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Case Report

A rare case of spontaneous thrombosis in saccular cerebral aneurysm in a patient with subarachnoid hemorrhage ☆,☆☆

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ABSTRACT

Cerebral aneurysm is a known cause of spontaneous subarachnoid hemorrhage (SAH). Furthermore, this condition is often asymptomatic, but the occurrence of a rupture can lead to fatal complications. The incidence of spontaneous thrombosis in saccular aneurysm is rare, with an incidence rate of 1%-2%. The most common sites include the middle cerebral artery (MCA) (41%), posterior communicating artery (PCOMM) (15%), and posterior inferior cerebellar artery (PICA) (11%). A head computed tomography angiography (CTA) with contrast is a common diagnostic tool for detecting SAH in the temporoparietal area, hippocampal gyrus, and right fissure of Sylvie. In some cases, saccular aneurysm can be found in the segment bifurcation of the right middle cerebral artery. A cerebral angiography was carried out, specifically digital subtraction angiography, which revealed the presence of visible blister remnants of aneurysm in the form of spontaneous thrombosis. After 1 year, another angiography evaluation was performed to assess the condition of the patient. Furthermore, the results showed no evidence of recanalization and there were no new neurologic deficits. Although spontaneous thrombosis led to the healing of aneurysm in some cases, secondary recanalization remained a possibility. Therefore, it was essential to monitor any incidence of this complication. Precise knowledge of the mechanism of spontaneous thrombosis could lead to the development of new therapeutic approaches. Spontaneous thrombosis in cases of saccular aneurysm is a rare occurrence, which can provide temporary or permanent benefits to the patient. Therefore, periodic evaluation is required to assess the condition of the patient.

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Introduction

Cerebral aneurysm is often caused by the protrusion of a brain blood vessel, leading to the weakening of its wall [1]. An unruptured type is usually asymptomatic in some patients and can be detected using *computed tomography angiography* (CTA) or a detailed *medical check-up*. Furthermore, enlarged and ruptured aneurysm often caused brain hemorrhage in the cranial cavity, ventricles, and subarachnoid [2]. Saccular aneurysm is generally considered to have a higher risk of rupture compared to fusiform [3].

This condition can occasionally present with several complications, including partial or total spontaneous thrombosis. The occurrence of this complication is very rare, with the prevalence of thrombosis in saccular aneurysm estimated to be around 1%-2% [4]. Spontaneous intra-aneurysmal thrombosis is also a well-documented phenomenon and has been noted in approximately 50% of giant aneurysm. However, the incidence of total aneurysmal thrombosis is significantly lower and ranges between 13% and 20% [5]. Spontaneous thrombosis is uncommon in non-giant aneurysm, and its occurrence is rarely reported. The most common sites include the *middle cerebral artery* (MCA) (41%), *posterior communicating artery* (Pcomm) (15%), and *posterior inferior cerebellar artery* (PICA) (11%) [4].

The occurrence of a thrombotic cerebral aneurysm after rupture is associated with hypotension, vasospasm, and damage to the arterial wall. The reported incidence is 1%-2%, which can increase to 3% in patients treated with antifibrinolytic agents [5]. This report presents a case of a patient with an unruptured cerebral aneurysm that was completely thrombosed.

Case reports

A woman aged 40 years came to the Emergency Room at Dr Soetomo General Hospital Indonesia, with the main complaint of severe and consistent headaches for the last 1 day. The patient also complained of seizures, which were characterized by unconsciousness, as well as stomping of the hands and feet. Furthermore, there were no complaints of facial cramps, sluggishness, body weakness or disturbances of urination and defecation. The patient had underlying hypertension but was not consistent with the use of the prescribed medications. Physical examination showed normal vital signs and high alertness, with focal neurological deficits in the form of neck stiffness.

The complete blood count laboratory examination showed normal results. In the *computed tomography* (CT) scan of the head without contrast, there was an extensive subarachnoid hemorrhage, bilaterally, accompanied by mild diffuse cerebral edema, as shown in Fig. 1. Follow-up examination with CTA of the head showed SAH in the temporoparietal region, hippocampal gyrus, and right sylvian fissure. There was also saccular aneurysm in the M2 segment of the right MCA, positioned posterosuperior with a length of 4.3 mm and width of 3.4 mm, as shown in Fig. 2.

The patient was given medications including 60 mg nimodipine every 4 hours and 100 mg phenytoin 100 mg every 8 hours, with other supportive therapy. Subsequently, the conditions improved and there was no headache and recurrent seizures. The patient was then subjected to DSA ad hoc coiling for post-treatment examination. The results of the DSA examination showed no dome aneurysm in the right MCA bifurcation, but aneurysmal remnants in the form of blisters were still visible. A diagnosis of spontaneous total thrombosis in aneurysm was then made, as shown in Fig. 3.

During the 1-year follow-up, the patient did not experience any focal neurological deficits. Re-evaluation was also carried out using DSA, with the results still showing a residual neck aneurysm resembling a blister formation on the right MCA bifurcation and there was no recanalization and infarction, as shown in Fig. 3.

