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# A hypermotor seizure with a focal orbital frontal lesion originating in the insula: A case report

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Received 13 March 2008; received in revised form 23 June 2008; accepted 26 June 2008

Available online 30 August 2008

## KEYWORDS

Frontal lobe epilepsy;  
Hypermotor seizure;  
Insular epilepsy;  
Epilepsy surgery

**Summary** We present herein the case of a patient with a focal orbital frontal lesion on magnetic resonance imaging (MRI), but an insular onset of seizures. A 15-year-old boy suffered from hypermotor seizures for 9 years. In his seizures, he initially had a sensation that sounds were distant, and then his consciousness became impaired. After a short period of tonic activity, violent activities occurred, such as kicking or gripping some objects and shaking. MRI showed a focal cortical abnormality in the right orbital frontal lobe. [(18)F]FDG-PET revealed diffuse hypometabolism in the right frontal lobe, especially in the same site as the cortical lesion on MRI. The seizure onset zone was localized in the right anterior insula by intracranial recording. A resection of the right anterior insula and a partial disconnection of the frontal lobe were performed, rendering the patient seizure-free.

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## Introduction

Hypermotor seizures are thought to be closely associated with frontal lobe epilepsy (Lüders and Noachtar, 2000). However, it has been reported that hypermotor seizures can originate from the extrafrontal area (Sussman et al., 1989; Swartz, 1994; Nobili et al., 2002, 2004). A few cases of hypermotor seizures with onset in the insula have been suc-

cessfully treated (Isnard et al., 2000, 2004; Kaido et al., 2006; Ryvlin et al., 2006; Dobesberger et al., 2008). In those reported cases, the insular lobes were either with or without lesions on magnetic resonance imaging (MRI). To our knowledge, there are no reported cases of insular epilepsy with hypermotor seizures without an insular lesion on MRI, but with a frontal lesion which is more likely associated with hypermotor seizures.

Herein we report a patient with a focal orbital frontal lesion on MRI who underwent successful surgical treatment. The semiology of the seizure and imaging of the orbital frontal lesion suggested frontal lobe epilepsy; however, intracranial recording and stimulation revealed an insular seizure onset.

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## Case report

A 15-year-old, right-handed boy presented with a 9-year history of seizures. He had no personal or family history of neurologic disease, including epilepsy. At the onset of the seizures, the patient was 6 years of age. In his habitual seizures, he initially had a sensation that sounds were distant, after which he felt as though he wanted to hug something, then his consciousness became impaired. After a short period of symmetrically tonic activity in lower extremities (approximately 10–15 s), violent activities occurred, such as kicking or gripping some objects and shaking. The seizure lasted approximately 30–40 s and then the patient recovered fully without postictal symptoms. According to international league against epilepsy (ILAE) classification of seizure type, the patient suffered from a kind of partial seizure. The frequency of seizures was 3–6 times/day and all seizures occurred in the daytime. Valproate and phenobarbital were administered, but did not control the seizures. One year later, the seizures began to occur during sleep with a frequency similar to that in the daytime. Valproate was changed to phenytoin. Seizures were then controlled satisfactorily, with a frequency of 1 every 2–3 months for 3 years. In the last 3 years, the seizures have recurred with increasing frequency, up to about 20–50 times/day. Secondary generalized tonic-clonic seizures (GTC) have occurred several times. Various medications have been prescribed, but they have exerted poor control.

