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Case Report

A difficult-to-treat Acom aneurysm with the combined vascular anomaly of Acom fenestration and accessory anterior cerebral artery

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Abstract

Background: Vascular anomaly and aneurysmal formation of an anterior communicating artery (Acom) complex has often been reported. Because of such a complicated relationship between the vascular structure and aneurysms, Acom aneurysm is one of the most difficult aneurysms to treat among other common anterior circulation aneurysms. We experienced an extremely rare and difficult-to-treat case of ruptured Acom aneurysm with the combined vascular anomaly of the Acom fenestration and an accessory anterior cerebral artery (ACA).

Case Description: A 29-year-old man underwent a clipping surgery for a ruptured Acom aneurysm with an Acom fenestration and an accessory ACA. By reasons of the complicated Acom structure and a posteriorly-projecting aneurysm, the patient was operated via an interhemispheric approach, which is generally reported to provide the best operative view for all types of Acom aneurysms. However, we could not help applying a clip in the narrow working space and the limited operative view, due to the poor mobilization of Acom complex and the interruption by an accessory ACA.

Conclusion: The interhemispheric approach may exceptionally have a blind area behind the Acom complex in the case of Acom aneurysms with an accessory ACA. Additionally, the aneurysms arising from the Acom fenestration strongly require neurosurgeons to take a more accurate surgical approach to obtain a direct visualization for an aneurysmal neck.

Key Words: Acom fenestration, accessory anterior cerebral artery, interhemispheric approach, pterional approach, subarachnoid hemorrhage

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INTRODUCTION

Vascular anomaly of the anterior communicating artery (Acom) complex and anterior cerebral artery (ACA) relates closely with the cerebral aneurysm formation. [3,5,11,13,15,17,18] We experienced a rare and difficult-to-treat case of ruptured Acom aneurysm with the combined vascular anomaly of the Acom fenestration and an accessory ACA. As far as we reviewed the literature, Acom aneurysm

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with these combination vascular anomaly was the first case. The interhemispheric approach, which is generally reported to provide the best operative view for all types of Acom aneurysms, [6,9,15] was exceptionally inappropriate approach for a neck clipping in the present case. Either Acom fenestration or accessory ACA was reported to be often observed in ruptured Acom aneurysms, [11,15,17] and therefore, neurosurgeons should know the surgical pitfall in the treatment of Acom aneurysm with such vascular anomaly. Herein, we describe the pitfall obtained from this case.

CASE DESCRIPTION

A 29-year-old man was taken to our hospital by ambulance, suffering from severe headache. He had no past medical history and did not take any medication. Head computed tomography (CT) revealed an extensive subarachnoid hemorrhage (SAH) filling basal cisterns, especially the thick hemorrhage in the interhemispheric fissure [Figure 1a]. CT angiography showed an Acom with fenestration, but no evidence of aneurysm [Figure 1b]. Three-dimensional-digital angiography (3D-DSA) subtraction demonstrated the fenestration of Acom with a small saccular aneurysm (2 mm) [Figure 2a-d]. The aneurysm arose from the inferior limb of the fenestrated segment, and it projected posteriorly [Figure 2b-d]. In addition, an accessory ACA arose from the upper limb of the

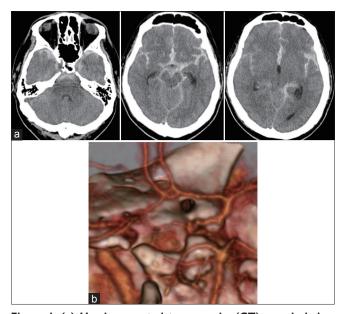


Figure 1: (a) Head computed tomography (CT) on admission. Diffuse subarachnoid hemorrhage in the basal cistern. In particular, a thick subarachnoid hemorrhage in the proximal portion of interhemispheric fissure. (b) Three-dimensional-CT Angiography (3D-CTA) on admission. 3D-CTA showed the anterior communicating artery (Acom) fenestration, but no evidence of aneurysm. The quality of this study was poor, probably due to high intracranial pressure

fenestrated segment [Figure 2c and d]. This patient underwent a clipping surgery via an interhemispheric approach. At the operation, we had difficulty in getting a good surgical view for an aneurysm existing behind the Acom, because we could not mobilize the Acom complex enough. Additionally, the surgical dissection behind the Acom complex was interfered by an accessory ACA [Figure 2e]. The intraoperative premature rupture occurred, and a fenestrated clip was applied to be able to preserve an accessory ACA [Figure 2f]. Immediately after the clip application, the bleeding from the ruptured aneurysm stopped. The remnant of aneurysm was not identified in the visible range, and then we finished off the operation. Postoperatively, we performed the management of brain swelling and cerebral vasospasm. Unfortunately, the re-rupture from an Acom aneurysm occurred on Day 13 [Figure 3a-c]. 3D-DSA demonstrated the recurrence of an aneurysm next to the clip [Figure 3d]. Cerebral vasospasm became progressively worse under the influence of re-rupture, and the patient died on the same day.

DISCUSSION

Vascular anomalies of the circle of Willis frequently associated with the aneurysm, and several anomalies of the Acom complex and ACA have been described.[3,5,11,13,15,17,18] The relationship fenestration and aneurysm formation has ever been reported.[3,5,11,15,18] It has been speculated that defects of the tunica media at the proximal and distal ends of the fenestration arise turbulent flow, which results in the aneurysm formation.^[5] Increased hemodynamic stress at these gaps of the media is thought to lead to the increased prevalence of aneurysms among patients with fenestration. [5] The fenestration of Acom is reported to be present in 12-21% of the population. [5,11,15,17] Ân accessory ACA is defined as the occurrence of three A2 segments. The reported prevalence of an accessory ACA arising from the Acom ranges from 2% to 13%. [3,5,13,14] This normal variant most likely represents persistence of the median callosal artery. The aneurysmal formation of an accessory ACA is quite rare, and only a few cases have been reported previously. [7,10,12] According to these case reports, aneurysms related to an accessory ACA have a tendency to arise at the distal portion of ACA, not Acom. [7,10,12] In contrast, in the present case, the aneurysm did not originate from the upper limb of fenestrated segment, where an accessory ACA arose. Thus, the aneurysmal formation in the present case is speculated to be mainly due to the fenestration.

