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Letter to the Editor

A case of right orbitofrontal epilepsy featuring ictal swearing

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Dear editor,

Ictal swearing in focal epilepsy is rare and only a few reports are available. Previous studies have reported a variety of ictal speech automatisms, which frequently have been associated with seizures originating from the temporal lobe [1], and more often in seizures originating from the nondominant hemisphere [2]. However, ictal swearing as a rare phenomenon of speech automatisms, the localizing value and the mechanism underlying ictal swearing have not been well determined. In this article, we present a case with swearing during seizures arising from the right orbitofrontal cortex, confirmed by neuroimaging and stereotactic electroencephalography (SEEG) monitoring.

1. Case report

A 36-year-old, right-handed man, was referred for pharmacoresistant epilepsy with seizures occurring every night. His seizures started at the age of 26 years old, refractory to several antiepileptic drug regimes. His seizures attacked with no aura, the majority of seizures were characterized by sudden awakening and quickly followed by swearing and repetitive purposeless movements (right upper limb). There was no history of perinatal complications, febrile convulsion, CNS infection or trauma. Family history was also uneventful.

Scalp video-EEG (VEEG) monitoring revealed the interictal spikes were located in the left temporal and frontal region (SPH-L, F7, T3, F3), the spikes appeared not frequently. After five days of VEEG, eight stereotypical seizures were recorded, one of which evolved into a generalized tonic-clonic seizure. Ictal EEG consistently showed low amplitude theta frequency activity in right temporal region followed by diffuse spike–wave discharges. A 3.0 T MRI disclosed no epileptogenic lesion on T1 and T2 imaging. T2 fluid-inversion recovery (FLAIR) MR imaging revealed a suspicious high intensity signal in the posterior part of the right gyrus rectus (Fig.1a). Fluorodeoxyglucose positron emission tomography (FDG-PET) imaging showed focal hypometabolism in the same area (Fig. 1b). Voxel-based morphometry (VBM) analysis showed abnormal high signal in the posterior part of right gyrus rectus (Fig. 1c). The MEG examination (Elekta Neuromag Vector View 306 Channel Meg) showed the interictal spike dipoles were located in the left temporal lobe, insular cortex and right dorsolateral frontal cortex. Considering there were only a few spikes with no consistent localization, the test was interpreted as an inconclusive result.

Through a comprehensive analysis on various data including semiology, scalp EEG and neuroimaging diagnosis (MRI, PET, MEG, VBM), an intracranial evaluation was performed using SEEG. Ten depth electrodes were implanted to sample targets in bilateral hemispheres. Three electrodes were implanted in left insular cortex, anterior cingulate cortex and orbitofrontal cortex. Three electrodes were implanted in the left temporal lobe and hippocampus. In the right hemisphere, four electrodes were implanted in the posterior part of the right gyrus rectus, anterior cingulate cortex, insular cortex, temporal cortex and hippocampus. Intracranial EEG monitoring showed interictal spikes were located in the left temporal lobe and right gyrus rectus. EEG onsets were characterized by paroxysmal fast activity arising from the right gyrus rectus, followed by rhythmic activities in the right dorsolateral prefrontal cortex and left temporal region (Fig. 1f). There were eleven seizures were recorded via SEEG, ten of them (90.9%) featured ictal swearing, the mean delay from EEG onset to appearance of swearing was 25.4 s. When the discharge involved the right dorsolateral frontal cortex and the left temporal region, the swearing and repetitive limb movements appeared. Cortical stimulation (0.2 ms width pulse, 50 Hz, 1–6 mA, 3 s duration) of electrode contacts in the EEG onset region did not elicit swearing but repetitive purposeless movements in right upper limb which was consist with stereotypical seizure. The patient underwent a resection of the anterior part of right prefrontal lobe including the frontal pole, gyrus rectus and subgenual cingulate cortex (Fig. 1d). Histopathology demonstrated a focal cortical dysplasia (FCDIIb) (Fig. 1e). The patient has been seizure free for 8 months at last follow up.

