

Aneurysm

A saccular-like dissecting aneurysm of the anterior cerebral artery that developed 2 years after an ischemic event

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Abstract

Background: A rare case of a growing dissecting aneurysm, which was located at the horizontal (A1) segment of the anterior cerebral artery (ACA), is reported.

Case Description: A 53-year-old woman experienced left hemiparesis and alien hand syndrome. A computerized tomography scan showed an infarction in the right frontal lobe, and cerebral angiography revealed a false lumen and intimal flap at the A1 segment of the ACA. Magnetic resonance angiography demonstrated that the stenosis progressed 6 months later and improved 1 year later. Cerebral angiography showed a saccular-like aneurysm 2 years later. The surgery was planned for prevention of aneurysmal rupture. The aneurysm, which was cocoon shaped, was exposed surgically and was resected. Histological examination of the aneurysm showed arterial dissection. The postoperative course was uneventful without additional neurological deficits.

Conclusion: This is the first case report of A1 dissecting aneurysm presenting with an ischemic event in the literature. The sequential change of the configuration was curious to develop aneurysmal dilatation in 2 years. Long-term follow-up is necessary even after disappearance of the arterial dissection.

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Keywords:

Anterior cerebral artery; Cerebral infarction; Dissection; Regrowth

1. Introduction

There are several reports of dissections of the ACA. Of ACA dissections, those of horizontal segment (A1) are likely to induce hemorrhagic events, and those of postcommunicating segment (A2) induce ischemia [3,6,9,12,15]. We report a rare case of dissection at A1 segment of the ACA presenting with ischemia, which developed a cocoon-shaped aneurysm later.

2. Case report

A 53-year-old woman suffered from left hemiparesis and “alien hand syndrome¹.” A computerized tomography scan showed an infarction in the right frontal lobe. Cerebral angiography revealed a false lumen and intimal flap at the A1 segment of the right ACA and an occluded A2 (Fig. 1). There was no other vascular lesion determined by a 4-vessel study; the flow at the right A2 via the anterior communicating artery was not visualized. Anticoagulant therapy was performed and her symptoms improved. Magnetic resonance angiography showed that the stenosis progressed

Abbreviations: A1, Horizontal segment; A2, Postcommunicating segment; ACA, Anterior cerebral artery; MRA, Magnetic resonance angiography; ON, Optic nerve.

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¹ The alien hand syndrome is a condition characterized by unwilling, uncontrollable, but seemingly purposeful movements of an upper limb joined to a strange or foreign feeling of one's hand, such as presenting with nonpurposeful behavior such as lifting the arm or writhing fingers. It was initially used to describe interhemispheric disconnection phenomena in patients with lesions in the anterior corpus callosum [1,2].

