



# Ictal swearing network confirmed by stereoencephalography: a case report

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## Abstract

Ictal swearing, characterized by episodic utterance of deistic, visceral and other taboo words and phrases, is a rare manifestation of epilepsy. Limited manifestation of that elucidate neural network about manifestation of. Here we report a 43-year-old right-handed man who has suffered from refractory epilepsy manifestation of ictal swearing. The stereoencephalography(SEEG) investigation located the seizure onset zone in left orbito-frontal cortex and the involvement of left orbito-frontal and right hippocampus in the propagation of epileptic activities. Surgical resection has made the patient seizure-free. The present case gives us better understanding of these ictal phenomena and helps to optimize the placement of SEEG electrodes in refractory epilepsy patient with ictal swearing.

**Keywords** Ictal swearing · Epilepsy · SEEG · Orbitofrontal cortex

## Introduction

Epileptic seizure semiology, produced by epileptic discharges spreading in both time and space within certain neural pathways, is fundamental to epileptology. As the direct reflection of pathological brain function, remarkable variability depending on the degree of involvement of the primary or associative cortex has been recognized in individual patient. Great effort has been made to clarify semiology notably over last century. Undoubtedly, the better understanding of seizure semiology has significant implication for clinical evaluation of lateralization and localization. Moreover, it provides new insights into how the cerebral cortex is organized to execute distinct function.

Ictal speech is not uncommon during seizure which is observed in 12 to 39% of patients with temporal lobe epilepsy,

more frequently originating from the nondominant than the dominant temporal lobe. Nevertheless, ictal swearing, described as episodic utterance of deistic, visceral, and other taboo words and phrases are rarely reported in epilepsy patients. In contrast to the notion of ictal speech resulting from activation or inhibition of languish cortical function, dysregulation of emotional and behavioral function is hypothesized to underlie ictal swearing. Clinically, the application of stereoencephalography (SEEG) in presurgical evaluation for substantial patient with refractory focal seizure because of insufficient information from noninvasive investigation provides the chance to probe the epileptic network. Here, we study the involved semiological network of ictal swearing by means of SEEG in an epilepsy patient with refractory ictal swearing.

## Patient case

The patient is a 43-year-old right-handed man. He developed prominent ictal swearing at age 28 without any past history. The episodes manifest as sudden onset of loss of consciousness, repetitive “I fuck” lasting about 10–40 s without any tonic and clonic component and no postictal confusion. The episodes occurred 1–3 times every month. Ploy-antiepileptic drugs including phenobarbital, carbamazepine, oxcarbazepine, and levetiracetam failed to control the seizure.

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## Noninvasive evaluation

The patient was monitored by video-EEG (Micromed S.p.A) for 5 days. Three habitual seizures were captured. Interictal discharges of sharp and spike wave were detected over left frontotemporal area predominantly (Fp1, F7). Focal attenuation of voltage over left frontotemporal area was noted at the seizure onset, then mixed with massive muscle and movement artifacts. MRI showed subtle abnormality on left orbitofrontal region with blurring of gray-white matter junction and slightly thickness of gray matter. Consistent with the findings of MRI, fluorodeoxyglucose positron emission tomography (FDG-PET) imaging showed focal hypometabolism on the same area (Fig. 1).

## Invasive evaluation

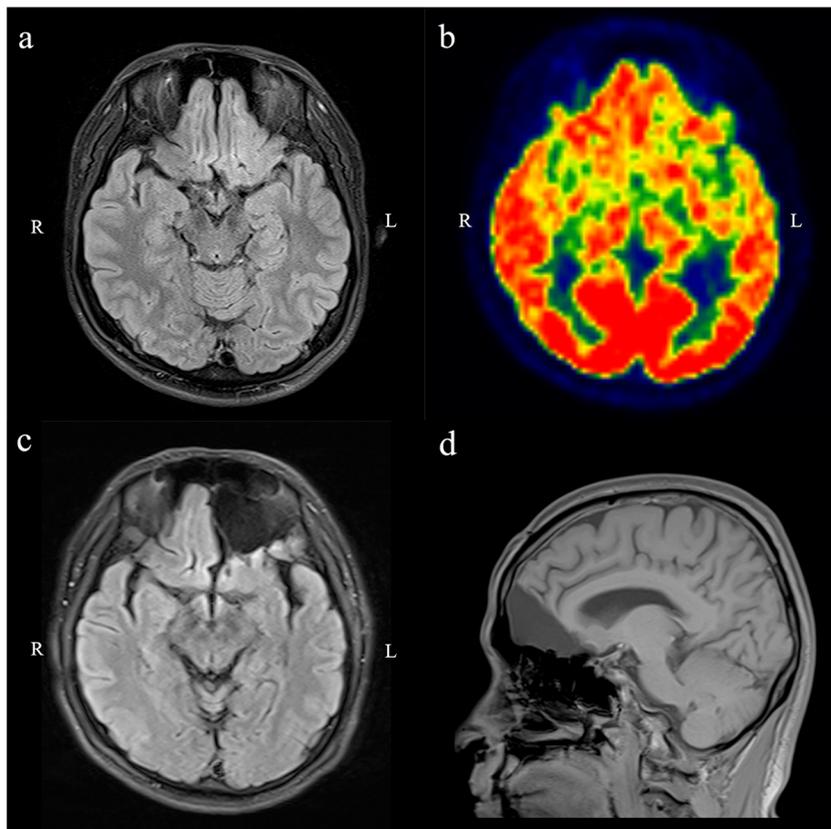
After comprehensive evaluation by multi-disciplinary team in our hospital, invasive recordings were scheduled to map the seizure onset precisely. For the design of SEEG trajectories, the primary target is left orbitofrontal area. To conceptualize the dynamics of seizure onset and propagation, left hippocampus, medial, and lateral orbitofrontal cortex, anterior cingulate cortex, Broca's area, motor cortex, anterior cingulate cortex, and right hippocampus were also explored. The implantation of depth electrodes was conducted under general anesthesia according to the

