Radiology Case Reports 19 (2024) 3405–3410

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Case Report

Spontaneous thrombosis of a large unruptured intracranial aneurysm causing ischemic stroke due to occlusion of the parent artery: A case report and literature review ☆

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ARTICLE INFO

Article history:
Received 1 May 2024
Accepted 3 May 2024

Keywords:
Spontaneous thrombosis
Unruptured large or giant intracranial saccular aneurysm
Simultaneous thromboses of the aneurysm and its parent artery
Ischemic stroke

ABSTRACT

Spontaneous thrombosis of an unruptured large or giant saccular intracranial aneurysm is a well-known phenomenon and can cause ischemic stroke (IS), which is a rare event. The possible pathogenic mechanisms of IS include distal embolic occlusion secondary to migration of the intra-aneurysmal thrombus, occlusion of the parent artery lumen caused by the retrograde extension of the aneurysmal thrombosis, external compression of the parent artery due to the increased aneurysmal mass effect. Among these, IS due to simultaneous thromboses of the aneurysm and its parent artery is extremely rare, with only a few cases reported in the literature. Herein, we present a case of a 18-year-old woman who suffered an acute IS, attribute to spontaneous complete thrombosis of an unruptured large saccular aneurysm of the right middle cerebral artery with occlusion of the parent artery, and we review the literature simultaneously.

Introduction

A brain aneurysm is an abnormal bulge at one or more locations that are weak spots on the wall of a cerebral artery. Morphologically, most IAs are saccular, which leads to the name "saccular aneurysm". IA is relatively common, with the prevalence of 0.4% on autopsy, 3.6% on biopsy, 3.7% on retrospective cerebral angiography, 6% on prospective cerebral angiography, 2.3% in healthy adults and increases with age [1]. Most

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2024.05.017

IAs present without causing symptoms or complications. The most common complication is rupture into the subarachnoid space causing subarachnoid hemorrhage. Fortunately, most IAs will not rupture during their lifetime. The annual rate of IA rupture is only about 1%-2% [2].

Spontaneous thrombosis forming in the lumen of unruptured IA is a common phenomenon, in most cases associated with large (>15 mm) and giant (>25 mm) aneurysms. About 50%-60% of large and giant IAs have partial thrombosis and 13%-20% have complete thrombosis [3–6]. Hemodynamic stress on the aneurysm wall leads to damage of the endothelial layer, triggering blood clot formation, which is considered the main mechanism of this phenomenon [7–9]. Most thrombi will stabilize within the aneurysm and have no consequence, but in some cases, this is the cause of IS. The mechanism of IS may be due to the thrombus moving from the lumen of the aneurysm (the aneurysm in this case is usually a partially thrombosed aneurysm) to occlude a distant cerebral artery—called distant thromboembolism mechanism (thromboemboli mechanism). Another mechanism is due to thrombus which developes to fill the lumen of the aneurysm (the aneurysm in this case will be a completely thrombosed aneurysm), invading retrograde towards and cause occlusion of the parent artery – called local invasive thrombosis mechanism (local extension mechanism). The final mechanism is because of the increased mass effect from an IA containing thrombus causing compression of the parent artery from the outside (aneurysm in this case usually a giant aneurysm) – called mass effect mechanism. All three mechanisms are rare causes of IS. The rate of giant aneurysms causing IS by thromboemboli mechanism is 5-8%, while by local extension and mass effect mechanisms are extremely rare, with only a few reported cases in the literature and all involve large or giant aneurysms [4,5,10–14].

Notably, diagnosing a case of IS based on the mechanism of thrombosis developing to fill the lumen of an unruptured aneurysm and invading the parent artery is not simple for several reasons. First, the imaging of a completely thrombosed aneurysm is difficult to distinguish from other lesions, especially brain tumors [6,15–18] and cavernoma [5,19–21]. Even the “gold standard” for diagnosing cerebrovascular disease, digital subtraction angiography (DSA), is also difficult to distinguish and the results are usually “negative”. Second, even in cases that the IA has been identified, determining the causal relation between this type of aneurysm and IS events requires extreme caution because there may be confusion and missing of other causes.

