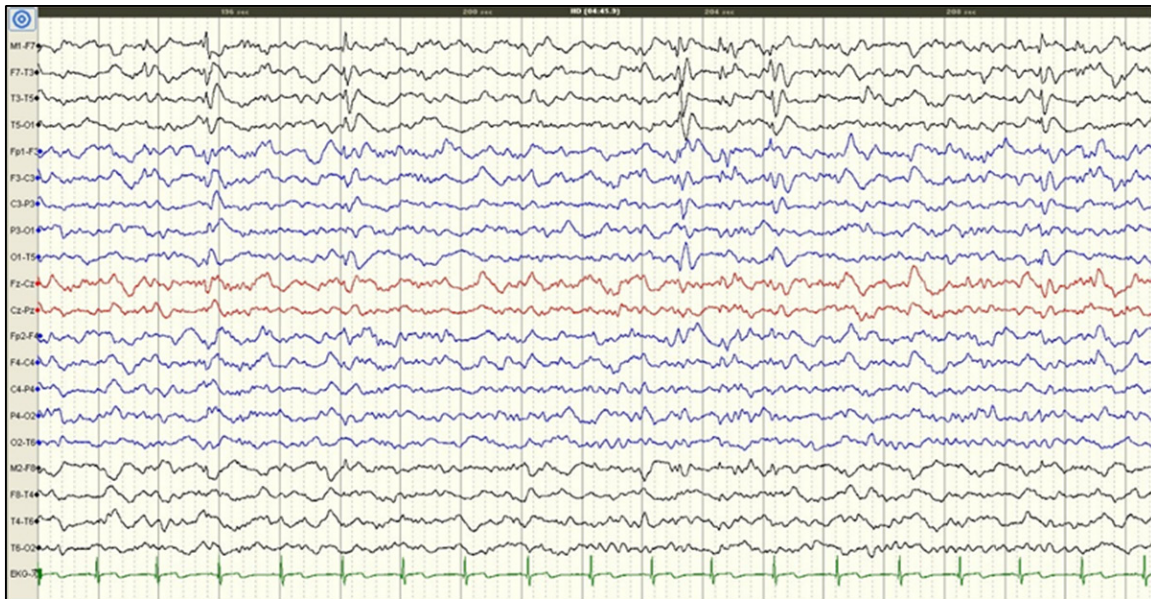


Figure 2. Epileptiform discharges during the intermittent stage in the left temporal region, especially in the anterior and middle temporal lobes.



Figure 3. A-D. Facial twitching in the acute stage.

vascular malformations, hippocampal sclerosis, and cerebral infarction. A few patients also exhibit crying seizures without apparent brain lesions. In our case, crying seizures were associated with a cavernous hemangioma, as has been previously reported [6, 7].

Relationship between crying seizures without tears and the position of the lesion

Many authors have reported that crying seizures are associated with lesions in the anterior temporal or frontal lobe (non-dominant hemisphere) [1-5]. Blumberg and his colleagues [8] studied the etiology of crying seizures and found that the seizures accompanied with out-

bursts of crying and laughing originated from a hypothalamic hamartoma; seizures with crying but no laughing originated from the cortex (anterior temporal lobe). Blumberg and his colleagues [8] reported four crying seizure patients who exhibited crying and no laughter, and were all affected in the left temporal region. Three of the patients had left hippocampal sclerosis and one had a glioma in the left temporal lobe [8]. Crying seizures associated with amygdaloid nucleus lesions are rarely reported. The patient described herein had amygdaloid nucleus lesions in the left dominant side, and the non-dominant hemisphere was not particularly prone to the lesions, as reported by Blumberg et al [8].

Figure 1 consists of two panels, A and B, showing axial brain MRI scans. Panel A is a T2-weighted scan showing a small, bright, hyperintense lesion in the left temporal lobe. Panel B is a corresponding axial scan showing a color-coded lesion in the same location, indicated by a white arrow. The color scale ranges from red (low) to yellow (high). The lesion is located in the left temporal lobe, near the lateral ventricle.

ed and ended suddenly, always in a fixed pattern, but without evidence of motor seizures. According to the medical history presented by the relatives, the patient had generalized tonic-clonic seizures secondary to the paroxysms of crying. Moreover, the patient would show signs of fear and cowering with her eyes open, which are typical features of seizures [9]. During the episodes, the heart rate increased from 72 to

120 beats/min, likely due to fear or suffering. The frequency of episodes increased with the dose. There was 1 episode of seizures on the first day and 3 episodes on the second day, with each lasting 20-30 seconds. On the third day, the seizures were aggravated both in frequency and duration (30 min). The symptoms resolved after a stabilizer was administered. The above symptoms were relieved by medication in a dose-dependent manner. The patient was diagnosed with crying seizures without tears.

Mechanism of crying seizures without tears

The distorting and fearful face displayed during the episodes of crying seizures may be associated with functional disorders of the amygdaloid nucleus. The periphery of the amygdaloid nucleus receives sound signals from the thalamus to the cortex and the sensory signals reach the sensory cortex. These signals are then sent to the central amygdaloid nucleus, which control fears [10, 11]. According to functional MRI (fMRI), the amygdaloid nucleus is the site where the fear is produced [12] (**Figure 5**). Our case had crying seizures without tears due to lesions in the periphery of the amygdaloid nucleus. This finding was probably caused by an abnormal electrical discharge in the brain that affects the functional region of the amygdaloid nucleus. As a result, the specific type of seizure (with or without tears) was highly related to amygdaloid nucleus function. That is, the abnormal electrical discharge resulting from the lesions of the amygdaloid nucleus led to paroxysms of crying and facial expressions of fear and pain. The position of the lesion was highly consistent with the activated region of the amygdaloid nucleus in fMRI under pictorial stimuli.

Clinical significance

Non-motor symptoms during acute stage deserve attention and timely treatment. Our patient was mistakenly diagnosed with a mental disorder. The clinical features of our patient can be summarized as follows: (1) The episodes started and ended suddenly, lasting for dozens of sec to several min and in a fixed pattern. (2) The patient cried out loud and painfully with her eyes open but without tears, which are important diagnostic features of epileptic seizures. Patients with non-epileptic seizures usually have their eyes shut and shed tears, with

increased then decreased vocalization [9]. (3) The heart rate increased during the episodes. (4) After the episodes, the patient was semi-conscious or had signs of mental aberrations [8]. For patients with simple crying seizures, the lesions may be localized to the temporal lobes or the amygdaloid nucleus based on the beginning and ending of seizures and the existing literature.

To conclude, the patient described in this study can shed new light on our understanding about the role of the amygdaloid nucleus in crying seizures and the feeling of fear.

Disclosure of conflict of interest

None.

Address correspondence to: Zhi-Pei Ling, Chinese PLA General Hospital, 28 Fuxing Road Beijing (Wukesong), China. Tel: 13811895827; E-mail: zhi-peilxx@163.com

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