classical approach. In brief, stereotactic digital cerebral angiography was first conducted and a three-dimensional cerebral contrasted T1-weighted MRI scan was obtained in stereotactic conditions. Each trajectory was plotted onto the stereotactic digital cerebral angiogram to avoid major vessel injury in the design of SEEG trajectories. Then, electrode implantation was performed one by one using an oblique approach in the same stereotactic conditions with a robotized arm (Medtech, Montpellier, France) connected to the stereotactic frame and driven by stereotactic planning software under general anesthesia.

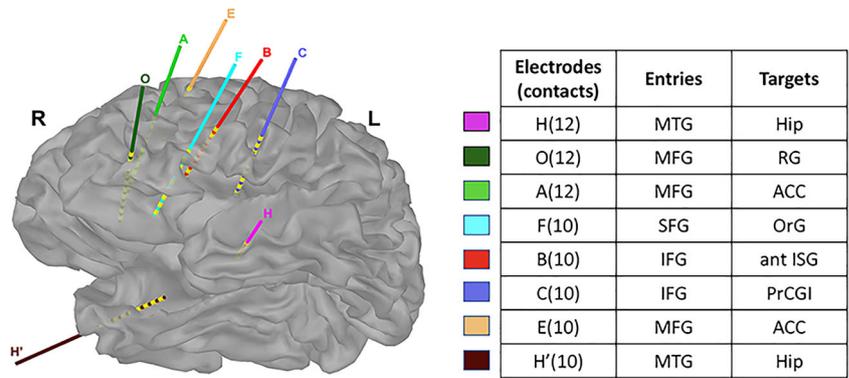
To determine the location of the SEEG electrodes, postsurgery CT was co-registered to the presurgery three-dimensional T1 image using brainstorm software. The depth electrodes were readily detected on three dimensions by visual inspection. Once the deepest point and surface of each electrode trajectory were determined, all locations of others contacts were calculated sequentially. The reconstructed electrodes were showed in Fig. 2.

Finally, 6 stereotypic seizures of ictal swearing were captured during the long-term SEEG monitoring. Initially, low voltage fast activities occurred over the lateral orbitofrontal cortex lasting about 3 s, followed by propagation to the left medial orbitofrontal cortex and frontal operculum, then followed by involvement of right hippocampus before the end of the seizure. Notably, ictal cursing words developed when ictal discharges of lateral orbitofrontal cortex were synchronized with left medial orbitofrontal cortex and frontal operculum (Fig. 3).