Principles of treatment of IS due to spontaneous thrombosis of an unruptured IA include recanalization treatment, secondary IS prophylaxis and treatment of the underlying cause. Recanalization treatment follows general treatment guidelines for acute IS, i.e. intravenous thrombolysis and/or endovascular mechanical thrombectomy if indicated. Antiplatelet drugs are the foundation of secondary IS prophylaxis, especially in cases where the aneurysm is only partially thrombosed. For underlying cause treatment aiming at preventing recurrent IS, and at the same time preventing the risk of aneurysm rupture, for an unruptured IA containing thrombus, the guidelines up to now have not been established. The consensus of opinion among experts is to remove the aneurysm by surgery or endovascular intervention in cases where the aneurysm is only partially thrombosed. On the other hand, if the aneurysm is completely thrombosed, conservative treatment and monitoring are considered most of the time, with surgery or endovascular intervention only being considered if during the monitoring process, spontaneous lysis of the thrombus and recanalization of the IA are noted [8]. However, spontaneous recanalization of a completely thrombosed IA is rare, with only a few cases reported in the literature [11,22,23]. Similarly, the risk of rupture of a completely thrombosed IA is extremely rare [8,9].

Case presentation

A female patient, 18 years old, is a high school student with no previous medical history. The day before admission around 5 a.m., the patient suddenly had difficulty speaking with mouth distorted to the right and left-sided weakness. She was admitted to a local hospital and diagnosed with an IS. Her condition then continued to deteriorate and she was transferred to the Emergency Department of our hospital at nearly 5 a.m. the next day, at the 24th hour of the disease. She was admitted in the state of drownsiness with severe dysarthria and left-sided weakness. The NIHSS (National Institutes of Health Stroke Scale) score was 8 points.

Paraclinical tests in the diagnostic protocol for young onset IS were urgently performed, and a definite diagnosis was established: Acute ischemic stroke in the right putamen and internal capsule due to occlusion of the right middle cerebral artery (MCA) by an invasive thrombus from a large unruptured right MCA aneurysm (Figs. 1 and 2).

The patient was treated according to the current treatment protocol for IS at our hospital. Right MCA revascularization was not recommended because the patient was admitted to the hospital at the 24th hour after onset, having already passed the treatment window. Antiplatelet drugs for secondary prevention of IA were not prescribed. Conservative treatment and follow-up planning to check whether spontaneous recanalization happens or not at 3 months, 6 months, 1 year, 2 years, 3 years were the choice for the etiology of completely thrombosed unruptured aneurysm of right MCA in this patient.

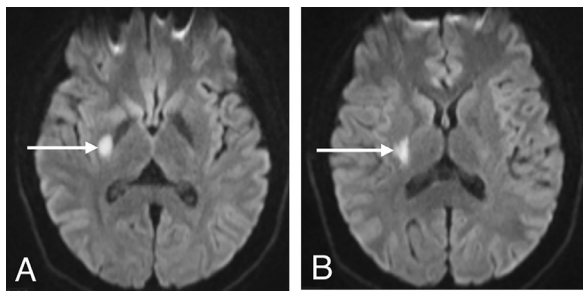

Fig. 1 – Infarction of the right putamen and internal capsule. (A and B) Hyperintensity of the right putamen and internal capsule on MRI-DWI (arrow).

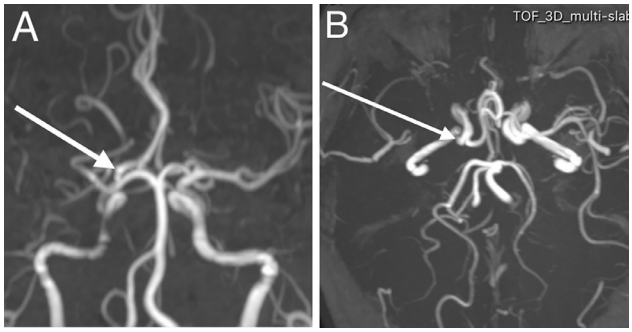

Fig. 2 – Occlusion of the right middle cerebral artery. (A and B) Loss of the right middle cerebral artery signal on 3D MRI-TOF.