Discussion

Treatment to prevent further complications from an aneurysm typically involved occlusion through *endovascular coiling* or *clipping*. However, in some cases, the aneurysm could occlude naturally, which was commonly referred to as spontaneous thrombosis. The reported incidence of spontaneous thrombosis was between 1% and 2%, with the prevalence of thrombosis in saccular aneurysm being 1%-2%. The most common sites for aneurysmal thrombosis included MCA (41%), Pcomm (15%), and PICA (11%) [4].

Several literature reports showed that spontaneous thrombosis could lead to complications in the form of ischemic stroke. For example, Whittle described the success of aneurys-

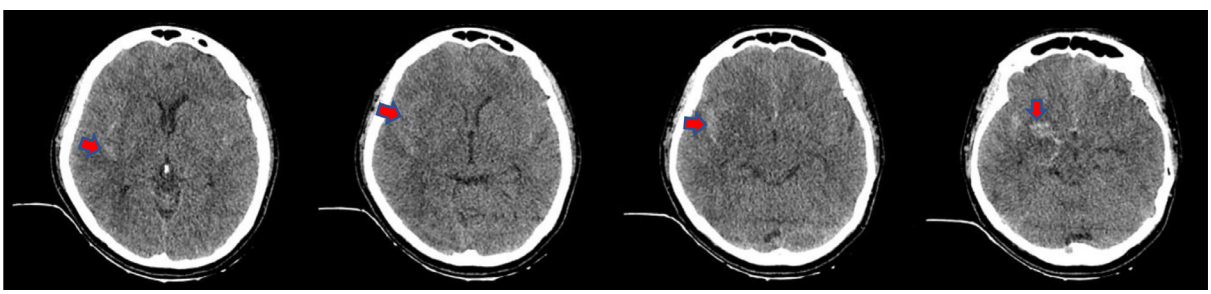

Fig. 1 – CT scan results dominant SAH on the right side.

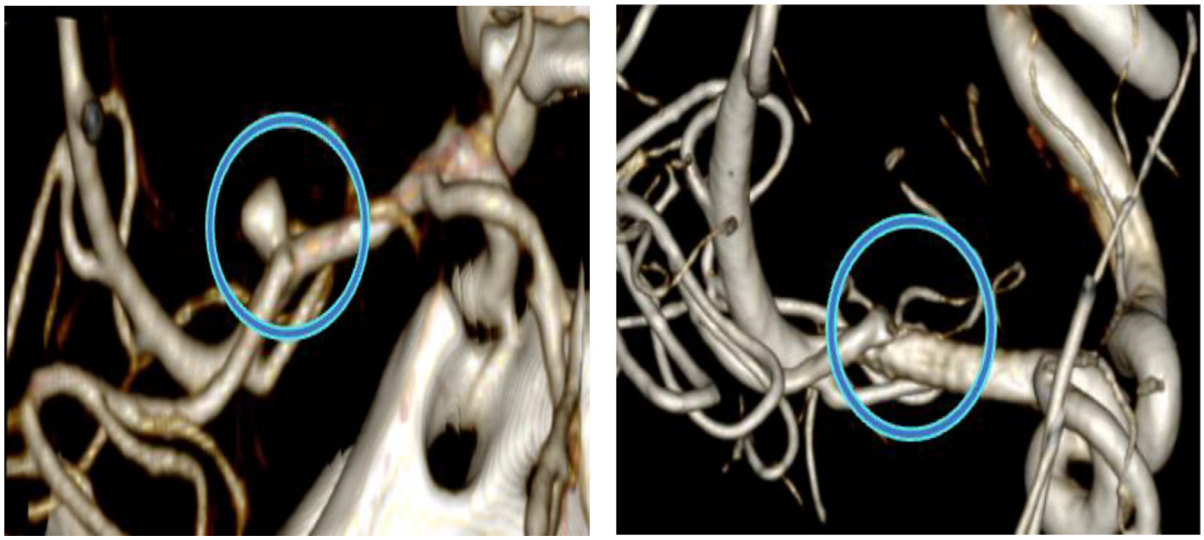

Fig. 2 – CTA results (left): saccular aneurysm at right MCA, cerebral angiography 1 month after CTA (right): no dome aneurysm visible, still visible aneurysm remnants resembling an aneurysm blister.