2. Discussion

We present a patient with ictal swearing and repetitive motor automatisms due to non-dominant right frontal lobe epilepsy. SEEG electrodes were implanted in the bilateral frontal, temporal and insular lobes, the result demonstrated that the EEG ictal onset was localized in the right gyrus rectus, when the seizure activity involved the right dorsolateral prefrontal cortex and left temporal region, the ictal swearing appeared. Ictal swearing is probably related to the recruitment of an orbitofrontal–temporal networks activated by the epileptogenic zone. The favorable post-surgical outcome is further proof of

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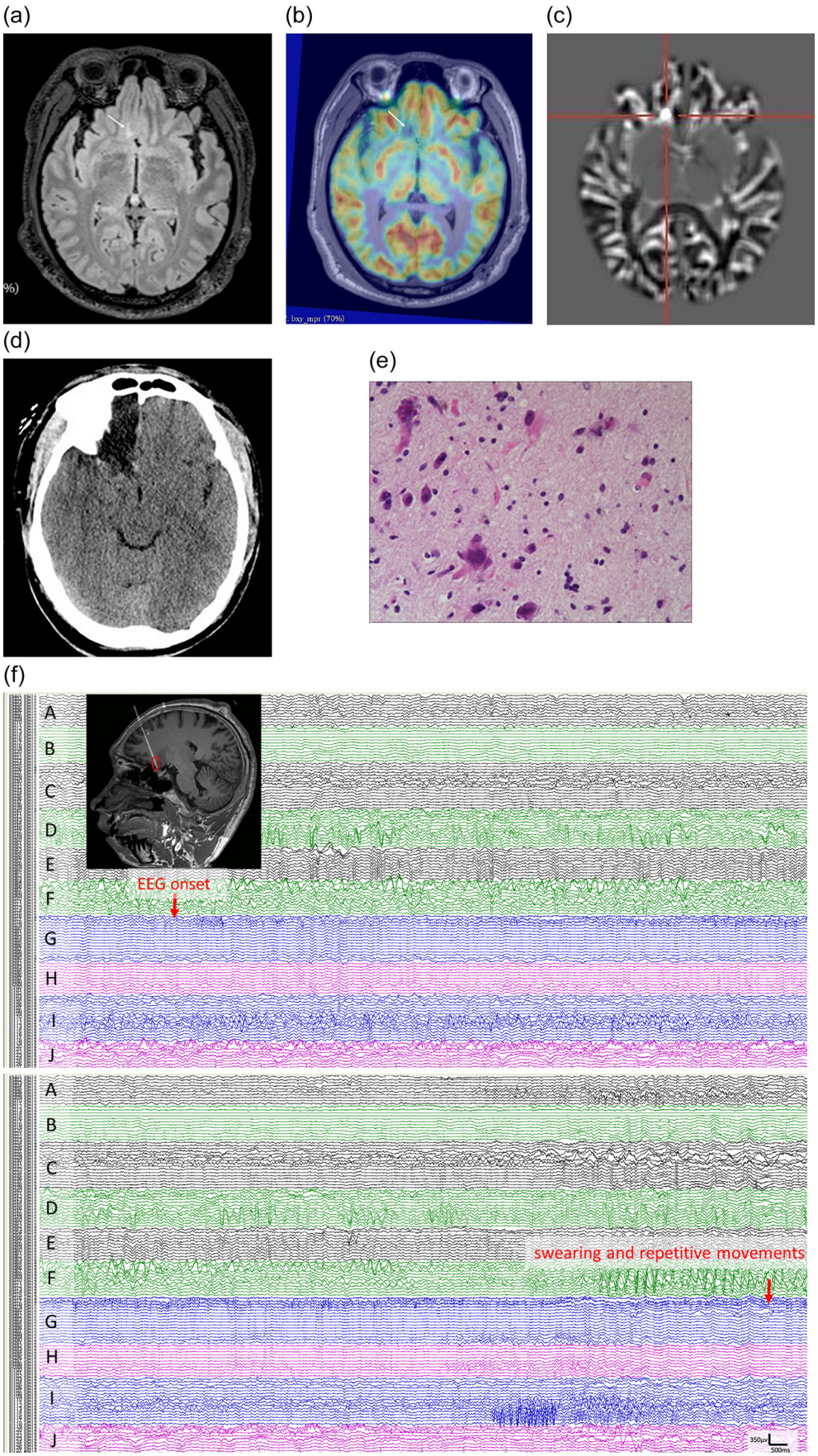