After 10 days of treatment, the patient recovered well from her symptoms and was discharged from the hospital in a state of complete alertness, almost normal speech, and improved muscle strength on the left side of the body. She was able to walk independently and was advised to return for follow-up appointments.

Discussion

The diagnosis of IS in this patient was completely convincing based on clear clinical (sudden onset, focal neurological signs, symptoms lasting more than 24 hours) and paraclinical (signal of cerebral infarction in anatomical location on MRI consistent with clinical symptoms) evidences.

The IS lesion was located in the right putamen and internal capsule, so we determined that the culprit artery (clogged artery) was the MCA on the same side, more specifically the perforating artery branches.

Five groups of etiologies of IS according to the TOAST (The trial of Org 10172 in acute stroke treatment) classification [24], the most commonly used classification in the world, are large artery atherosclerosis, cardioembolism, small vessel occlusion, other determined etiology and unknown etiology. Our patient is only 18 years old, which means young onset IS, so we focused our attention on 2 groups of etiologies: cardioembolism and other determined etiology, in addition to excluding 2 common etiological groups in general: large artery atherosclerosis and small vessel occlusion. We thoroughly explored the patient's medical history as well as performed a series of tests including complete blood coagulation, echocardiography, systemic vascular ultrasound, transcranial ultrasound with air bubble test, chest X-ray, holter ECG, hypercoagulable state testing, inflammatory markers, lipid profile, especially brain imaging tests including CT/CTA, MRI/MRA and even DSA. Analyzing the collected data, the etiological groups of large atherosclerosis, small vessel occlusion, and cardioembolism were not difficult to rule out due to lack of evidence. On the contrary, a lesion suspected of having a causal relationship with IS was noticed on imaging. The imaging characteristics of this lesion were as follows: located in the right temporal fossa, size 23 × 18 mm, clear boundary, quite

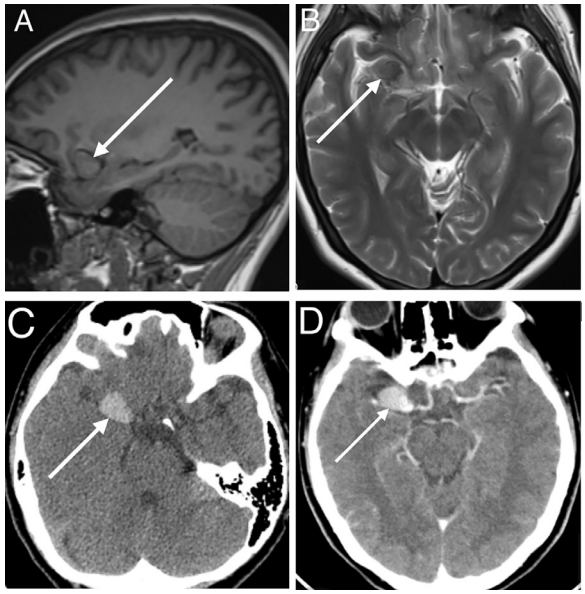

Fig. 3 – The right middle cerebral artery aneurysm contained an early subacute thrombus. (A) Slightly homogeneous hyperintensity on MRI-T1 (arrow). (B) Slightly homogeneous signal reduction on MRI-T2 (arrow). (C) Homogeneous enhancement on non-contrast CT (arrow). (D) No enhancement on contrast-enhanced CT (arrow).

