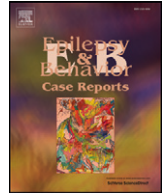




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Case Report

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ABSTRACT

Epileptic olfactory auras manifesting as simple partial seizures are rare. We report a patient who presented with olfactory auras after hemorrhage from a cavernous angioma in the left mesial temporal region. His olfactory auras persisted 12 years after two surgeries for a cavernous angioma. Intracranial depth electrodes revealed a very focal isolated epileptogenic zone in the amygdala. Olfactory auras were successfully treated by focus resection.

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1. Introduction

A subset of patients with head injury, migraine, schizophrenia, depression, tumor, and epilepsy experience olfactory auras. Jackson and Stewart named them “uncinate fits” [1]. Chen et al. [2] reported that 5.5% of patients with temporal lobe epilepsy experienced olfactory auras. They are usually accompanied by other types of auras such as rising epigastric discomfort and a sensation of fear, followed by complex partial seizures. Although mesial temporal structures such as the uncus, amygdala, and orbitofrontal cortex appear to be implicated, the definite site of the epileptic focus eliciting olfactory auras and the effectiveness of surgical intervention remain unknown [3]. We report the clinical course of a patient with intractable olfactory auras alone secondary to a left mesial temporal angioma who underwent intracranial EEG recordings to resect residual very focal isolated epileptic networks in the amygdala after the two surgeries for angioma.

2. Case report

This 42-year-old right-handed male had been healthy before he experienced the sudden onset of headache, nausea, and the subsequent perception of a foul smell at the age of 27 years. Computed tomography (CT) showed a small hemorrhage in the left mesial temporal lobe (Fig. 1A). His neurological examinations were normal. He had a mild headache and experienced a putrid odor several times a day without loss of consciousness. Magnetic resonance imaging (MRI; Fig. 1B) showed a fresh 20-mm diameter hemorrhage in the left amygdala.

He underwent resection of the anteromesial part of the left temporal lobe via the transylvian approach. Intraoperative electrocorticography (ECoG) showed spikes from the uncus. The spikes disappeared after we totally evacuated the hematoma and partially removed small vessels and membranous tissues in the mesial part of the hematoma. The histopathological diagnosis was cavernous angioma.

Although his olfactory auras abated immediately after the surgery, they recurred two years later. Magnetic resonance imaging showed a small enhanced lesion along the surgical cavity, and at the age of 30 years, he underwent a second resection of residual angioma for recurrent olfactory auras without intraoperative ECoG. Again, the histopathologic diagnosis was cavernous angioma.

His olfactory auras persisted during the next 12 years despite the administration of nine different antiepileptic drugs. His perception of a putrid odor worsened and occurred more than 10 times a day and was followed by listlessness without unconsciousness or convulsive

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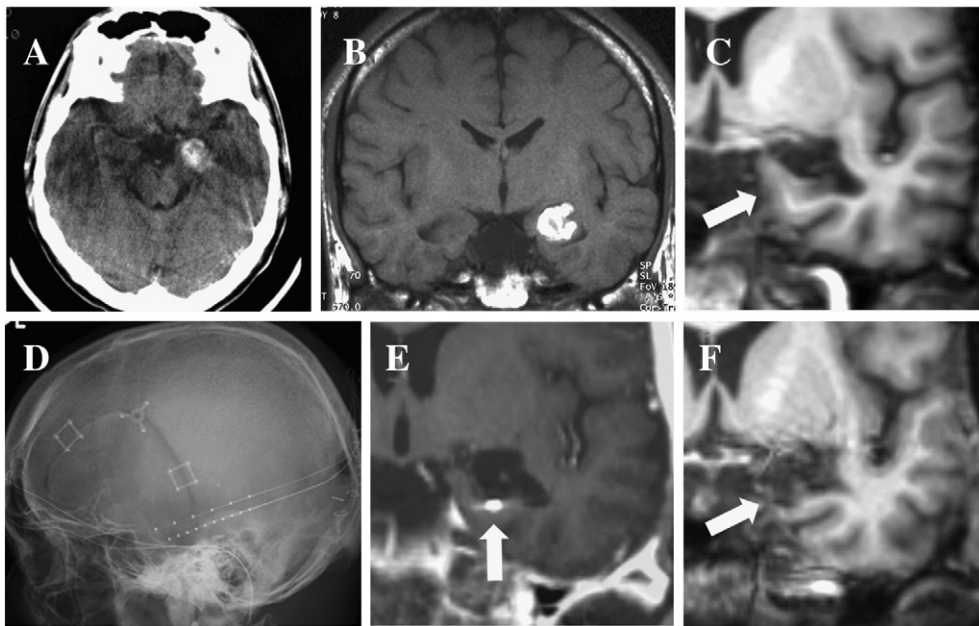