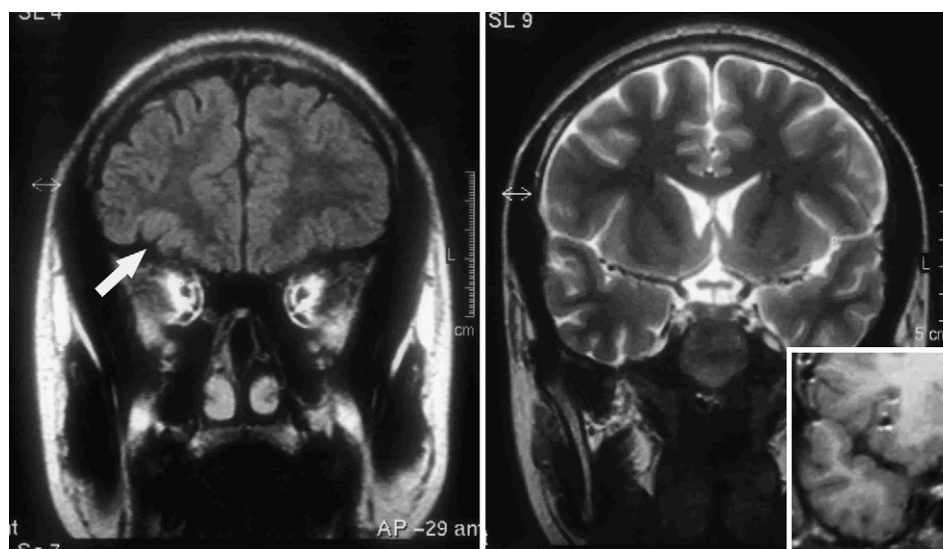
The neuropsychologic and neurologic examinations were normal. A MRI showed a focal intensity lesion with FLAIR images involving the right orbital frontal area (Fig. 1), which was confirmed as abnormal by PET scanning. PET scanning also showed diffuse hypometabolism in the frontal lobe and insular cortex on the right side, which was more obvious in the right orbital frontal area and consisting with the focal lesion shown by the MRI. Ictal scalp electroencephalogram

(EEG) showed a focal seizure onset from the right anterior frontal lobe. Intracranial electrodes were implanted on the surface of the frontal lobe, including the mesial and orbital frontal areas, and part of the lateral temporal and parietal lobes on the right side. Because the lesion in orbital frontal lobe was close to insular cortex and hypometabolism of insular cortex was showed on PET scanning, the right insula was recorded by depth electrodes implanted by stereotactic means (Figs. 1 and 3). Thirty-eight seizures were recorded and ictal intracranial EEG patterns were almost same. Ictal intracranial EEG showed a focal seizure onset from the right anterior part of the insula with fast propagation (within 3 s) to the right frontal lobe (Fig. 3). Electrical cortex stimulation (0.2 ms width pulse, 50 Hz, 1–15 mA and 5–10 s duration) was performed. Four habitual seizures were elicited in the insula.

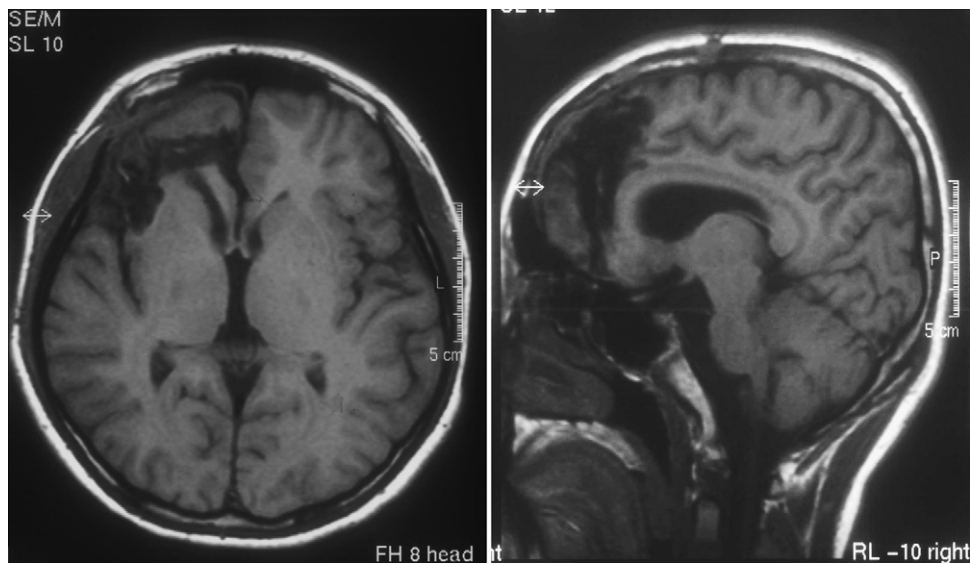
A resection of the right anterior insula and medial frontal area, and a partial disconnection of the frontal lobe were performed (Figs. 2 and 3). The patient became seizure-free after surgery for 17 months while being maintained on lamotrigine monotherapy, but his habitual auras still existed. Because of disconnection of the orbital frontal lobe with the lesion, the pathology of the lesion cortex was unavailable. Other brain tissue sections showed no significant diagnoses.

