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Case Report

A case of unruptured aneurysm of the internal carotid artery presenting as olfactory hallucinations

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Abstract

Background: Olfactory hallucination, a symptom of medial temporal lobe epilepsy, is rarely associated with unruptured intracranial aneurysms.

Case Description: We encountered this situation in a 70-year-old woman with an unruptured aneurysm at the bifurcation of the internal carotid and posterior communicating artery. We were able to achieve epileptic control by craniotomy clipping and medial temporal lesionectomy.

Conclusion: According to our knowledge, previous reports are limited to cases of large middle cerebral artery aneurysms compressing the lateral orbitofrontal cortex, and this is apparently the first report of a case where olfactory hallucinations occurred from direct stimulation of the entorhinal cortex by an internal carotid and posterior communicating artery bifurcation aneurysm. We examined the pathophysiology underlying the development of olfactory hallucinations. We found craniotomy clipping and focal resection to be useful from the standpoint of seizure control. Whether seizure control can also be obtained with intracranial aneurysm coiling should be investigated in the future.

Key Words: Neck clipping, olfactory hallucination, resection of epileptogenic foci, surgery, unruptured aneurysm

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INTRODUCTION

Having olfactory hallucinations is a symptom of medial temporal lobe epilepsy syndrome. It is believed that they occur from stimulation of the region where the lateral olfactory stria passes through the amygdala to the entorhinal cortex in the anterior parahippocampal gyrus.

In this article, we report our experience with a patient, presenting with olfactory hallucinations, who underwent aneurysmal clipping and resection of epileptic foci for an unruptured aneurysm of the internal carotid artery.

CASE PRESENTATION

The patient was a 70-year-old woman. She had an elder sister with oculomotor nerve palsy who had undergone

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clipping for an internal carotid artery aneurysm. The patient was also concerned about an aneurysm but had never experienced a severe headache. She visited a local clinic because she suddenly developed the symptom of smelling burning rubber, which occurred a few times per week for the previous month. She underwent magnetic resonance imaging (MRI) of the head and was found to have a right internal carotid artery aneurysm. She was then referred and admitted to our hospital. She had clear consciousness, without any particular neurological deficits or any abnormalities in blood chemistry. Even after hospitalization, she had sudden episodes of phantosmia, which, however, were not associated with loss of consciousness. No obvious spikes were observed on scalp electroencephalogram (EEG). On magnetic resonance angiography (MRA) of the head, we noted a dumbbell-shaped aneurysm, 10 mm in diameter, appeared to have extended from the right internal carotid artery to pierce the medial temporal lobe cortex [Figure 1a]. Plain T1-weighted MRI revealed an area of high signal intensity in the distal dome of the aneurysm, suggesting thrombosis. The surrounding brain showed high signal intensity on the T2-weighted image [Figure 1b]. Cerebral angiography revealed that the principal site of the aneurysm was the posterior communicating artery (P-com) bifurcation, and contrast medium retention was observed in the distal dome [Figure 1c]. Post-contrast constructive interference in the steady state (CISS) imaging revealed that the aneurysm dome intruded into the entorhinal sulcus and reached the amygdala [Figure 1d]. As its long axis and shape suggested a risk of rupture and its nature being an unruptured intracranial aneurysm presenting with olfactory hallucinations, the condition was determined to be an indication for surgery.

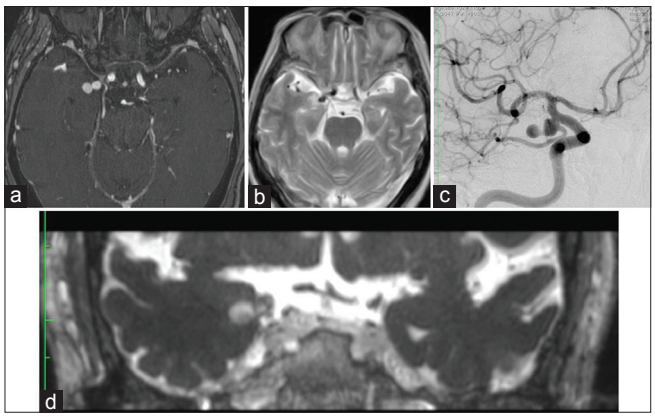

Figure 1: (a) An original image of gadolinium-enhanced MR angiography. A dumbbell-shaped aneurysm is intruding into the entorhinal sulcus from the right internal carotid artery. (b) A T2-weighted horizontal MRI image. A high signal intensity area suggesting edema around the aneurysm is shown in the medial temporal lobe. (c)The right internal carotid artery angiography shows an aneurysm with a narrow neck at the posterior communicating artery bifurcation. (d) A coronal section image of gadolinium-enhanced T2-weighted CISS MRI. The tip of the aneurysm reaches the vicinity of the amygdala nuclei