Fig. 1. Consecutive CT scan, MR images, and skull X-ray with depth electrodes. (A, B) CT scan (A) and coronal MRI (B) acquired at the start of the seizures. A hematoma is seen in the left medial temporal lobe. (C) Coronal MRI after hematoma evacuation in the first surgery. The arrow indicates the parahippocampal gyrus located mesially to the surgical cavity. (D) Lateral skull X-ray shows the bilateral intracranial electrodes inserted via the occipital lobes. The left and right depth electrodes feature 10 and 6 contacts, respectively. Both depth electrodes are stereotactically located from the hippocampus to the amygdala. (E) The fusion image of MRI and CT identifies the location of L2 of the left depth electrode. The arrow indicates L2 placed in the inferomedial part of the previous surgical cavity. (F) Coronal MRI after the third surgery. The arrow indicates the cavity of the resected left prepiriform cortex and the residual mesial-cortical part of the amygdala.

seizures. It severely interfered with his daily activities. His listlessness was not associated with any changes in the EEG recorded from hippocampal depth electrodes and was considered to be a postictal symptom. He knew that the smell did not arise from the external environment; while the odor was always unpleasant, it varied from faint to strong.

Magnetic resonance imaging showed the prior surgical resection in the lateral-basal part and residual tissue in the mesial-cortical portion of the amygdala (Fig. 1C). A scalp EEG revealed interictal spikes at T1, T3, and F3. There were subtle EEG changes consisting of left frontotemporal theta activities after the olfactory auras without any preictal and ictal changes. Magnetoencephalography (MEG) demonstrated spike dipoles in the mesial temporal lobe with horizontal orientation indicative of mesial temporal epileptic networks. Iodine-123-iomazenil single-photon emission computed tomography (SPECT) revealed hypoperfusion in the left temporal lobe. The intracarotid sodium amyltal test confirmed left hemispheric dominance for language.

We stereotactically placed two depth electrodes in the bilateral amygdalae and hippocampi via the occipital lobes of the patient at the age of 42 years (Fig. 1D). While he was asleep, ictal low-amplitude fast (around 40 Hz) appeared only at electrodes L1, L2, and L3 in the anterior contacts placed in the mediocortical part of the amygdala (Fig. 2). They lasted up to 60 s without evolution. After the prolonged ictal discharges, the sensation of a putrid odor awakened him. The ictal patterns were stereotypic when he experienced “stronger” olfactory auras regardless of whether he was awake or asleep. Interictal high-amplitude spike and waves were recorded at only the same electrodes from L1 to L3.

The ictal and interictal zones were identified in the left prepiriform cortex and the residual mesial-cortical part of the amygdala on the second postoperative MRI (Fig. 1C). We planned to perform selective resection of the presumed epileptogenic zone in the residual amygdala without involving the hippocampus. The previous surgical cavity was