## Discussion

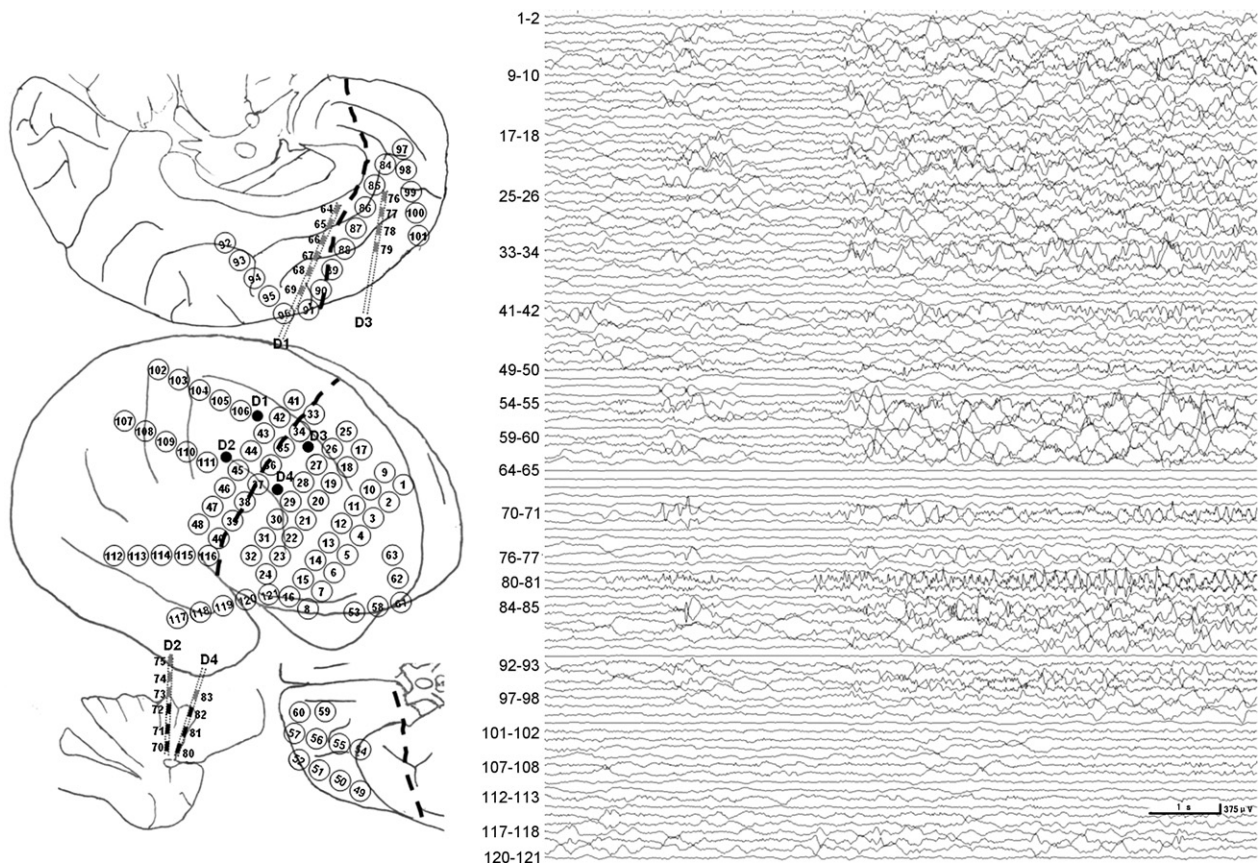
According to the review (Lüders and Noachtar, 2000), hypermotor seizures are most frequently associated with frontal lobe epilepsy, auras precede hypermotor seizures in the majority of cases, and hypermotor seizures are of short duration and often combined with tonic activity. This case consisted of the above descriptions, except for the origin of the seizures. Hypermotor seizures also can originate in the temporal lobes (Sussman et al., 1989; Swartz, 1994; Nobili



