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Case Report

Olfactory hallucinations caused by an unruptured posterior communicating artery aneurysm improved by clipping: A case report with literature review

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ABSTRACT

Background: Unruptured cerebral aneurysms that lead to epilepsy are rare and olfactory hallucinations caused by such an aneurysm are extremely rare. Various treatments have been proposed, including wrapping, clipping with or without cortical resection, and coil embolization, but there is no consensus on the best approach.

Case Description: We present a case of a 69-year-old female who experienced olfactory hallucinations caused by a posterior communicating artery aneurysm and was treated with clipping without cortical resection, with a positive outcome.

Conclusion: According to our knowledge, there has been only one report of a posterior communicating artery aneurysm presenting with olfactory hallucinations has been reported, where clipping and cortical resection were performed. This is the first report of a posterior communicating artery aneurysm with olfactory hallucinations that was effectively treated with clipping alone. There have been a few similar reports of large middle cerebral artery aneurysms, most of which are believed to be caused by entorhinal cortex compression. Although a definitive treatment protocol for this condition remains elusive, we suggest that elimination of the pulsatile compressive stress exerted on the cerebral cortex through surgical clipping or coil embolization is crucial for achieving efficacious seizure management.

Keywords: Clipping, Epilepsy, IC PC aneurysm, Olfactory hallucination, Unraptured aneurysm

INTRODUCTION

Olfactory hallucinations is a symptom of medial temporal lobe epilepsy and is mainly seen in brain tumors of the temporal lobe and hippocampal sclerosis, but is very rare when caused by a unruptured cerebral aneurysm. It is believed that are associated with entorhinal cortex, amygdala, rostral insula, and even with olfactory bulb.[1] This is the first report of treatment of olfactory hallucination resulting from the posterior communicating artery aneurysm through clipping alone, without cortical resection, and achieving a positive outcome.

CASE REPORT

History and imaging

A 69-year-old female patient with no significant prior medical history presented at a local clinic complaining of a persistent olfactory disturbance, characterized as a smell resembling burnt dust, which had persisted for 1 month before her visit. A magnetic resonance imaging (MRI) scan revealed the presence of an aneurysm at the bifurcation of the right internal carotid artery (ICA) and posterior communicating artery. Subsequently, the patient was referred to our clinic for further management. The MRI scan revealed a substantial aneurysm at the bifurcation of the right posterior communicating artery, and the aneurysm was situated within the amygdala, showing high intensity on the fluid attenuated inversion recovery image in the cerebral parenchyma surrounding the aneurysm [Figure 1]. An electroencephalogram revealed no evidence of spikes or abnormal waveforms. A cerebral angiogram confirmed the presence of a 14-millimeter aneurysm in the right posterior communicating artery [Figure 2]. A frontotemporal craniotomy with aneurysmal neck clipping was performed to prevent rupture of the aneurysm and alleviate the patient's olfactory hallucinations.

Surgery and outcome

Under general anesthesia, the ICA was exposed at the level of the neck. A frontotemporal craniotomy was then performed, and the aneurysm was visualized utilizing a trans-sylvian approach. Because the aneurysm was large, neck clipping of the aneurysm was carried out without viewing the entire aneurysm following temporary occlusion of the cervical segment of the ICA [Figure 3]. In the postoperative period, the patient experienced no new symptoms, and the smelly symptoms were resolved by the 2nd postoperative day. Her postoperative course was uneventful, the smelly symptoms were resolved by the 2nd postoperative day, and she was discharged home 2 weeks following the procedure. No antiepileptic drugs were administered postoperatively as no seizures other than olfactory hallucinations had been observed in preoperatively. One year after surgery, an MRI revealed resolution of aneurysm-induced pressure drainage at the medial temporal lobe, and the high intensity surrounding the aneurysm had also disappeared [Figure 1]. Two years postoperatively, there was no recurrence of smelly symptoms, and no seizures were reported.