Surgery

A right pterional craniotomy was performed using a trans-sylvian approach to the right internal carotid artery. After confirmation of the presence of the aneurysm, 16 electrodes were placed over the lateral temporal lobe cortex to record temporal cortical EEG. As a result, clear spike discharges were detected from some electrodes [Figure 2a]. After reaching the aneurysm, we found that the P-com was separated from the proximal neck, and the right oculomotor nerve was compressed. The aneurysm intruded into the medial temporal lobe cortex, which was slightly discolored to a brownish tone [Figure 2b]. We performed neck clipping after confirming the position of the anterior choroidal artery distal to the aneurysm neck as well as the positions of the P-com and its perforators. Thereafter, the discolored medial temporal lobe cortex was incised and resected, and thrombosis of the distal dome was confirmed [Figure 2c]. After detaching it circumferentially, we removed the dome as a mass at a position slightly distal to the clip. Thereafter, the medial temporal lobe cortex was resected by aspiration until normally colored white matter was exposed, and the posterior cerebral artery was visually identified at the bottom [Figure 2d]. After these procedures, EEG was again recorded from the lateral temporal cortex. In the EEG, although slow waves appeared in part, no obvious spikes were detected, and the operation was completed [Figure 2e].

Pathology

The resected aneurysm wall was a pseudoaneurysm without an arterial wall structure; the internal elastic lamina was composed of thin fibrous tissue with a mild inflammatory response [Figure 3a]. The excised temporal lobe tissue showed degenerated, ganglion cell-like neurons in the cortex, and edematous changes in the white matter accompanied by the presence of reactive astrocytes. There were no changes suggestive of an old hemorrhage [Figure 3b].

Postoperative course

The patient had an uneventful clinical course, and the olfactory hallucinations resolved. Postoperative 3D-CT angiography confirmed the disappearance of the aneurysm and the patency of the P-com [Figure 3c]. In addition, contrast-enhanced MRA showed the extent of resection of the medial temporal lobe cortex. She was discharged ambulating independently under treatment with oral levetiracetam 1000 mg, which was discontinued at the sixth postoperative month. Since then, she has not experienced seizures.

DISCUSSION

A study analyzing 1423 patients with partial epilepsy reported that olfactory hallucination was a rare symptom, occurring in only 13 patients (0.9%).[1] Of the 13 patients,