Fig. 1. (a) Axial sections of T2 flair MR showing a high intensity signal in posterior part of the right gyrus rectus. (b) PET imaging showing focal hypometabolism in the same area. (c) Voxel-based morphometry (VBM) analysis showing abnormal high signal in posterior part of the right gyrus rectus. (d) Postoperative CT imaging of the patient. Anterior part of the right frontal lobe including the frontal pole, gyrus rectus, subgenual cingulate cortex was resected. (e) The postoperative pathology findings: focal cortical dysplasia (FCDIIb). (f) SEEG of an ictus. Above: Ictal low voltage fast activities originate from the right gyrus rectus recorded by the electrode G (G1-G5, arrow). Below: When the discharge involves the right dorsolateral frontal and left temporal regions, the swearing and repetitive limb movements appears (arrow). The delay from EEG onset to appearance of swearing is about 20s.
The legend of SEEG location: A: L (mFG)-IC; B: L (mFG)-ACC; C: L (iFG)-OG; D: L (sTG)-Hip; E: L (mTG)-TP, F: L (mTG)-FG; G: R (mFG)-GR; H: R (mFG)-ACC; I: R (mFG)- IC; J: R (sTG)- Hip.
mFG: middle frontal gyrus, iFG: inferior frontal gyrus, sTG: superior temporal gyrus, mTG: middle temporal gyrus, ACC: anterior cingulate cortex, IC: insular cortex, GR: gyrus rectus, OG: orbital gyrus, TP: temporal pole, Hip: hippocampus, FG: fusiform gyrus.

seizure onset zone localization. To our knowledge, this is the first surgically proven report of ictal swearing originated from right gyrus rectus.

Reports of ictal swearing are rare, the lateralization value of this complex speech automatism remain uncertain. The latest case series was reported by Birca et al. (2013) reported eight patients with ictal swearing seen between 1974 and 2012. Focus localization included: orbitofrontal and frontopolar ($n = 1$), inferior dorsolateral frontal ($n = 2$), medial parietal (n = 1), parieto-occipital (n = 1) and medial temporal ($n = 3$) [3]. Their results indicated that ictal swearing is more commonly seen in male subjects, and lateralises to the non-dominant hemisphere, but has poor localization value [3]. The main limitation of the previous study mentioned by the author was that patients were not able to be assessed in a standardized way. Our case is well in line with some of the observations made by Birca: a) a male subject; b) EEG onset was localized in the non-dominant hemisphere; and c) one patient in the Birca et al.'s series had an orbitofrontal lesion as well. We present a patient with seizures arising from posterior part of right gyrus rectus, and the EEG activity always spread to the right dorsolateral frontal cortex and the left temporal region when swearing appears. The propagation pattern was not quite consistent in what these authors described: ictal swearing could not be attributed to a lateral or medial origin in the frontal or temporal lobes and a tendency for ictal discharge to propagate to medial structures [4]. Besides, the mean delay of our case from EEG onset to appearance of swearing was 25.4 s. The time of swearing occurrence suggests that the extrafrontal networks may participate in ictal swearing. A circuit that includes the prefrontal cortex (PFC), amygdala, hippocampus, hypothalamus, anterior cingulate cortex (ACC) and insular cortex has been implicated in emotion regulation [5].