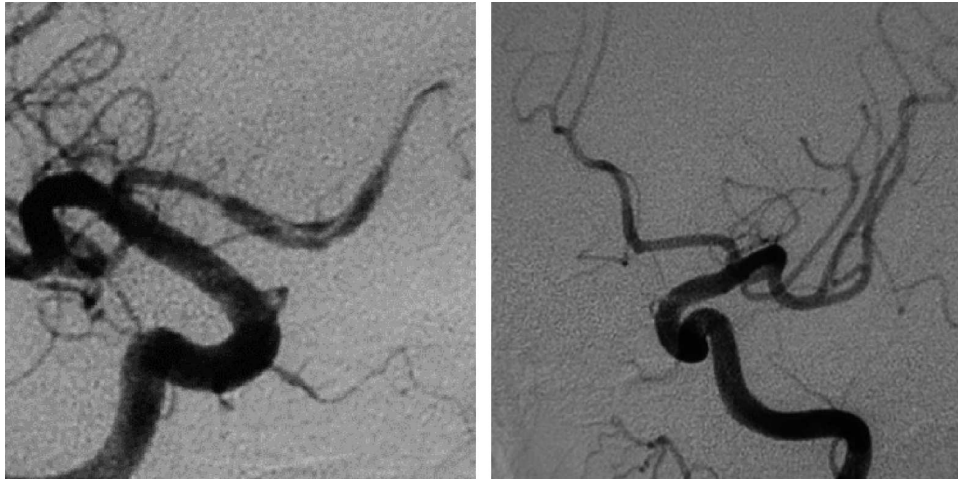


Fig. 1. An initial angiogram of the bilateral internal carotid artery showing a false lumen and intimal flap at the horizontal (A1) segment of the right ACA and occluding the postcommunicating (A2) segment of the right ACA (left). There is no flow of the right A2 via the anterior communicating artery (right).

6 months later, the stenosis resolving at 1 year, and the dissection growing an aneurysmal shape in 2 years (Fig. 2). Carotid angiography demonstrated a saccular-like aneurysm, and the right A2 was well visualized (Fig. 3). We performed surgery for prevention of aneurysmal rupture and for discrimination between a saccular aneurysm and a dissecting aneurysm. The pterional approach was made through the right frontotemporal craniotomy with orbitozygomatic osteotomy. The aneurysm, which was white and dilated in A1's itself, looked like a cocoon. There was no branch originating from the aneurysmal dome. The anterior communicating artery was not hypoplastic, and the aneurysm was resected after trapping the proximal and distal site of the aneurysm (Fig. 4). The adequate flow at the right A2 was confirmed by using a Doppler flowmeter. Histological examination of the resected aneurysmal wall showed that the vessel wall was hypertrophic, lacked the internal elastic lamina, and had no inflammation (Fig. 5). The postoperative

course was uneventful and the patient returned to the previous occupation without additional neurological deficits.

3. Discussion

Dissecting aneurysms have been related to arteriosclerosis, cervical manipulation, cystic necrosis of the media, fibromuscular dysplasia, hypertension, Marfan's syndrome, renal polycystic disease, syphilis, and head trauma. Most intracranial arterial dissections involve the posterior circulation, predominantly the distal vertebral and basilar arteries [4]. In the anterior circulation, arterial dissections have been known to involve the internal carotid artery and the middle cerebral artery [8,16], whereas dissections of the ACA had been considered rare.

In reviewing the literature of 62 ACA dissections [3,5,6,9,10,12,14,15], ischemic events were twice as many as hemorrhagic events for an initial stroke. Regarding the

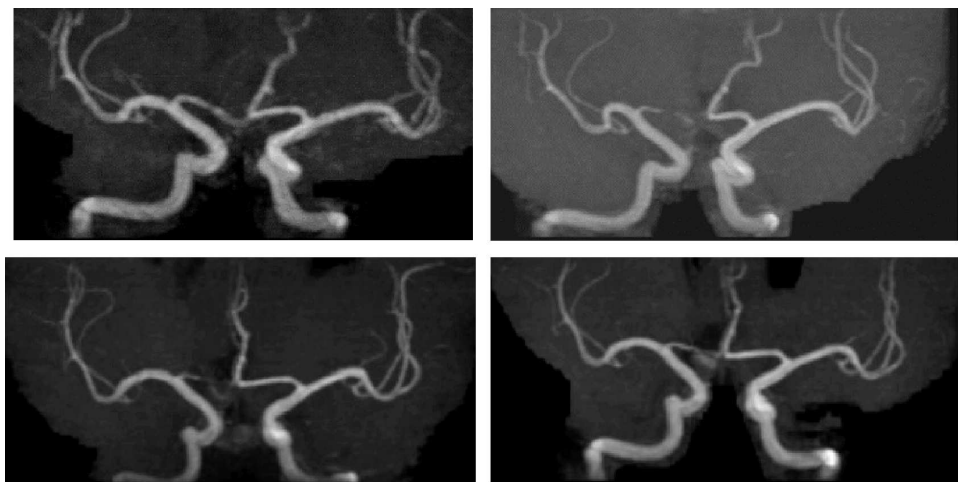


Fig. 2. Magnetic resonance angiography on admission revealed a focal stenosis at midportion of A1 (upper left). The stenosis developed at 6 months (upper right) and improved at 1 year (lower left). However, the dissection had changed to an aneurysmal shape in 2 years (lower right).



