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*Case Report*

# Difficulty of diagnosing a mucor-induced aneurysm arising in segment P4 of the posterior cerebral artery – A case report

Takao Koiso[1], Yoji Komatsu[1], Yuji Matsumaru[2], Eiichi Ishikawa[2]

[1]Department of Neurosurgery, Hitachi General Hospital, Hitachi, Ibaraki, Japan, [2]Department of Neurosurgery, Faculty of Medicine, University of Tsukuba, Tsukuba, Ibaraki, Japan.

E-mail: *Takao Koiso - s0201534@hotmail.co.jp; Yoji Komatsu - komatsu@md.tsukuba.ac.jp; Yuji Matsumaru - yujimatsumaru@me.com; Eiichi Ishikawa - e-ishikawa@md.tsukuba.ac.jp

***Corresponding author:**
Takao Koiso,
Department of Neurosurgery,
Hitachi General Hospital,
Hitachi, Ibaraki, Japan.

s0201534@hotmail.co.jp

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## ABSTRACT

**Background:** Identification of causative pathogen for fungal aneurysm is frequently difficult. We reported the case of a fungal aneurysm caused by Mucor arising in segment P4 of the posterior cerebral artery (PCA) detected only by histopathological examination.

**Case Description:** A 50-year-old female complained of nausea and vomiting. Computed tomography showed an intracranial hemorrhage in the left occipital lobe and acute hydrocephalus due to intraventricular hemorrhaging. Digital subtraction angiography performed after external drainage showed a cerebral aneurysm in segment P4 of the left PCA. Surgical excision of the aneurysm and end-to-end anastomosis of the PCA were performed. A histopathological examination revealed that the aneurysm had been caused by a Mucor infection.

**Conclusion:** In fungal aneurysm cases, especially those involving Mucor infections, it is difficult to identify the causative fungal infection based on cultures, imaging, and serological tests. Therefore, surgical excision and histopathological diagnosis are important for diagnosing such cases if possible.

**Keywords:** Fungal aneurysm, Histopathological examination, P4 segment

## INTRODUCTION

Infectious cerebral aneurysms include both bacterial and fungal aneurysms. Typically, bacterial aneurysms exhibit a distal location.[4] On the other hand, most fungal aneurysms arise in the proximal portion of the cerebral artery because fungi usually invade from an adjacent organ.[1,6,10,12,14,16,22] To diagnose, fungal infection is often difficult based on clinical manifestations, serological methods, cultures, molecular methods, or imaging.[11] This is a case report of a fungal aneurysm caused by Mucor arising in segment P4 of the posterior cerebral artery (PCA) in which only a pathological examination was diagnostically useful. Our discussion includes details of diagnostic methods for fungal aneurysm.

## CASE REPORT

A 50-year-old female presented with nausea and vomiting. She had a history of ulcerative colitis and had used steroids for over 30 years. She also had undergone aortic root surgery and

mechanical valve insertion for an aneurysm of the ascending aorta at the age of 48, and hence, was taking a Vitamin K antagonist. A clinical examination showed an impairment of consciousness (Glasgow Coma Scale [GCS] score: E3V4M6). Her systolic blood pressure had increased to >200 mmHg. Her body temperature was normal.

Laboratory studies showed a slightly elevated white blood cell (WBC) count and anemia. Her WBC was 11300/μL (normal, 3500–9000) and her hemoglobin level was 11.1 g/dL (normal, 11.5–16.6). In addition, her C-reactive protein level was 0.17 mg/dL (normal, <0.3). In coagulation tests, it was found that the patient's prothrombin time-international normalized ratio was elevated to 2.47, and her D-dimer level had also increased to 5.7 μg/mL (normal, <1.0). Two sets of blood cultures and a urinary culture were negative.

Brain computed tomography (CT) revealed a subcortical hemorrhage in the left occipital lobe and acute hydrocephalus due to intraventricular hemorrhaging [Figure 1a]. No fluid or bone destruction was seen in the paranasal sinuses [Figure 1b]. 3D-CT angiography revealed a cerebral aneurysm in the distal left PCA [Figure 1c]. During the examination, the patient suddenly became comatose. Emergency intubation and bilateral external ventricular drainage were performed under general anesthesia. On day 2, her consciousness level improved (GCS score: E1VtM4), and digital subtraction angiography was performed. The left vertebral angiography revealed a wide-necked aneurysm in the left parietooccipital artery (POA) [Figures 2a and b]. Its maximum diameter was 5.6 mm. To prevent rebleeding, direct surgery was performed on the same day.