The pterional approach and interhemispheric approach are standard surgical procedures for clipping Acom aneurysms. [1,2,4,6,16] The advantage of interhemispheric approach is that it provides better visualization and

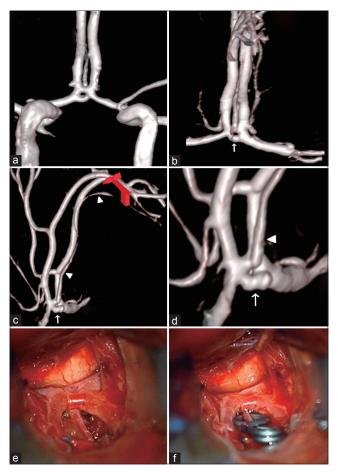


Figure 2: (a-d) Three-dimensional-digital subtraction angiography (3D-DSA) on admission. (a) water's view, (b) A-P view, (c) L-R view, (d) magnification picture of Figure 2c. 3D-DSA revealed the vascular anomaly [the anterior communicating artery (Acom) fenestration and an accessory anterior cerebral artery] and small Acom aneurysm projecting posteriorly. An accessory anterior cerebral artery (arrow head) arose from the upper limb of the fenestrated segment. In contrast, the Acom aneurysm (arrow) arose from the inferior limb of the fenestrated segment. (e-f) Intraoperative photographs. The surgical view obtained from the interhemispheric approach showed a blind area behind the Acom complex with limited mobilization. A fenestrated clip was applied to be able to preserve an accessory anterior cerebral artery

better understanding of the vascular structures near the Acom complex. [6,15] An interhemispheric approach is thought to be suitable, especially for Acom aneurysms projecting posteriorly or Acom aneurysms with vascular anomaly. In the present case, both these factors representing good indication of the interhemispheric approach were observed. However, there were two reasons why we had the difficulty in getting the good visualization of Acom aneurysm in this case. First, the accessory ACA directly prevented us from getting the good view in the back of Acom complex. Second, Acom complex was very firmly fixed by more vessels than usual. Compared to normal, Acom complex consisted of two Al, three A2, and fenestrated Acom with the double vascular diameter in this case. As a result, we could not get the mobilization of Acom complex, and obtain the restricted view in the back of Acom complex. On the other hand, some neurosurgeons take a pterional approach for clipping of Acom aneurysms with posterior projection. [6,15,16] In such case, the aneurysms are usually

approached from the side where the proximal A2 portion is located anteriorly (i.e. the side where the A2 fork is close). Sano et al. warned that neurosugeons should consider the appropriate surgical approach for Acom aneurysms with Acom fenestration, because the wrong approach may make the neck clipping very difficult.[16] Akashi et al. also reported that they could not apply a clip for a posteriorly-projecting aneurysm of fenestrated Acom via a pterional approach from the side where the A2 fork was open.[8] They concluded that one of the major reasons for their failure was the poor mobilization of fenestrated Acom, and Acom aneurysms with fenestration should be approached to be able to obtain the direct operative view to the aneurysmal neck. Thus, in the present case, the only surgical procedure to clip an aneurysm was thought to be a pterional approach from the side where the A2 fork was close. On the other hand, the endovascular treatment should be considered as an alternative therapeutic option. However, even the endovascular coiling was thought to be technically

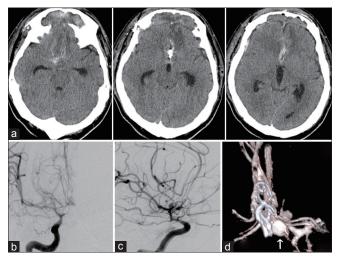


Figure 3: (a) Head computed tomography (CT) on Day 13. The focal subarachnoid hemorrhage was observed around the aneurysmal clip, suggesting the re-rupture from the remnant of anterior communicating artery (Acom) aneurysm. (b-c) Two-dimensional-digital subtraction angiography (2D-DSA) after re-rupture. b: P-A view, c: R-L view. 2D-DSA showed the recurrence of Acom aneurysm. (d) Three-dimensional-digital subtraction angiography (3D-DSA) after re-rupture, posterior view with caudal angulation. The recurrence of an aneurysm (arrow) was observed next to the clip

difficult in the present case, because the aneurysm size was too small (2 mm).

CONCLUSION

Vascular anomaly associated with Acom complex is often observed. The aneurysms of fenestrated Acom should be approached to be able to obtain a direct visualization for an aneurysmal neck, because the Acom complex may be fixed and poorly mobilized. In addition, an interhemispheric approach may exceptionally have a blind area behind the Acom complex in the case of Acom aneurysms with an accessory ACA, although an interhemispheric approach is generally reported to provide the best operative view for all types of Acom aneurysms.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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Commentary

The authors courageously report the persistence (not recurrence) of an aneurysm of a complex anterior communicating complex following an inter hemispheric approach.

Their chosen operative approach, one of a number of described approaches, was particularly ineffective in this complex configuration.

I would point out that proximal control, enabling a safer and more thorough evaluation of complex configurations, is also a surgical goal. For this reason, a pterional approach from the dominant ACA side, is a preferred approach in this surgeon's experience.

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