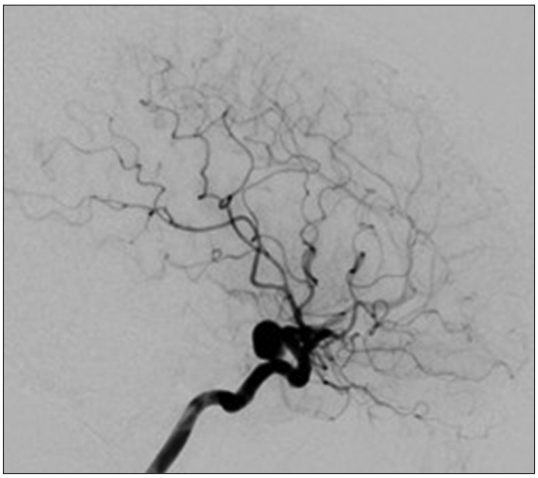

Figure 2: The preoperative cerebral angiogram confirmed the presence of a 14-millimeter aneurysm in the right posterior communicating artery.

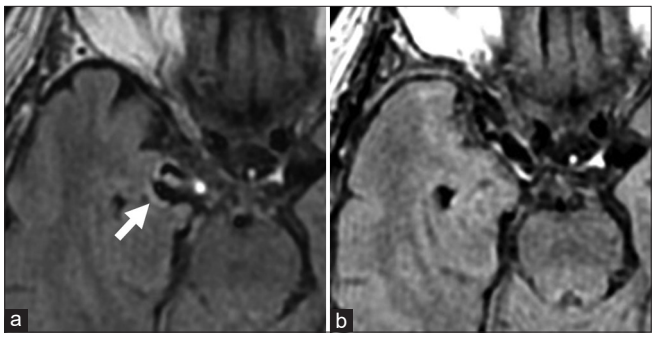

Figure 1: (a) The preoperative state fluid attenuated inversion recovery image in magnetic resonance imaging detected a substantial aneurysm located in the right posterior communicating artery, displaying high intensity in the brain parenchyma surrounding the aneurysm (arrow). (b) The fluid attenuated inversion recovery image of the magnetic resonance imaging scan taken 1 year postsurgery showed resolution of aneurysm-induced pressure drainage at the medial temporal lobe, and the high intensity surrounding the aneurysm had also disappeared.

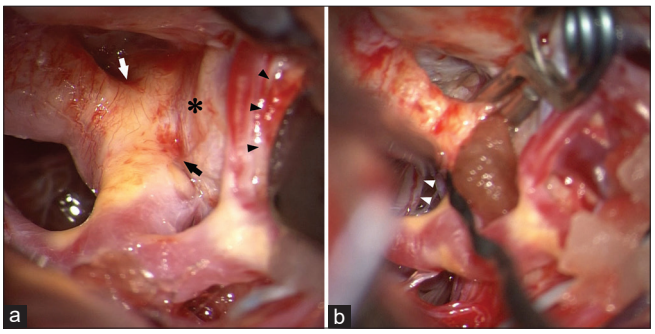

Figure 3: Intraoperative findings of surgical clipping (a) We attained access to the aneurysm (asterisk) with trans-sylvian approach. The aneurysm was found to be in proximity to the temporal lobe (black arrowheads). The black and white arrows indicate the proximal and distal neck of the aneurysm, respectively. (b) Following temporary occlusion of the cervical segment of the internal carotid artery (ICA), neck clipping of the aneurysm was performed with a straight clip. The posterior communicating artery (white arrowheads) was confirmed between the optic nerve and the ICA and preserved.

