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Case Report

# Ruptured Mycotic Cerebral Aneurysm Secondary to Disseminated Nocardiosis

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## Abstract

We report a case of a ruptured mycotic cerebral aneurysm caused by Nocardia infection. A 22-year-old immunocompromised woman with adult-onset Still's disease developed a subarachnoid hemorrhage (SAH). Digital subtraction angiography revealed a small aneurysm at the M2-3 bifurcation of the right middle cerebral artery. Cardiac ultrasonography showed vegetation at the posterior cardiac wall, suspecting infective endocarditis (IE). Gram-positive filamentous bacteria were observed in the necrotic tissue surrounding the aneurysm obtained during trapping surgery. Long-term blood culture showed that the cause of her cerebral mycotic aneurysm was nocardiosis. A mycotic ruptured cerebral aneurysm is an important cause of SAH in immunocompromised patients. Early diagnosis of IE, detection of gram-positive rods by Gram staining, and long-term culture to identify the bacteria is crucial in diagnosing nocardiosis.

**Keywords:** *Immunosuppressed host, nocardiosis, ruptured mycotic cerebral aneurysm*

## Introduction

Nocardia is a gram-positive rod belonging to the actinomycete Nocardia family, Nocardia order.[1] They are widely present in clod, and recently, the prevalence of the opportunistic infection nocardiosis, which affects compromised hosts, has increased.[2,3]

Here, we present the case of a 22-year-old woman, who was immunosuppressed, with a ruptured intracranial aneurysm caused by a Nocardia infection and required surgery. Pathological analysis of the aneurysm clearly indicated the presence of Nocardia.

Cases of ruptured cerebral mycotic aneurysm caused by Nocardia infection and showing the pathology of intraaneurysm Nocardia are rarely reported.[4]

## Case Report

The patient was a 22-year-old woman diagnosed with adult-onset Still's disease and received chronic steroid and immunosuppression therapy for 5 years. She presented with a sudden disturbance in consciousness. The head computed tomography scan revealed subarachnoid hemorrhage (SAH) (World Federation of Neurosurgical Societies: Grade III) and intracerebral hemorrhage [Figure 1].

Digital subtraction angiography (DSA) showed a small aneurysm at the bifurcation of the right middle cerebral artery (M2-3) [Figure 2]. Preoperative cardiac ultrasonography revealed vegetation on the posterior wall of the heart, measuring approximately 7 mm × 8 mm × 10 mm; therefore, mycotic ruptured aneurysm with infective endocarditis (IE) was suspected. Emergent trapping of the ruptured aneurysm of M2-3 middle cerebral artery, hematoma removal, and decompression craniotomy was performed. In the pus-filled aneurysm, the wall of the aneurysm was turbid and white and had irregularities [Figure 3a]. Pathological analysis showed the destruction of the aneurysm wall structure [Figure 3b], and Gram-positive filamentous bacteria were observed in the necrosis surrounding the aneurysm [Figure 3c and d].

Blood culture was performed after the surgery because mycosis was observed in the aneurysm sample. Gram-positive rods were also noted in the blood culture another 4 days later, and nocardiosis was identified 11 days after performing the blood culture.



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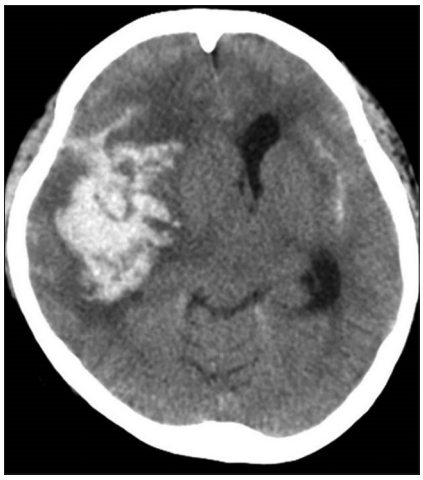

**Figure 1: Head computed tomography scan at the onset of subarachnoid hemorrhage demonstrates diffuse subarachnoid hemorrhage with intracranial hematoma**

We determined that the cause of her cerebral mycotic aneurysm ruptured was nocardiosis. Combination therapy with imipenem/cilastatin (IPM/CS) and amikacin was started and was continued for 3 months. As a hyposensitization therapy, trimethoprim/sulfamethoxazole (TMP/SMX) administration was initiated at 40 days postoperatively and was continued for 6 months. Her symptoms of disturbed consciousness, cognitive function, and hemiparesis gradually improved. She was transferred to another hospital 7 months after medical treatment and rehabilitation.

## Discussion

Nocardia is a Gram-positive rod bacterium belonging to the actinomycete Nocardia family, Nocardia order, which is widely present in clod.[1] The risk of infection has recently increased, especially in patients on steroid and immunosuppressant therapy for a long time and those with high blood sugar levels, lymphoma, malignant tumor, and human immunodeficiency virus infection.[2,3]

Our present case is a 22-year-old woman diagnosed with adult-onset Still's disease and received chronic steroid and immunosuppression therapy for 5 years.

There were not any clinical signs of endocarditis or multi-organ failure, but cardiac ultrasonography immediately after the onset of SAH showed vegetation of the posterior wall of the heart, so mycotic ruptured aneurysm with IE was suspected.