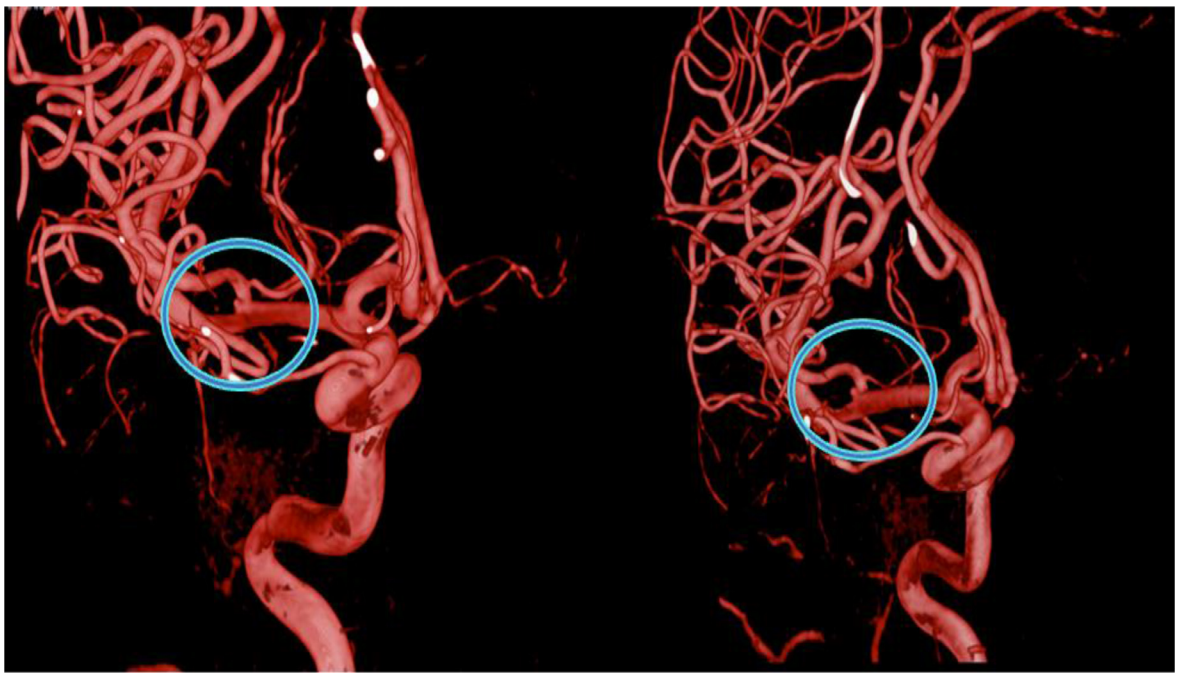

Fig. 3 – Cerebral angiography in 2020 (left) with cerebral angiography in 2021 (right) same as before, no more visible dome.

mectomy and thrombectomy in large intracavernous carotid aneurysm that was thrombosed and occluded in the carotid arteries [6]. Furthermore, Cohen reported a case series of 3 patients who had a completely thrombosed aneurysm causing infarction distal to the vascularized area of the anterior communicating artery, PICA, and PCA [5].

Spontaneous thrombosis could occur due to calcification within the atherosclerotic aneurysm wall and loss of the elastic lamina. Aneurysm size, volumeter door aneurysm, and high aneurysmal dome diameter had been reported to be associated with this condition. High spontaneous aneurysmal thrombosis was related to a narrow aneurysmal orifice and a wide dome. Compared to giant aneurysm, saccular aneurysm accompanied by spontaneous thrombosis was rarely reported [7].

The vasa vasorum fragile that surround *giant aneurysm* can bleed into the inner wall *subadventitial*, contributing to the occurrence of intramural thrombosis. This mechanism was

similar to the neovascularization of the bleeding membrane of chronic subdural hematomas [8]. Factors that precipitate spontaneous thrombosis included vasospasm, compressive effects of an adjacent hematoma, hypotension induced during anesthesia, and arrest of blood flow during angiography. Most of these factors were related to some aspect of the *trias virchow* for vascular thrombosis, including stasis, hypercoagulability, and endothelial lesions [9].

Vasospasm could cause temporary low blood flow and aneurysmal thrombosis. Furthermore, spontaneous thrombosis of a ruptured aneurysm, confirmed by angiography within 24 months of demonstrating severe vasospasm played a crucial role in aneurysmal thrombosis [9]. One of the risk factors of this condition was the use of antifibrinolytic drugs, especially tranexamic acid, which inhibited plasminogen activating factors in and around the aneurysm wall [10].

In this current case, the patient presented with a stiff neck, severe headaches and seizures. The CTA examination revealed the presence of an aneurysm, while the DSA showed spontaneous thrombosis. Upon follow-up with another DSA within 1 year, the aneurysm remained occluded, no recanalization was found, and there were no changes in neurological deficits. The occurrence of spontaneous thrombosis in the patient was likely due to vasospasm, which was associated with factors leading to *thrombogenesis*. The use of the prescribed antifibrinolytic agents also contributed to the incidence of spontaneous thrombosis. It was important to note that the occurrence of this complication does not rule out the possibility of bleeding, recanalization, or rupture. Therefore, it was recommended that patients with spontaneous thrombosis undergo a DSA evaluation at least 3 months after the onset. Another alternative that can be used to evaluate the blood vessels of the brain was Ultrasound fusion imaging system. This examination is noninvasive and can be done repeatedly with more comfort [11].

Conclusion

The occurrence of spontaneous thrombotic events in saccular aneurysm is a rare case. The patient in this report had total spontaneous thrombosis in segment M2 of Right MCA. This provided temporary and permanent benefits to the patient. However, spontaneous thrombosis requires periodic evaluation, and proper understanding of the mechanism can serve as a guide during the selection of therapeutic approach to aneurysm.

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Patient consent

Written informed consent for the publication of this case report was received from patient's family.

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