**Figure 1** Preoperative MRI FLAIR imaging, T2-weighted imaging and T1-weighted imaging after implantation of intracranial electrodes. *Left:* Preoperative coronal FLAIR imaging showed an abnormal cortex signal in the right orbital frontal area (white arrow) compared with the left orbital frontal area, which was confirmed as abnormal by PET scanning. *Right:* Coronal T2-weighted imaging showed structural normality of the insular cortex. Small size coronal T1-weighted imaging showed depth electrodes inserted in the right anterior insula.



**Figure 2** Postoperative MRI imaging of the patient. *Left:* Axial T1-weighted imaging showed the resection of the right anterior insula and the junction cortex between the insula and the frontal lobe. *Right:* Sagittal T1-weighted imaging showed the resection of the medial frontal area and disconnection of the frontal pole and the orbital frontal area (disconnection lines were shown in Fig. 3).



**Figure 3** Extraoperative intracranial EEG of an ictus. *Left:* Schematic representation of electrode locations. The frontal disconnection lines of surgery are showed by black dash lines. D1, D2, D3 and D4 represent four depth electrodes. *Right:* Low voltage fast activity is recorded over the electrodes 80–81 (anterior insula) before any propagation to medial frontal, orbital frontal and dorsolateral convexity regions.

et al., 2002, 2004) and insular cortex (Isnard et al., 2000, 2004; Kaido et al., 2006; Ryvlin et al., 2006; Dobesberger et al., 2008).

Guillaume and Mazars (1949a,b) were the first authors to call attention to the concept of insular epilepsy. Due to the lack of intracranial EEG recording direct from the insular cortex, firm confirmation of seizures with insular onset was not available until the work of Isnard et al. (2000). Since then, more cases of insular epilepsy with stereo-electroencephalograph confirmation have been reported (Isnard et al., 2004; Ryvlin et al., 2006; Dobesberger et al., 2008). For clinical characterization of insular seizures, laryngeal discomfort with thoracic oppression or dyspnea, unpleasant paresthesias, or a sensation of warmth focused on the perioral region or extended to a large somatic territory and dysarthric or dysphonic speech at the very beginning of the seizure suggests an insular seizure (Isnard et al., 2004). Our patient had none of the aforementioned symptoms at the onset of the clinical seizures, but a sensation that sounds were more distant. Auditory auras are a relatively rare manifestation of focal epilepsy and generally originate from the temporal lobe (Mulder and Daly, 1952; Currie et al., 1971; Gupta et al., 1983; Palmi and Gloor, 1992; Acharya et al., 1996). Schneider and colleagues (Schneider et al., 1961, 1965) described auditory symptoms in three patients with frontal, parietal, and occipital lesions, presumably due to a spread of epileptic discharge to the temporal lobe. Acharya et al. (1996) also reported two patients with auditory auras who had frontal lobe epilepsy.

For this case, besides the depth of the electrode in the insular cortex, extensive subdural electrodes covered the convexity of the frontal lobe, mesial and orbital frontal lobes, and part of the temporal and parietal lobes, but only the depth electrode elicited aura and habitual seizures. Only one subdural electrode in the right frontal operculum, just on the upper bank of the right lateral fissure, elicited auras but no seizures. The results of stimulation were consistent with the results of recording. Even if semiology of seizures, neuroimaging, and scalp EEG data all support a diagnosis of frontal lobe epilepsy, however, it is still possible the real seizure onset zone is in the insula in such cases.

Although intracranial EEG recording and electrical cortex stimulation suggest an insular onset, we still performed a resection of the medial frontal area and a partial disconnection of the frontal lobe, including the orbital frontal and polar frontal lobes. There are two reasons for our final decision to do such resection. First, fast propagation of ictal discharges after insula onset quickly caused abnormal discharges in other parts of the frontal lobe. Second, there is still a possibility that orbital frontal lesion and its vicinity may be epileptogenic because of relatively limited sampling from the orbital frontal lobe, but the remains of patient's aura after surgery suggest the seizure onset zone should be in the insular cortex.

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