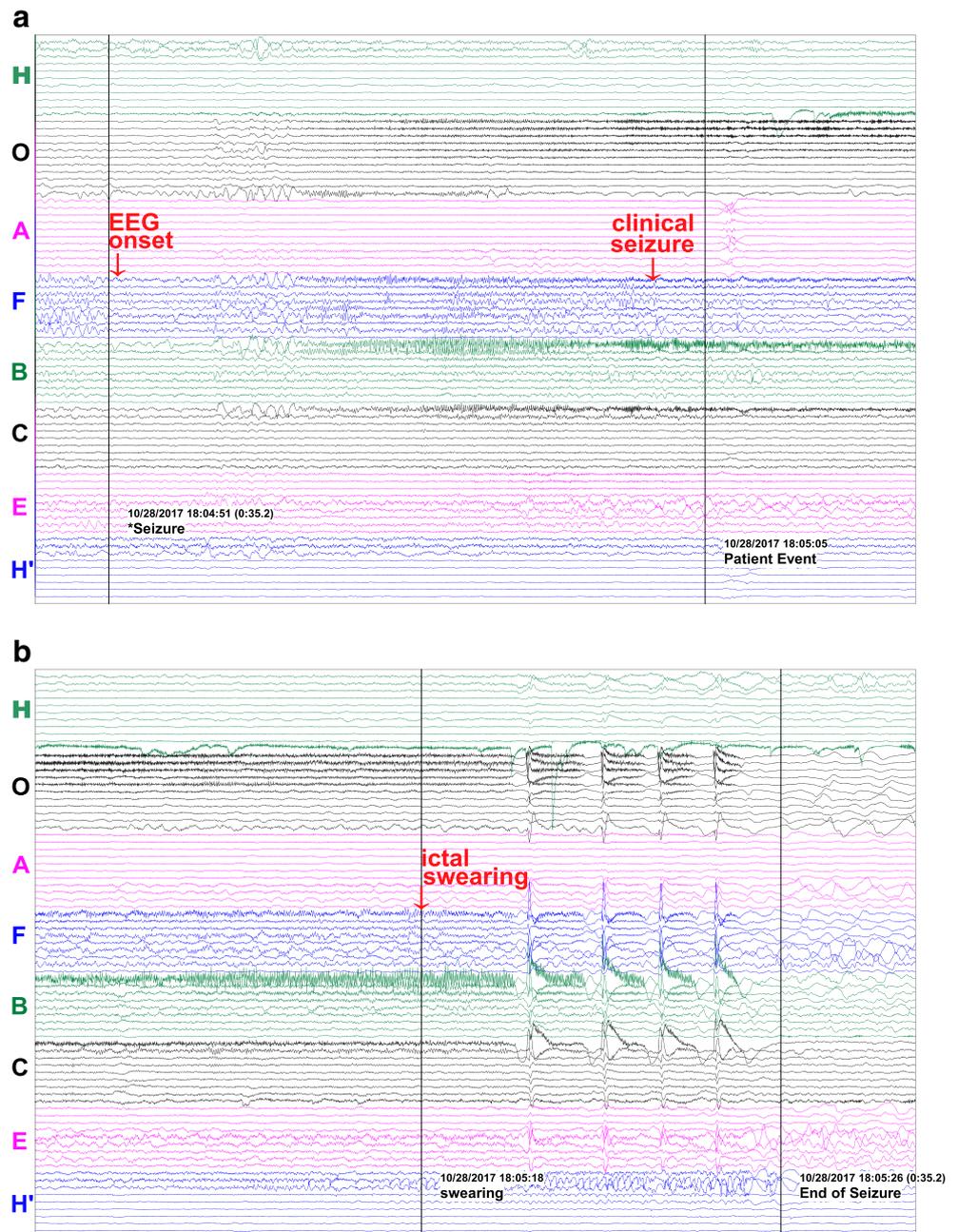
**Fig. 1** The findings of structural and functional images. **a** Axial FLAIR MR image, **b** axial FDG-PET image, **c** axial postoperative MR image, **d** sagittal postoperative MR image. R, right; L, left



**Fig. 2** The reconstruction of SEEG electrodes into brain



**Fig. 3** SEEG of an ictus. **a** ictal low voltage fast activities originated from left orbital gyrus recorded by electrode F (F1–3, arrow). **b** When the epileptic discharge involves to left orbito-frontal cortex, frontal operculum and the right hippocampus, the ictal swearing occurred (arrow), the delay from EEG onset to appearance of swearing is about 27 s



Next, we assessed the epileptogenic index of explored cortical areas (Fig. 4). The calculation of the epileptogenic index has been validated previously. It showed that the epileptogenic index on left orbitofrontal cortex was the largest, suggesting epileptic zone in consistent with observation by visual inspection.

## Epilepsy surgery and follow-up

Surgical resection of the region of PET hypometabolism including the left frontal pole and orbitofrontal cortex was completed without complication (Fig. 1). The neuropathology findings confirmed the diagnosis of FCD type Ib. He has remained seizure-free since surgery.