homogeneous signal which is consistent with the signal of blood in the early subacute stage (hyperenhancement on CT, mild hyperenhancement on MRI-T1, mild hypoenhancement on MRI-T2), no peripheral hemosiderin rim, no contrast enhancement, no surrounding cerebral edema, no mass effect, located in the anatomical area of the right MCA (Figs. 1-3). Our clinicians and radiologists discussed and proposed a number of possibilities for lesions that could be consistent with the above imaging characteristics including brain parenchymal hemorrhage, hemorrhagic brain tumor, hemorrhagic cavernoma, large unruptured MCA aneurysm that was completely thrombosed. In-depth analysis helped us rule out brain parenchymal hemorrhage because of the clear boundary characteristics of the lesion as well as the absence of cerebral edema around the lesion. Hemorrhagic brain tumor and cavernoma were not considered because the lesion did not enhance on contrast, there was no surrounding brain edema (not consistent with a brain tumor) and no hemosiderin rim surrounding the periphery (not suitable for cavernous angioma). Finally, a large unruptured MCA aneurysm with complete thrombosis was the possibility that we thought of the most because it matched all the described imaging characteristics, especially more convincing with the lesion located on the anatomical direction of the MCA on the same side. We also concluded that there was a causal relationship between this aneurysm and the IS that occurred in the blood supply area of the MCA on the same side. More specifically, a large unruptured right MCA aneurysm which was completely spontaneously thrombosed, and then the invasive thrombus caused occlusion of the right MCA (parent artery) was the possible pathogenesis for IS in this case. We did not think that

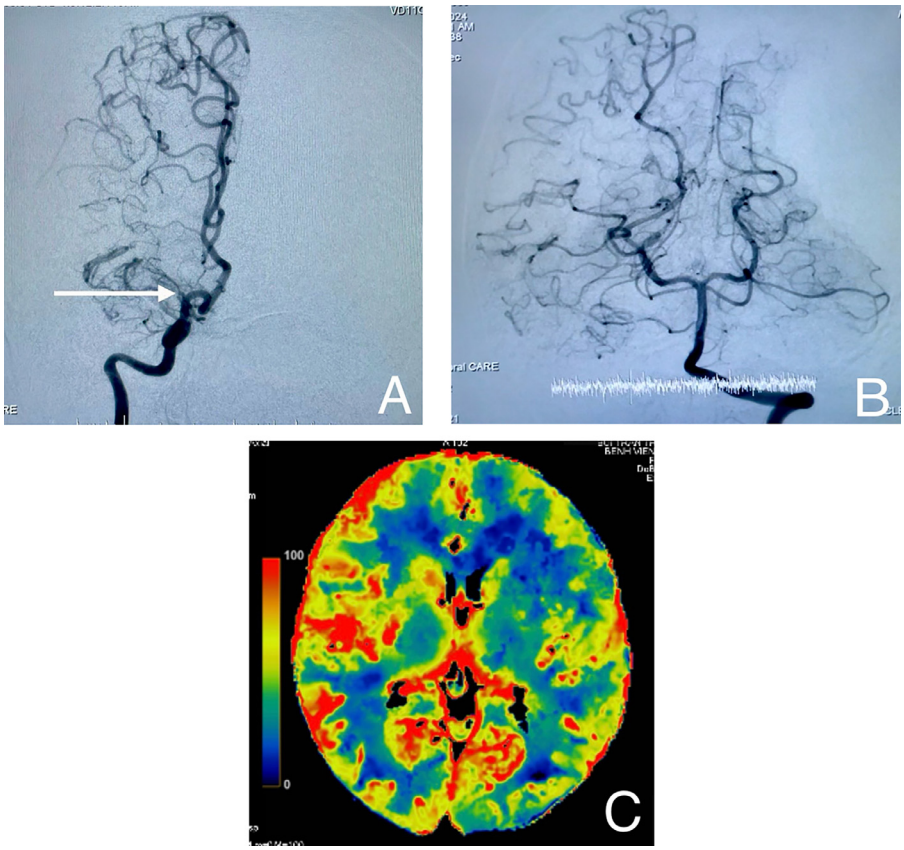

Fig. 4 – Collateral circulation and cerebral perfusion after the right middle cerebral artery occlusion. (A) The right middle cerebral artery occlusion (arrow), rich collateral circulation from the right anterior cerebral artery on DSA. (B) Rich collateral circulation from the right posterior cerebral artery on DSA. (C) Abundant cerebral perfusion after the right middle cerebral artery occlusion on CTP.