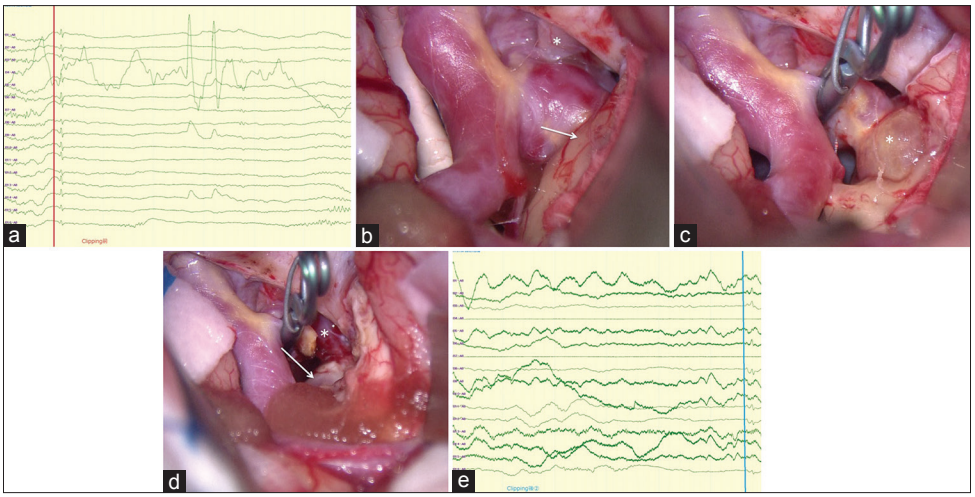

Figure 2: (a) Clear spikes were evoked in several electrodes of intraoperative cortical EEG. (b) The exposed aneurysm intruded into the right medial temporal lobe cortex, where brownish discoloration (arrow) occurred (*right oculomotor nerve). (c) After detachment of the posterior communicating artery and its perforators, neck clipping was performed, followed by corticotomy of the brownish discolored area of the medial temporal lobe cortex. With these procedures, the thrombosed distal dome (*) was exposed. (d) After resection of the aneurysmal dome, the right medial temporal lobe cortex was removed until normally colored white matter was exposed. (e) No obvious spikes were detected on the intraoperative cortical EEG after removal of the medial temporal lobe cortex

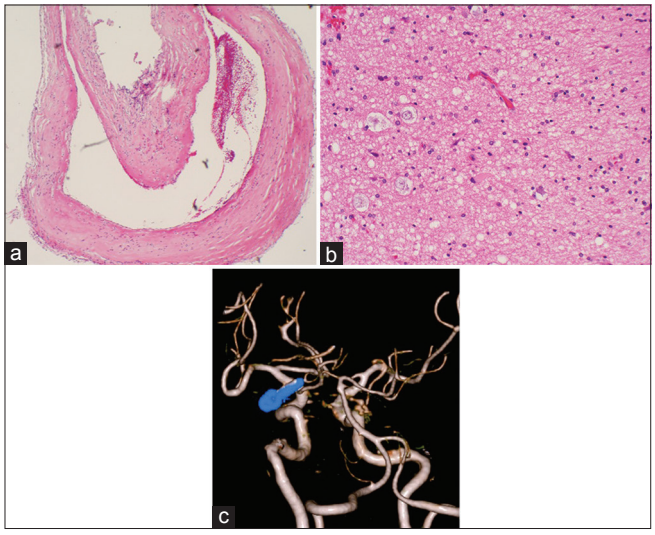

Figure 3: (a) The resected aneurysm wall was a pseudoaneurysm without an arterial wall structure. (b) The medial temporal lobe resected at the same time showed degenerated neurons and edematous tissue accompanied by gliosis. There was no clear hemosiderin deposition. (c) Postoperative 3D-CT angiography confirmed the complete disappearance of the aneurysm and presented a favorable image of the posterior communicating artery

10 patients developed tumors in the area from the entorhinal cortex to the amygdala. In the remaining 3 patients, the cause of olfactory hallucination was considered to be medial temporal sclerosis.[1] In a study where temporal lobectomy was performed in 217 patients with intractable temporal lobe epilepsy, 12 patients (5.5%) were found to have olfactory aura. The cause in these patients was considered to be medial temporal sclerosis in 7 patients, tumors in 4 patients, and arteriovenous malformation in 1 patient.[2]

Unruptured intracranial aneurysms rarely lead to partial seizures. According to a study conducted by Solomon *et al.* in 202 patients with unruptured intracranial aneurysms, 5% of the patients had seizures, and all of them had large aneurysms of ≥1 cm in diameter.[7] On the other hand, a study conducted by Currie *et al.* in 666 patients with temporal lobe epilepsy reported that the cause was unruptured intracranial aneurysm in only 1 patient.[3] There have been rare reports, i.e., only a few cases, of olfactory aura caused by unruptured intracranial aneurysms[5,6,8] [Table 1]. As described earlier, for the development of olfactory aura, it is necessary to apply stimulation to some part of the olfactory neural network. Therefore, it is reasonable to consider an aneurysm that is in contact with the olfactory tract, lateral olfactory stria, amygdala, parahippocampal gyrus, or entorhinal cortex to be the cause. On the other hand, it has been clarified that the orbitofrontal cortex in the frontal lobe is involved particularly in olfactory discrimination as the secondary olfactory cortex. In fact, as to all the previously reported cases listed in Table 1, the lateral orbitofrontal cortex was considered to be stimulated by a large middle cerebral artery aneurysm exceeding 1 cm in diameter. Therefore, this is apparently the first report of a case where an internal carotid and posterior communicating artery bifurcation aneurysm extended laterally, growing in size to ≥1 cm, and intruded into the entorhinal cortex, thereby causing olfactory aura. In the present case, the patient complained of an unusual smell of something like burning rubber. However, the nature of the unusual smell perceived was completely different from the smell reported in the cases of middle cerebral artery aneurysms, such as the sweet pleasant smell of flowers, sweet fruits, or roses. Interestingly, this may reflect a difference in the affected site.