## Discussion

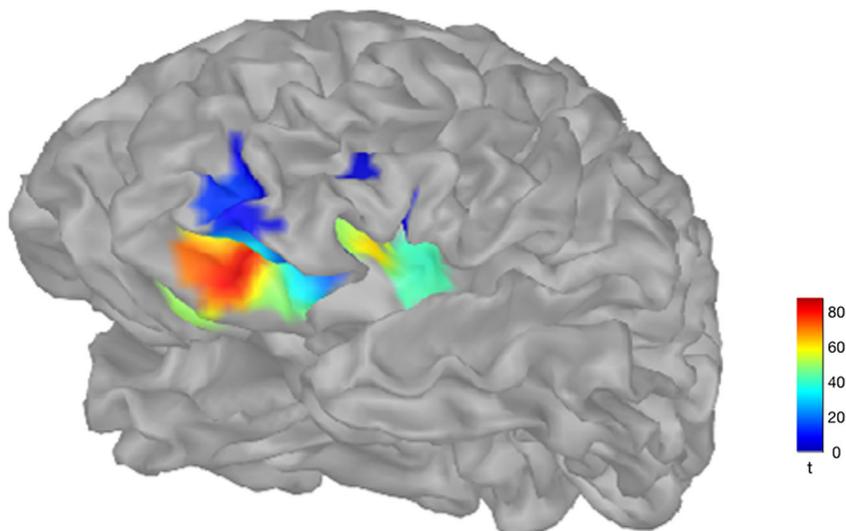
Ictal swearing is a rare epileptic ictal sign thus cohort studies are lacking. Birca et al. [2] reported the largest case series including eight ictal swearing cases but lack of neuro-electrophysiological evidences. We present a male patient with ictal swearing whose seizure onset zone studies are located in left orbitofrontal cortex by SEEG investigation, which provides us a clear characterization of the neural activity.

Swearing is a common human speech and may be present in different neurological disorders. Coprolalia, a type of involuntary swearing, is most common in Tourette syndrome (TS). Our data show that ictal swearing may share the neural network of coprolalia. Brain imaging, neurophysiological, and postmortem research have identified changes in cortical-basal ganglia structure, connectivity, and function in TS [3, 6, 9]. Worbe and colleagues presented evidence on widespread structural abnormalities in cortical-basal ganglia circuits in patients with TS using

MRI tractography and functional connectivity methods [9] and proposed that abnormalities represent immature and atypical development of these circuits. In another fMRI study on 120 participants, TS group had more activation in frontal cortex, dorsal anterior cingulate, and temporal cortices [8], which was consistent with cortical-basal ganglia circuit abnormalities in TS. Recent developments in sociality and emotion science suggest additional brain systems may be altered in TS. Albin [1] described that TS may be a disorder of social communication resulting from developmental abnormalities at several levels of the social decision-making network. In accordance with TS, previous studies showed ictal swearing more often involved prefrontal circuit [2, 5]. The orbitofrontal cortex receives inputs from all the sensory modalities, including amygdala, hippocampus, cingulate cortex, and other brain areas and projects back to temporal lobe areas [4, 7], which is a major component of social decision-making network. Consistent with previous observation, this case also suggests the critical role of orbitofrontal lobe in the ictal swearing. Our data demonstrated that the synchronization of ictal discharges on orbitofrontal cortex, frontal operculum, and mesial temporal lobe is crucial for ictal swearing, suggesting ictal swearing may share the anatomy and physiology of coprolalia.

Ictal swearing is rare in epilepsy patients, which epileptic zones have not been consistent in previously studies. Under certain circumstance, the invasive investigation is necessary to identify the epileptic seizure zones. We report a case with prominent ictal swearing originating from left orbitofrontal cortex. The SEEG recordings illustrate the involvement of orbitofrontal, frontal operculum, and mesial temporal structures in the propagation of epileptic activities. These findings may help optimize the placement of SEEG electrodes in patient with refractory epilepsy with ictal swearing. It is also a supplement of the pathological research in the involuntary swearing.

**Fig. 4** Epileptogenicity mapping of SEEG group analysis with the 6 seizures together showed that the epileptogenic index on left orbitofrontal cortex was the largest. R, right; L, left



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## Compliance with ethical standards

**Patient consent** The patient has consented to the submission of the case report for submission to the journal.

**Conflict of interest** The authors declare that they have no conflict of interest.

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**Comments** Huaqiang Zhang et al. are providing us with the interesting case report of an epileptic patient presenting with ictal swearing bringing argument in favor of the frontoorbital origin of this semiology or at least the involvement of this cortical region. It is well known that correlating univocally one clinical symptom to a specific area of the brain is always a dangerous exercise due to the network organization of the brain. Swearing is also a key symptom in a different neurological disease which is the Tourette syndrome. Sudden, stereotypic explosive coprolalic short sentences are likely to involve subcortical motor scheme.

Like pointed by the authors in Tourette syndrome several studies have underlined both the role of prefrontal cortex and cortical-basal ganglia circuit abnormalities 1. The VIP1 area in the hypothalamic area in connection with the tegmental periaqueductal area has been demonstrated in diverse animals to generate vocalization 2. The activation of the equivalent of this hypothalamo-mesencephalic circuitry in human has been suspected to play a role in the ictal laugh observed frequently in hypothalamic hamartomas

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