the mechanism was due to mass effect from the aneurysm causing compression of the MCA because on imaging there was no evidence of mass effect. In general, the diagnosis of determining the nature of the lesion with the described imaging characteristics was a difficult diagnosis. World medical literature even recorded cases of misdiagnosis, in which the results were only clear when the lesions were remove from patients by surgery and then underwent pathology [7,17]. This case was fortunate to have many relatively typical objective data at the same time, helping us analyze and establish a definitive diagnosis with high reliability as described.

An IA measuring 23 × 18 mm (width x height) is by definition a large aneurysm, nearly meeting the standard for giant aneurysm size, and thrombus forming naturally within is not difficult to explain [3,4,8,9,15]. According to the medical literature, the thrombus developing to fill the lumen of the IA, invading and occluding the parent artery, leading to IS is very rare. The clinical case we shared is one of them [4,5,13].

The patient had an occlusion of the root of the MCA, but the infarction core was limited to the putamen and the internal capsule which belonged to the blood supply area of the perforating artery branches. The reason was because of the abundant collateral circulation behind the occlusion site from the anterior cerebral artery and posterior cerebral artery with clear evidence on DSA as well as CTP (computed tomography perfusion) (Fig. 4). This was an important factor that helped the patient not have to endure a severe stroke which led to her well recovery.

Treatment of occlusion of the right MCA with intravenous thrombolytic drug as well as mechanical endovascular therapy was not recommended by us because the patient was hospitalized at the 24th hour of onset, beyond treatment window. The goal of preserving collateral circulation to supply blood to the brain parenchyma is achieved by medical treatment, with a focus on maintaining optimal blood pressure and circulatory volume level. For the goal of preventing recurrent IS, because the aneurysm was completely thrombosed which led to complete occlusion of the parent artery, we decided not to use antiplatelet drugs due to the assessment that the risk of new thrombi continuing to form and continuing to cause local as well as distant thromboembolism was absent. For the treatment of the large unruptured MCA aneurysm that was completely thrombosed, based on a highly consensus view in the medical literature, we chose conservative treatment with follow-up of checking for spontaneous recanalization. There was no indication for surgical treatment or endovascular intervention for the MCA aneurysm because the risk of spon-

taneous recanalization and rupture of this type of aneurysm is extremely rare [8,9,11].

According to our research, medical literature in our country has not recorded any reports of IS due to spontaneous thrombosis causing simultaneous occlusion of an unruptured IA and its parent artery. Our patient is the first case reported in Vietnam.

Conclusion

Thrombus forming spontaneously within a large or giant unruptured IA is not uncommon. Diagnosing a large or giant unruptured IA that has been completely thrombosed by imaging is not simple and can easily be confused with other type of lesions, especially brain tumor and cavernoma. The thrombus develops to fill the aneurysm lumen, invades the area, causes occlusion of the parent artery, leading to IS. This is a very rare condition, with only a few cases reported in the world. There have been no reports in the country and this is the first clinical case report in Vietnam according to our research. Conservative treatment and follow-up planning is the choice of treatment that has received high consensus for unruptured IA with complete thrombosis. Surgery or mechanical endovascular intervention for this type of IA is only recommended when there is spontaneous recanalization, which is extremely rare.

Author's contributions

Ta VK, Truong MT, and Nguyen MD: Case file retrieval and case summary preparation. Ta VK, Truong MT, and Nguyen MD: preparation of manuscript and editing. All authors read and approved the final manuscript.

Availability of data and materials

Data and materials used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

Our institution does not require ethical approval for reporting individual cases or case series. Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

Patient consent

Informed consent for patient information to be published in this article was obtained.

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