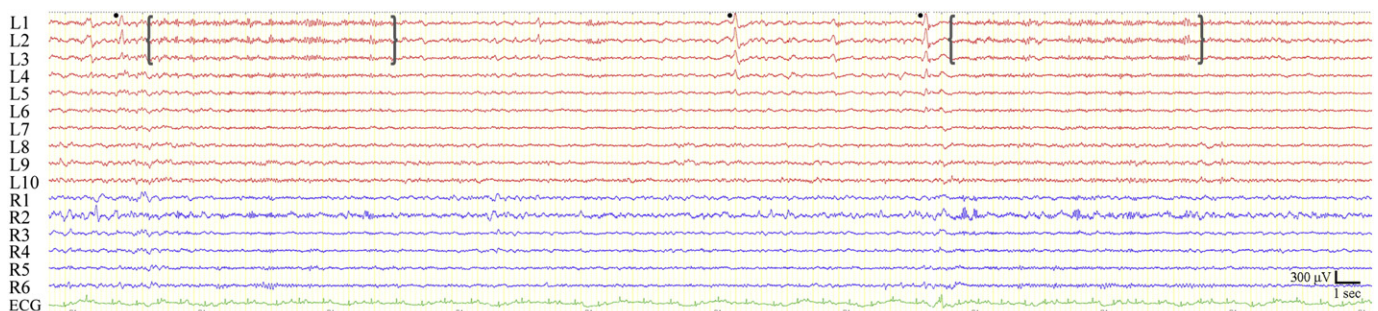


Fig. 2. Intracranial EEG findings. The 16 monopolar contacts of the bilateral depth electrodes are comprised of 10 contacts (red) in the left hemisphere and 6 contacts (blue) in the right hemisphere. Both depth electrodes are stereotactically located from the hippocampus to the amygdala via occipital lobes (Fig. 1D). Three interictal spikes are seen from L1 to L3 (closed circles). Two brackets indicate ictal low-amplitude fast activities (around 40 Hz) lasting around 10 s in duration each. These short-duration fast activities occurred frequently during sleep. When the ictal low-amplitude fast activities lasted longer than 20 s, the patient awoke and complained of a foul odor (sampling rate: 2 kHz; LFF, 1.59 Hz; HFF, 600 Hz).

opened via the transylvian approach. Using an intraoperative navigation system (Fig. 1E), we aspirated the left prepiriform cortex and the residual mesial–cortical part of the amygdala. Resection ended when the arachnoid membrane of the parahippocampal gyrus was exposed (Fig. 1F).

At 9-month follow-up, he reported being able to resume most of his normal daily activities while on antiepileptic drugs. There was no decline in his cognitive functions. He no longer experienced the strong unpleasant putrid odors and their effects. He did perceive a faint citrus smell that did not bother him.

3. Discussion

This patient never manifested consciousness alterations upon experiencing the olfactory auras. The two resective surgeries failed to block the epileptic network and did not change his seizure patterns. We posit that the absence of ictal discharges on the scalp EEG and very confined intracranial ictal discharges reflected the noninvolvement of the hippocampus and other temporal regions and accounted for the absence of complex partial seizures. Only three depth electrodes, which were located in prepiriform and entorhinal cortices and the corticomesial region of the amygdala, provoked just olfactory auras as simple partial seizures. West and Doty [4] reported that the specific cortical areas' directly axonal projections from the olfactory bulb terminate presenting olfactory auras.

A subset of patients with temporal lobe epilepsy may not show ictal discharges on scalp EEGs during simple partial seizures. The depth electrodes in the mesial temporal region identified the very focal and isolated epileptic network related to the olfactory auras in our patient. Because of the lack of hippocampal involvement and of complex partial seizures, scalp EEGs did not demonstrate ictal evolution. In the absence

of EEG changes, it is difficult to differentiate psychiatric problems from the simple partial seizures of uncinate fits and nauseating and cephalic auras without consciousness changes. Subtle interictal epileptiform discharges on scalp video-EEGs cannot exclude the existence of a very focal epileptogenic zone in a subset of various auras that are refractory to antiepileptic drugs in patients with temporal lobe epilepsy.

We report a patient who presented with olfactory auras secondary to a cavernous angioma in the mesial temporal region and underwent intracranial depth electrode recording to demonstrate a very isolated epileptogenic zone in the amygdala.

Acknowledgments

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Conflict of interest

None of the authors have potential conflicts of interest to be disclosed.

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