Nocardiosis is classified into three groups: pulmonary form, skin form, and dissemination form. Hematogenous dissemination could cause IE and lead to the development of a mycotic cerebral aneurysm.[5-7] Mycotic aneurysm associated with IE occurs in approximately 2% of all patients with cerebral aneurysms[8] and appears more commonly in the middle cerebral artery distal branch.[9]

In our case, DSA demonstrates a small aneurysm at the right M2-3 bifurcation and pathological findings showed an agglomeration of Gram-positive rods with a thread-like form in the disrupted aneurysm wall. Blood culture was performed after the surgery, and Gram-positive rods were also noted in the blood culture, so we determined that the cause of her cerebral aneurysm ruptured was IE due to the mold.

Actinomycetes, which are virulent, are categorized into two main types. One is anaerobes (Actinomycete genus) and another is aerobes (Nocardia genus); both cause opportunistic infections and result in systemic dissemination. Distinguishing the species is important to select the proper antibiotic therapy against actinomycetes,[10] but the diagnosis of nocardiosis is difficult because the growth speed of Nocardia is slower than other bacteria. Culturing for >1 week will be needed to identify Nocardia. Nocardia could be identified by Kinyoun staining, which distinguishes actinomycetes because the cell wall is stained by the mycolic acid stain.[11]

In our case, Nocardia was identified by Kinyoun staining, which took 11 days after the blood culture.

Previous reports showed that 4 of 10 patients (40%) treated only with antibiotics died, whereas 2 of 16 surgically treated patients (12.5%) died;[12] the mortality rate of the unruptured aneurysm is 30% and that of ruptured aneurysms is 80%.[7] Therefore, surgical intervention in the acute phase could reduce the mortality rate of ruptured mycotic aneurysms. As an antibiotic treatment, TMP/SMX could be administered[13] and TMP/SMX therapy should be continued for 12 months in an immunocompromised host with a central neurological disease, such as the presence of multiple brain abscesses.[13]

In our case, emergent trapping of ruptured aneurysm of M2-3 middle cerebral artery, hematoma removal, and decompression craniotomy was performed. After that, combination therapy with IPM/CS and amikacin was started considering the resistance of Nocardia to antibacterial drugs.[14,15] Then, TMP/SMX therapy was started as a hyposensitization therapy 40 days after onset and continued for 6 months. Finally, nocardiosis was ameliorated.

In conclusion, we reported a case of a ruptured mycotic cerebral aneurysm owing to nocardiosis. A mycotic ruptured cerebral aneurysm is an important cause of SAH in patients who are immunocompromised. Early diagnosis of IE, detection of Gram-positive rods by Gram staining, and long-term culture to identify the bacteria are crucial in diagnosing nocardiosis.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts

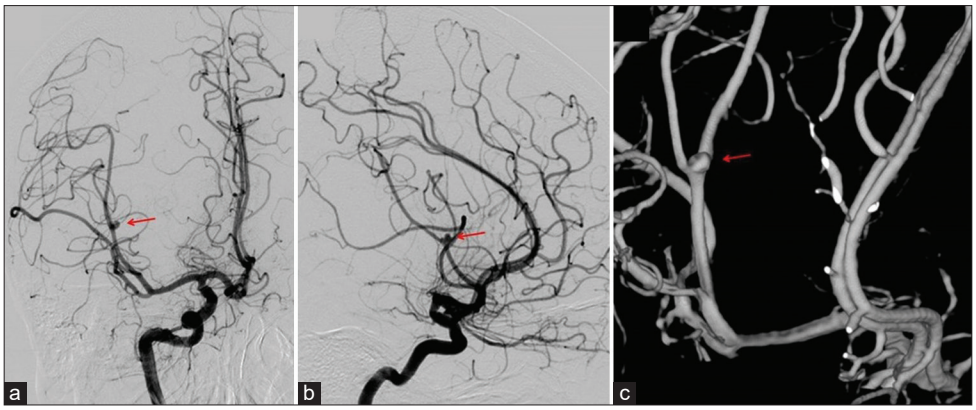

**Figure 2: Digital subtraction angiography at the onset of subarachnoid hemorrhage demonstrates a small aneurysm at the right M2-3 bifurcation (red arrow). (a) Anteroposterior view, (b) Lateral view, (c) 3D Rotational Angiography**

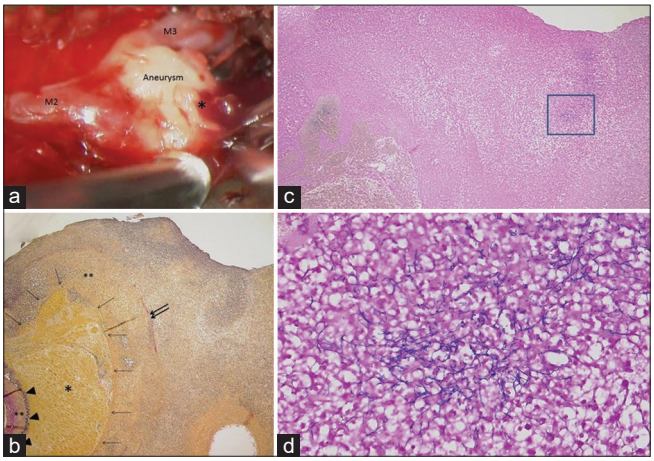


**Figure 3: Clinical and pathological findings of aneurysm (a) The aneurysm filled with pus, which leaked out from the point of destruction of the aneurysm wall (*). The aneurysm wall appears white and turbid with irregularities (b) Pathological analysis demonstrates an internal elastic lamina (arrow head), intravascular lumen (*), expanded media (**), destruction of internal elastic lamina (arrow), and persisting outer membrane (double arrows) (c and d) Gram-positive filamentous bacteria detected in the necrosis surrounding the aneurysm**

will be made to conceal identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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