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Case Report

A case of subarachnoid and intracerebral hemorrhages complicated by trichosporonosis

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ABSTRACT

Background: Trichosporonosis has an extremely poor prognosis. In this report, we describe a case of subarachnoid hemorrhage and intracerebral hemorrhage due to a fungal aneurysm caused by *Trichosporon*.

Case Description: A 71-year-old woman who experienced subcortical hemorrhage developed a subarachnoid hemorrhage. Endovascular parent artery occlusion was performed for a fungal aneurysm in the left posterior cerebral artery caused by *Trichosporon*. After surgery, voriconazole and liposomal amphotericin B were administered. The patient died of massive left putamen hemorrhage.

Conclusion: Effective treatment for intracranial hemorrhage due to trichosporonosis has not yet been established and an accumulation of cases is required.

Keywords: Fungal aneurysm, Intracerebral hemorrhage, Subarachnoid hemorrhage, Trichosporonosis

INTRODUCTION

Trichosporon is an opportunistically infective yeast-like fungus. About 74.7% of *Trichosporon* infections reach the bloodstream, which is associated with an extremely poor prognosis.[2] Infectious cerebral aneurysms are caused by cerebral artery wall destruction due to infection and are relatively rare, accounting for <1% of intracranial aneurysms.[7] Causative organisms are usually *Streptococcus* (30%–44%), *Staphylococcus* (14%–18%), or unknown (10–12.5%).[4] There have been no reports of infected aneurysms caused by *Trichosporon*.

We describe a case of subarachnoid hemorrhage due to rupture of a fungal aneurysm caused by *Trichosporon*, resulting in recurrent intracerebral hemorrhage and death.

CLINICAL SUMMARY

The patient is a 71-year-old woman who underwent surgical aortic mechanical valve replacement for aortic dissection and was started on warfarin postoperatively. Seven months after her aortic valve replacement surgery, she experienced her first intracerebral hemorrhage. No other underlying medical conditions, such as diabetes or immunodeficiencies, were noted. The hemorrhage was subcortical, in the right parietal lobe. The hematoma was removed by

craniotomy in the neurosurgery department of another hospital (day 0) [Figure 1].

Thirty-eight days after the onset of the first hemorrhage (day 38), the patient was transferred to our hospital for convalescent rehabilitation. At that time, neurological findings included a Japanese Coma Scale 1 and GCS 15 (E4 V5 M6) level of consciousness, left unilateral spatial neglect, left facial paralysis, and left hemiplegia. Thirteen days after transfer (day 51), the patient developed a fever of unknown etiology, and antibiotic therapy (levofloxacin, ceftriaxone, and meropenem) was administered for possible urinary tract infection and meningitis. The patient's laboratory results during the course of the fever included white blood cell count 11600/ul, C-reactive protein 3.76 mg/L, procalcitonin 0.18 ng/mg, and βD glucan 7.2 pg/ml. Cerebrospinal fluid examination revealed a cell count of 46.2/μL (57 mononuclear cells and 43 polymorphonuclear cells), 70 mg/dL protein, and 45 mg/dL sugar. Echocardiography showed an echo-free around the prosthetic valve, but coronary computed tomography (CT) showed no obvious abscess formation around the valve.

Three days later (day 54), the patient developed subarachnoid hemorrhage and was referred to our department [Figure 2a]. Cerebral angiography showed an aneurysm in the P4 segment of the left posterior cerebral artery (PCA) [Figure 2b], and endovascular parent artery occlusion was performed with a coil on the same day. A 5Fr Fubuki Dilator Kit (ASAHI INTECC, Aichi, Japan) was inserted into the left vertebral artery through the left brachial artery. A microcatheter (1.7Fr RESTAR, Medico's Hirata, Osaka, Japan) was guided to the distal part of the aneurysm neck using a 3.2Fr TACTICS (Technocrat, Aichi, Japan) as the distal access catheter [Figure 3a] and filled with a total of 12 coils in a sequential refilling fashion to complete the procedure [Figures 3b and c]. Intraoperatively, blood was collected from a microcatheter tip near the aneurysm, from which *Trichosporon* species were cultured.