Driver et al. proposed that swearing is a form of emotional or compulsive speech, and there were two possible mechanisms underlying ictal speech automatisms: 1. ictal speech results from release of the dominant hemisphere from the inhibitory action of the non-dominant one; 2. ictal speech results directly from epileptic activity of the non-dominant hemisphere [6]. Ictal swearing is a kind of verbal automatism correlated with negative emotional experience, it has been known that the ventral system, including the amygdala and ventrolateral prefrontal cortex, are associated with negative emotional processing [7]. It has been proposed that an inhibitory connection from the orbitofrontal cortex to the amygdala regulates negative emotion and related behaviors. This proposal is also supported by a PET study, which shows reciprocal relations of glucose metabolism between several areas of the frontal cortex (including the OFC) and the amygdala [8]. A functional magnetic resonance imaging (fMRI) study has shown that ventromedial prefrontal cortex (vmPFC) is a key region associated with multiple components of emotional and stress processing, the vmPFC is critical for successful emotion regulation through its effective connectivity with the amygdala [9]. In our case, the triggered seizures during cortical stimulation manifested only right upper limb repetitive purposeless movements without swearing. It is an additional proof that ictal speech manifestation is probably produced by the recruitment of broad and complex neural networks.

In the previously reported cases of ictal swearing, the symptomatogenic zones have not been consistent. We present a patient with ictal swearing caused by seizures originating from the posterior part of the right gyrus rectus, and proposed that orbitofrontal–temporal networks is closely associated with symptomatology of ictal swearing. In this patient, multi-methods of neuroimaging evaluation revealed a subtle focus that was likely to be missed by routine MRI scan. The case highlights the importance of comprehensive neuroimaging evaluations in identifying the epileptogenic focus. More studies are still needed to clarify the symptomatogenic mechanism of the phenomenon.

Conflicts of interest statement

The authors declare no conflict of interest.

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References

[1] M. Gabr, H. Luders, D. Dinner, H. Morris, E. Wyllie, Speech manifestations in lateralization of temporal lobe seizures, Ann. Neurol. 25 (1989) 82–87.
[2] E.A. Serafetinides, M.A. Falconer, Speech disturbances in temporal lobe seizures: a study in 100 epileptic patients submitted to anterior temporal lobectomy, Brain 86 (1963) 333–346.
[3] V. Birca, T. Tayah, J.M. Saint-Hilaire, D.K. Nguyen, Ictal swearing: a case series and review, Epileptic Disord. 15 (4) (2013 Dec) 377–381.
[4] F. Bonini, A. McGonigal, A. Trebuchon, M. Gavaret, F. Bartolomei, B. Giusiano, P. Chauvel, Frontal lobe seizures: from clinical semiology to localization, Epilepsia 55 (2) (2014) 264–277.
[5] G. Bush, P. Luu, M.I. Posner, Cognitive and emotional influences in anterior cingulate cortex, Trends Cogn. Sci. 4 (6) (2000 Jun) 215–222.
[6] M.V. Driver, M.A. Falconer, E.A. Serafetinides, Ictal Speech automatism reproduced by activation procedures. A case report with comments on pathogenesis, Neurology 14 (1964) 455–463.
[7] F. Dolcos, G. McCarthy, Brain systems mediating cognitive interference by emotional distraction, J. Neurosci. 26 (2006) 2072–2079.
[8] R.J. Davidson, K.M. Putnam, C.L. Larson, Dysfunction in the neural circuitry of emotion regulation — a possible prelude to violence, Science 289 (5479) (2000) 591–594.
[9] J.C. Motzkin, C.L. Phillipi, R.C. Wolf, M.K. Baskaya, M. Koenigs, Ventromedial prefrontal cortex is critical for the regulation of amygdala activity in humans, Biol. Psychiatry 77 (2015) 276–284.

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