The patient's head was fixed in the prone position using a Sugita frame and the left occipital craniotomy was carried out. The location of the hematoma was confirmed by ultrasonography. A corticotomy was made in the superior occipital gyrus and the hematoma was approached [Figure 3a]. The aneurysm was identified after removing the hematoma. The proximal and distal portion of parent artery were exposed and the location of the neck of the aneurysm was confirmed [Figure 3b]. The parent artery had adhered to the neck of the aneurysm and an intra-aneurysmal thrombosis was found. As neck clipping would have been difficult due to the fragility of the neck of the aneurysm, the aneurysm was removed, and end-to-end anastomosis of the normal parts of the proximal and distal POA was performed [Figures 3c and d]. A histopathological examination revealed that the elastic fibers had disappeared from the aneurysm wall and only fibrotic tissue was seen [Figures 4a and b]. On the outside of the aneurysm, neutrophils and lymphocytes gathered and necrotic tissue was seen [Figure 4c]. There were coenocytic hyphae in the necrotic tissue. Immunohistochemical analysis performed with Grocott's stain revealed that the coenocytic hyphae varied in width and exhibited right-angled branching, which are characteristics of zygomycetes [Figure 4d]. These findings were consistent with Mucormycosis.

Postoperative CT and magnetic resonance imaging only revealed a small hematoma in the left occipital lobe, but no cerebral infarction. As status epilepticus occurred on postoperative day (POD) 1, the propofol-induced general anesthesia was maintained, and anticonvulsant drugs were administered. The patient's cerebrospinal fluid (CSF) showed a slightly elevated cell count (117/μL) and a normal glucose level (96 mg/dL) on POD 7. Tests for serum β-D-glucan and *Aspergillus* antigen were negative on POD 9. After a definitive diagnosis was made based on a histopathological examination on POD 10, we administered 600 mg/day voriconazole. The patient's consciousness level gradually improved and she was extubated on POD 10. Heparinization was performed to prevent thromboembolic events from POD 18. The source of the infection was not found during transesophageal echocardiography performed on POD 21 or contrast-enhanced whole-body CT conducted on POD 22. Six blood cultures and a CSF culture obtained after surgery were all negative. The patient's condition suddenly worsened after vomiting on POD 31 and she died on POD 32. An autopsy revealed pneumonia in the dorsal section of the right lung. Aspiration pneumonia might have been the cause of death. No systemic fungal infection was found.

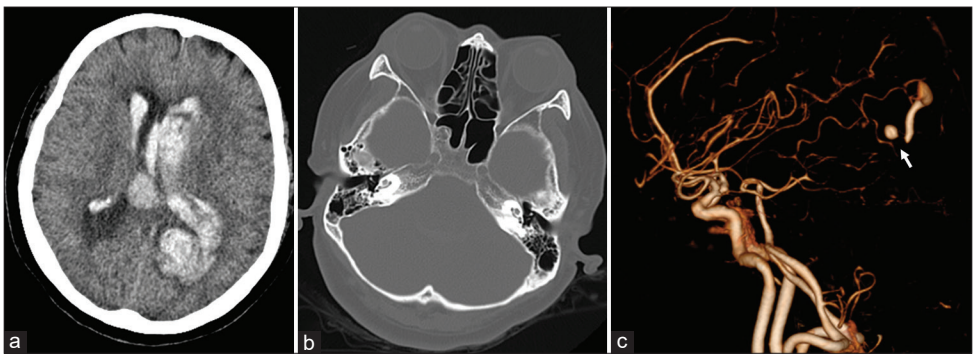


**Figure 1:** Computed tomography (CT) (a and b) and CT angiography (c) scans performed on admission (a) Brain window image showing an intracerebral hemorrhage in the left occipital lobe and an intraventricular hemorrhage (b) Bone window image showing no bone destruction or fluid storage in the paranasal sinus (c) CT angiography showed a cerebral aneurysm (arrow) in the distal left posterior cerebral artery.

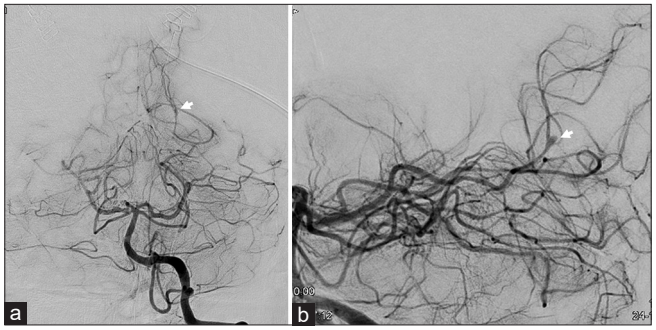


**Figure 2:** Anteroposterior (a) and lateral (b) left vertebral angiograms showing a cerebral aneurysm in segment P4 of the left posterior cerebral artery (arrow).

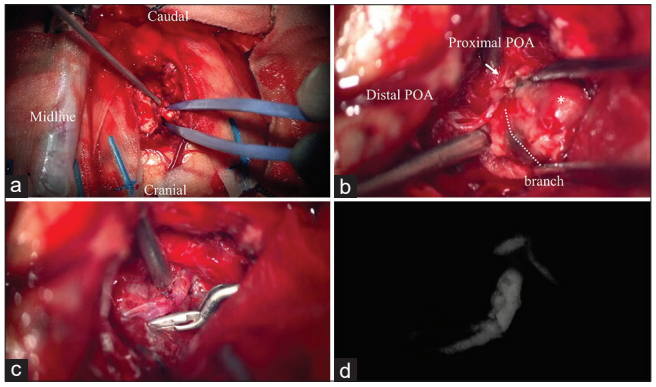


**Figure 3:** Intraoperative photographs. (a) Left occipital craniotomy and corticotomy in the superior occipital gyrus were performed. (b) The proximal (arrow) and distal (arrowhead) posterior occipital artery were exposed. Intra-aneurysmal thrombosis was found (asterisk). (c) The aneurysm was removed, and end-to-end anastomosis of the normal sections of the proximal and distal POA was performed. (d) Indocyanine green video angiography confirmed the patency of the bypass.