Table 1: Summary of clinical picture of unruptured intracranial aneurysms presenting with olfactory hallucinations

Author	Age/ Sex	Location	Size (mm)	Type of olfactory hallucination	Affected brain	Mechanism	Treatment	Postoperative AED	Seizure outcome
Whittle (1985)	52/M	MCA	25	Felt an overwhelming pleasant smell, likened to roses	Medial temporal cortex	Focal compression	Clipping	Maintenance of phenytoin and methylphenobarbitone	Controlled
	48/F	MCA	40	Felt a putrid smell, likend to peeled chokos	Lateral orbitofrontal cortex	Focal compression	Wrapping	Maintenance of phenytoin, methylphenobarbitone, carbamazepine, sodium valproate	Uncontrolled
Mizubuchi (1999)	49/F	MCA	10	Felt a sweet pleasant smell, likened to flowers or sweet fruits	Lateral orbitofrontal cortex	Focal compression	Clipping	Administration of zonisamide for two years	Cure
Miele (2004)	55/F	MCA	15	Felt a perception of a strange smell	Medial temporal cortex	Subclinical hemorrhage	Clipping	Maintenance of carbamazepine	Controlled
Presented case (2017)	70/F	IC-PC	12	Felt a smell of burnt rubber	Medial temporal cortex	Focal ischemia	Clipping and resection of epileptogenic focus	Administration of levetiracetam for six months	Cure

AED: Anti-epileptic drug, MCA: Middle cerebral artery, IC-PC: Internal carotid artery distal to the origin of the posterior communicating artery

Regarding the mechanism underlying the development of partial seizures due to unruptured intracranial aneurysms, potential causes are considered to be direct compression stimulation of the cortex by an aneurysm, minor hemorrhage from an aneurysm, or infarction caused by emboli from an aneurysm.[5,6] However, the present case is characterized by cortical neuron degeneration and edema with gliosis, with no clear hemosiderin deposition in the edematous brain parenchyma surrounding the aneurysm, suggesting that ischemic changes are primarily involved. On the other hand, the changes around the aneurysm on the MRI were restricted within a very narrow region. Therefore, the primary cause is considered to be local circulatory disturbance resulting from the compression by the aneurysm, rather than embolism from the aneurysm.

Majority of patients with unruptured intracranial aneurysms presenting with partial seizures progress to general convulsions. The occurrence of seizures in these patients is considered a warning sign of impending rupture of the aneurysm. Therefore, aneurysmal clipping is performed in most patients. However, the clipping alone, which can prevent the rupture, is considered inadequate from the standpoint of seizure control. Therefore, in the present case, we performed cortical EEG monitoring, resected the aneurysm following clipping, and then confirmed the disappearance of spikes on the cortical EEG after the macroscopically discolored area was completely removed. Consequently, the patient achieved a seizure-free state at the sixth postoperative month. In the literature, a study by Whittle *et al.*[8] in 4 patients with middle cerebral artery aneurysms presenting with focal seizures reported that 2 patients achieved a seizure-free state after clipping and focal resection, whereas seizures could not be controlled in patients receiving surgical wrapping only. Miele *et al.*[5] also reported that seizure control can be obtained only by a reduction of the mass effect of an aneurysm and removal of tissue degenerated by compression, which is detected by MRI. On the other hand, Kuba *et al.*[4] reported patients with middle cerebral artery aneurysms presenting with focal seizures in whom a seizure-free state was achieved within 4 years by coil embolization only. These reports suggest the possibility that seizure control can be obtained by eliminating a pulsatile compressive force applied to the cortex around the aneurysm.

To accurately determine whether resection of epileptic foci is required for control of seizures in such cases would require a randomized controlled trial. Considering the rarity of these lesions, it is unlikely such a trial will ever be conducted, and therefore, decisions regarding the management of these patients will need to be made on an individual basis.

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Conflicts of interest
There are no conflicts of interest.

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