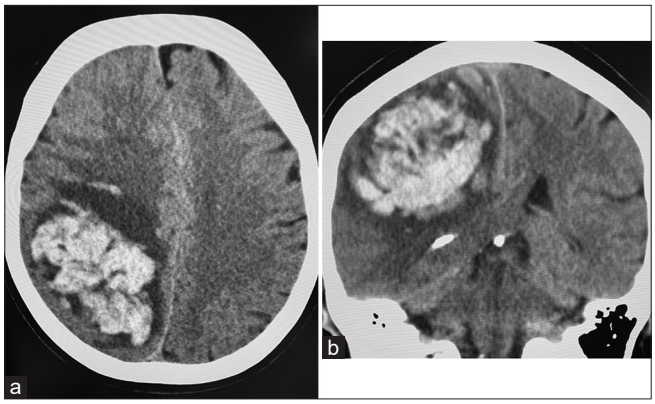

Figure 1: Head computed tomography (CT) at first onset of intracerebral hemorrhage. A hematoma 7 cm in size was found in the right parietal lobe in axial (a) and coronal (b) view.

Postoperatively, the patient was treated for vasospasm and started on voriconazole (VRCZ) as an antifungal. On the 3rd postoperative day (day 57), head CT showed asymptomatic subcortical hemorrhage in the left temporal lobe [Figure 4a]. Liposomal amphotericin B (L-AMB) was added to the VRCZ regimen and the blood culture was negative for *Trichosporon*. Contrast-enhanced CT 1 month after the operation (day 85) showed multiple diffuse microaneurysms [Figure 4b]. One week later (day 92), a massive intracerebral hemorrhage in the left putamen resulted in cerebral herniation and the patient died [Figure 4c]. The prothrombin time-international normalized ratio (PT-INR) immediately before death was 4.41, suggesting that the interaction between VRCZ and warfarin caused an overextension of the PT-INR. There were no obvious findings of disseminated intravascular coagulation. Autopsy was not performed because patient's family did not wish it.

DISCUSSION

Trichosporon species are present in the human microbiota of skin and gastrointestinal tract.[11] Deep-seated *Trichosporon* is an opportunistic infection with a poor prognosis. Risk factors for the onset of the disease include neutropenia, steroid therapy, intensive care unit admission, and highly invasive surgery. The mortality rate has been reported to be 60–90%.[16] Furthermore, *Trichosporon* infections of the central nervous system are rare. Although there have been reports of abscesses and meningitis, fungal aneurysms caused by trichosporonosis have not been reported.[1,5,6,8,10-15] This is the first report of an intracranial fungal aneurysm caused by trichosporonosis. *Trichosporon* species are naturally resistant to candin antifungals, so there is a risk of breakthrough infection in case of inadvertent use.[9] As for *Trichosporon* treatment, some reports indicate that both VRCZ and L-AMB improve prognosis. The patient was treated with VRCZ and L-AMB and blood cultures were negative for *Trichosporon*. However, it is thought that the VRCZ enhanced the effect of warfarin through interaction, resulting in fatal

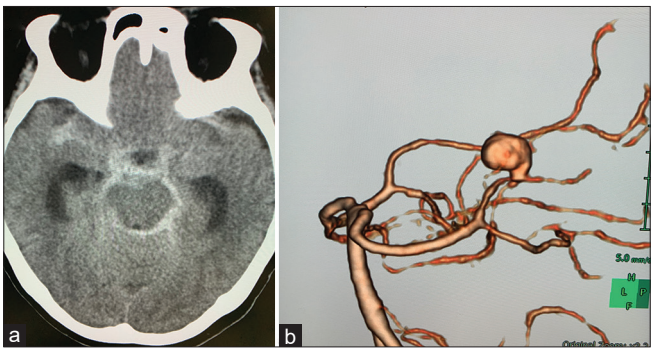

Figure 2: (a) Head CT showed subarachnoid hemorrhage. (b) Three-dimensional reconstruction of CT angiography showed aneurysm in the P4 segment of the left posterior cerebral artery.