## DISCUSSION

This was a reported case of a fungal aneurysm arising in segment P4 of the PCA. Mucor was the cause of the fungal aneurysm. A histopathological examination was required to reach a diagnosis in this case.

In cases of fungal aneurysms, confirming a fungal infection using cultures, imaging, or serological tests is often difficult.[6,12,16] In such cases, a histopathological examination is required to diagnose the fungal infection. In cases of mycotic aneurysms, the pathogen cannot be identified in 10–12.5% of cases.[3] Kannoth *et al.* reported that the causative pathogen could not be identified before death, but was identified at autopsy, in two of 21 cases of bacterial aneurysms and three of four cases of fungal aneurysms.[10] Therefore, it is more difficult to identify the causative pathogen in cases of fungal infections than in cases of bacterial infections. The positive predictive values of blood or CSF cultures for diagnosing infectious

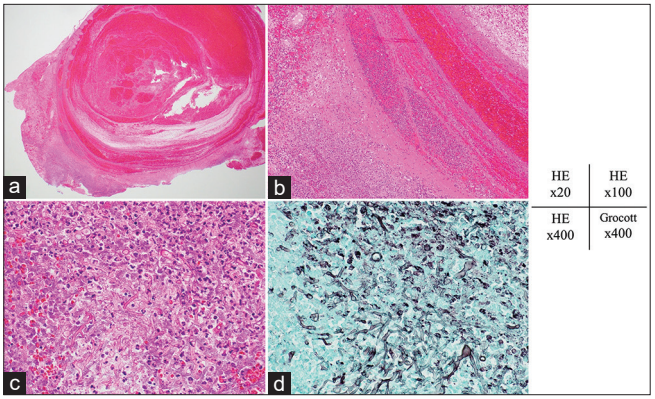


**Figure 4:** Hematoxylin and eosin staining. (a and b) Photomicrographs showing that the elastic fibers had disappeared from the aneurysm wall and only fibrotic tissue was present. (c) Photomicrograph of the outside of the aneurysm showing the accumulation of neutrophils and lymphocytes, and coenocytic hyphae in the necrotic tissue. (d) Photomicrograph obtained after staining using Grocott's method showing coenocytic hyphae, which varied in width and exhibited right-angled branching. These findings are characteristic of zygomycetes. Original magnification: (a) ×20, (b) ×100, (c) ×400, and (d) ×400.

aneurysms were reported to range from 28 to 47%.[10,15,20] Especially, in cases involving Mucor, cultures frequently produce negative results.[23] On the other hand, the negative predictive value of the serum β-D-glucan level for diagnosing fungal infections was reported to be 99.6%.[5] Therefore, the serum β-D-glucan is useful for excluding a diagnosis of fungal infection; however, it is not elevated in cases of Mucor infections because the amount of β-D-glucan produced by Mucor is very small.[5] In cases of fungal infections caused by *Aspergillus*, antigen tests are diagnostically useful. The sensitivity of such antigen testing was reported to range from 83 to 100% and its specificity was reported to range from 86 to 97.5%.[19] However, no Mucor antigen tests are currently available, and hence, a histopathological examination is needed if culture tests show no fungi or bacteria.

In some cases of fungal aneurysms due to sinusitis, it is possible to determine the causative fungi through a histopathological examination of the sinus.[1,13,14] On the other hand, in cases of fungal aneurysms caused by hematogenous seeding or in which the source of infection is unknown, it can be difficult to identify the causative fungi without performing a histopathological examination of the aneurysm.[2,9,16,18]

Some infectious aneurysms have recently been treated using endovascular therapy.[1,7,8,17,21] In some of these cases, relatively good outcomes were reported.[1,7] On the other hand, Hirai *et al.* reported a case in which an infectious aneurysm reruptured after coil embolization.[6] Endovascular treatment for infectious aneurysms is not well established. Moreover, pathological specimens cannot be collected without direct surgery. Direct surgery might be better for

suspected infectious aneurysms if the patient's condition allows them to tolerate surgery, and the aneurysm can be treated with minimal risk of neurological deterioration. Neck clipping is sometimes difficult due to the fragility of infectious aneurysms. The primary treatment for a ruptured infectious aneurysm is trapping, but bypass surgery should be employed if the parent artery perfuses a broad or eloquent area.

## CONCLUSION

We described a case of a Mucor aneurysm arising in segment P4 of the PCA. In this case, making a definitive diagnosis is difficult without a pathological examination. If you cannot identify pathogen, surgical excision and pathological examination of the aneurysm should be considered.

### 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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