DISCUSSION

The previous reports have described Olfactory hallucinations are rarely seen, with a reported prevalence of 0.9–16% in particular epileptic patient groups and have been shown to be associated with entorhinal cortex, amygdala, rostral insula, and even with olfactory bulb.[1] 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 one patient.[3] The development of olfactory hallucination in medial temporal lobe tumors and sclerosis is thought to be due to stimulation of part of the olfactory network. Therefore, it is consistent that aneurysms contacting the olfactory tract, lateral olfactory stria, amygdala, parahippocampal gyrus, or entorhinal cortex could be the cause. The previous reports have shown that the fronto-olfactory cortex is involved in olfaction as a secondary olfactory cortex, and there have been reports of symptoms caused by compression of the lateral orbitofrontal cortex by an unruptured middle cerebral artery (MCA) aneurysms. The olfactory hallucination is a rare phenomenon in unruptured cerebral aneurysms, with only five reported cases [Table 1], four of which were caused by a MCA aneurysm, and one case arising from a posterior communicating aneurysm.[2,5,6,9] Among the previous reports, three out of four patients with MCA aneurysm reported the sweet pleasant smell of flowers, sweet fruits, or roses, whereas those with posterior communicating artery aneurysm, the previous report and the present case reported the smell of burnt objects. We suggested that the smell may differ depending on the site of brain compression by the aneurysm. However, the previous studies of olfactory hallucination have reported no relationship between the location of the epileptic focus and the symptoms of olfactory hallucination in simple partial seizures.[8] Further case series are needed to elucidate the relationship between symptoms and site of epileptic focus. The mechanisms of seizure appearance due to unruptured cerebral aneurysms include direct compression stimulation of the cortex by an aneurysm, minor hemorrhage from an aneurysm, infarction caused by emboli from an aneurysm, or local circulatory disturbance resulting from the compression by aneurysm.[2,5,6] In the present case, there was no intraoperative hemosiderin deposition and an MRI revealed abnormal signals in the cortex surrounding the aneurysm. We postulate that the likely mechanism for the olfactory symptoms is local compression or focal ischemia resulting from the aneurysm's compression. In a previous study that examined four patients with MCA aneurysms presenting with focal seizures, reported that two patients achieved a seizure-free state after clipping and focal resection, while seizures could not be controlled in patients who received surgical wrapping alone.[9] Miele *et al*. 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.[5] Kamali *et al*. reported the outcome of epilepsy in four cases of unruptured cerebral aneurysms that developed focal seizures treated through coil embolization. While two patients experienced a few minor seizures post treatment, the seizures in the remaining two patients were fully resolved following treatment.[4] These findings suggest that elimination of the pulsatile compressive force exerted on the cortex through aneurysm clipping or coil embolization is crucial to achieving effective seizure control. However, there is no consensus as to whether cortical resection

Table 1: Reported cases of olfactory hallucination caused by unruptured cerebral aneurysm.

Year/Author	Age/sex	Location/size	Smell symptom	Mechanism of epilepsy	Treatment	Seizure outcome
1985/Whittle *et al*.[9]	52/M	MCA/25 mm	Like roses	Focal compression	Clipping	Controlled
	48/F	MCA/40 mm	Like peeled chokos	Focal compression	Wrapping	Uncontrolled
1999/ Mizobuchi *et al*.[6]	49/F	MCA/10 mm	Like flowers or sweet fruits	Focal compression	Clipping	Cure
2004/Miele *et al*.[5]	55/F	MCA/15 mm	strange smell	Subclinical hemorrhage	Clipping	Controlled
2017/Akimoto *et al*.[2]	70/F	IC-PC/12 mm	Like burnt rubber	Focal ischemia	Clipping and cortical resection	Cure
2023/Present case	69/F	IC-PC/14 mm	Like burnt dust	Focal compression/ focal ischemia	Clipping	Cure

M: Male, F: Female, MCA: Middle cerebral artery, IC-PC: Bifurcation of the internal carotid artery and posterior communicating artery

is imperative. This condition is very rare, and there is no consensus treatment regarding whether coil embolization, clipping, flow diverter, or focal resection should be added. However, it is suggested that wrapping may result in poor control, and at least loss of blood flow within the aneurysm is considered necessary. Further case accumulation is needed for the treatment of this condition in the future.

CONCLUSION

In this study, we present a case of an unruptured posterior communicating artery aneurysm that was accompanied by olfactory hallucinations and was effectively treated through surgical clipping. Although a definitive treatment protocol for this condition remains elusive, we posit that elimination of the pulsatile compressive stress exerted on cerebral cortex through surgical clipping or coil embolization is critical for attaining efficacious seizure management.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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