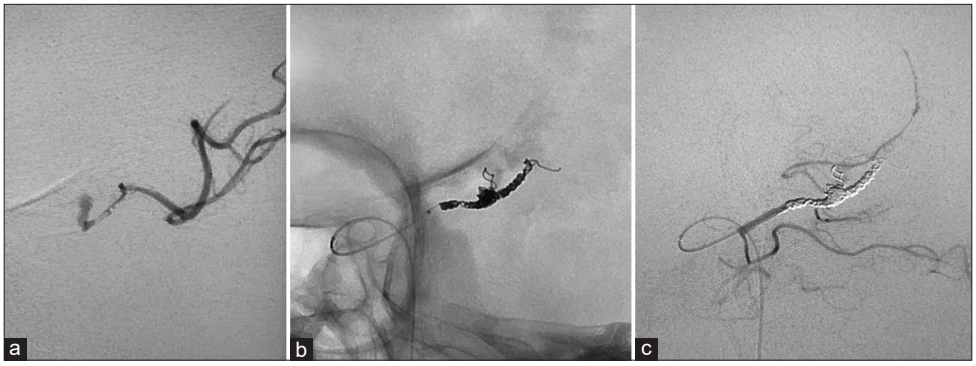

Figure 3: Endovascular parent artery occlusion, (a) A microcatheter (Lester STR 1.7–2.3Fr 157 cm) was guided to the distal neck of the aneurysm. (b) A total of 12 coils were filled with sequential backfilling. (c) The aneurysm was no longer depicted after occlusion of the parent artery.

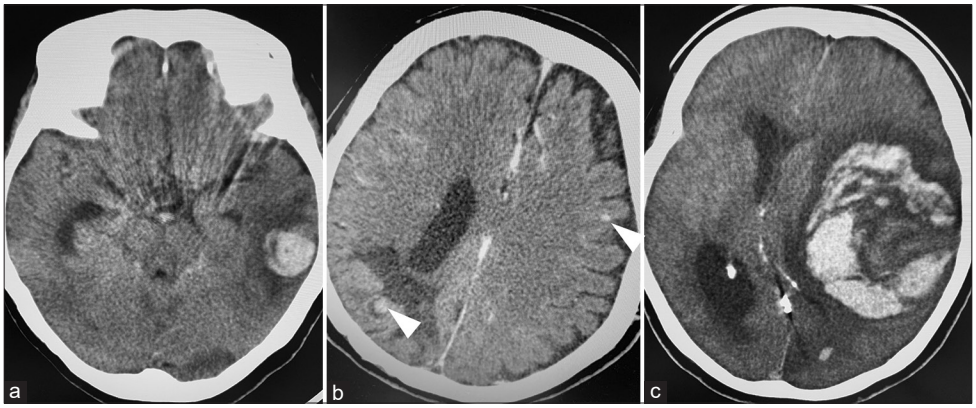

Figure 4: Time course of head CT scan after endovascular parent artery occlusion. On postoperative day 3 (day 57), head CT showed left temporal lobe subcortical hemorrhage (a) on day 85, head contrast-enhanced CT showed multiple microaneurysms (arrow heads) (b) on day 92, head CT showed left putamen hemorrhage with cerebral herniation (c).

bleeding. This could be considered a pitfall in the treatment of *Trichosporon*. The mechanism is inferred to be bleeding from an infected aneurysm, which was contributed to by an over-prolonged PT-INR, leading to a cerebral herniation. The route of invasion was unknown, but, as there was a history of valve replacement, one would suspect that it may have been the source of infection. Coronary CT did not demonstrate any abscess around the prosthetic valve so that remains unconfirmed. Despite successful treatment of the fungal infection using VRCZ and L-AMB, the patient developed multiple microaneurysms and passed away.

As for fungal aneurysms, the known routes of entry include hematogenous dissemination, direct invasion, and surgical contamination. The most frequently reported fungi are *Aspergillus, Candida, Zygomycetes,* and *Coccidiodes.* There are no reports of intracranial aneurysms caused by *Trichosporon*. Aneurysms caused by direct invasion are more common in proximal arteries, such as the internal carotid artery, while those caused by blood dissemination are more common in distal arteries, such as the anterior cerebral artery, middle cerebral artery, and PCA. Reports of both craniotomy and endovascular surgery indicate that the location and shape of the aneurysm determine which is more appropriate for management. Endovascular parent vessel occlusion may be indicated for distally infected aneurysms.[3,17] In this case, we performed endovascular parent artery occlusion for an aneurysm caused by *Trichosporon* distal to the PCA.

An accumulation of reports is needed to formulate a proper plan of management in such cases. Furthermore, the clinical summary may aid clinicians in the diagnosis and treatment of similar rare occurrences of trichosporonosis infection.

CONCLUSION

We report an unprecedented case of intracranial fungal aneurysm caused by trichosporonosis. Effective intracranial trichosporonosis management has not yet been established and the treatment for such cases with subarachnoid or intracerebral hemorrhage requires careful analysis of multiple cases.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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