Fig. 3. Right internal carotid artery angiography 2 years after initial symptom showing a saccular aneurysm at the A1 segment of the right ACA and a normal flow of the right A2.

location of the ACA dissection, all of 11 patients with A1 dissection suffered from subarachnoid hemorrhage, and most patients needed surgery [9,12]. On the other hand, most patients with A2 dissection suffered from ischemic event, and they were treated with medical therapy [3,6,9,12,15]. A1 dissection involving the cerebral infarction, such as the present case, appears to be extremely rare.

Ohkuma et al [12] reported that the stenosis often progressed on follow-up angiograms within 1 month after onset, and the stenosis improved in all 10 cases after 2 months in A2 dissection presenting with ischemia. These results indicate the dynamic nature of the pathological condition, with increased narrowing due to the intramural hematoma and the formation of an aneurysmal bulge in the acute to subacute stage and then the healing process in the

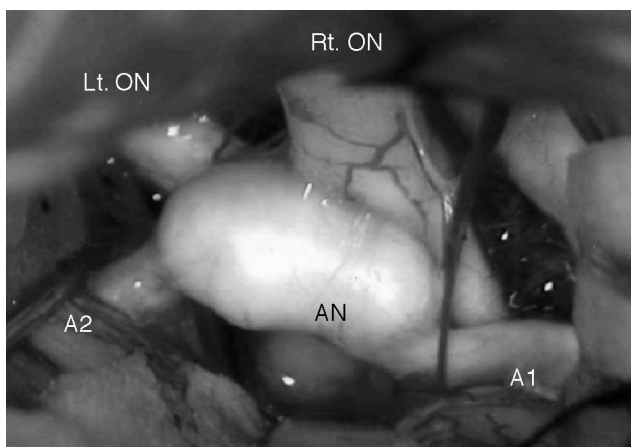


Fig. 4. Photographs of the operative field via the right pterional approach. The aneurysm located at the middle portion of the right A1 is white and looks just like a cocoon. A1 and A2 indicate A1 and A2 segment of the ACA; optic nerve, ON; AN, aneurysm.

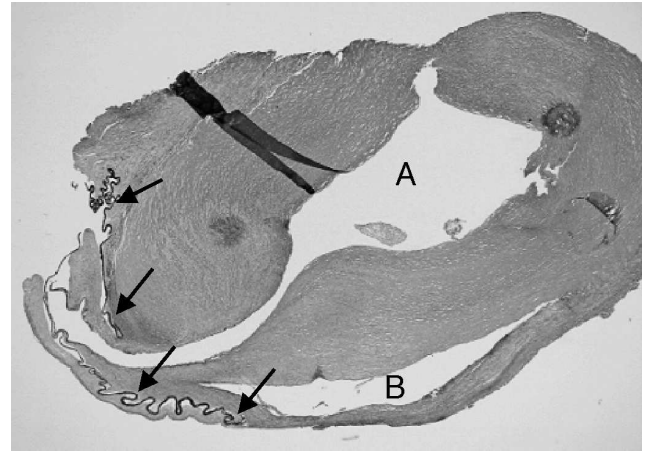


Fig. 5. Photomicrographs obtained at surgery. Cross section of the resected aneurysmal wall showing the hypertrophy of the vessel wall. Arrows indicate the disrupted internal elastic lamina (Elastica van Gieson stain, original magnification $\times 40$). A indicates the lumen of the aneurysm; B, artificial space.

chronic stage [13]. In the presented case, MRA demonstrated progression of the stenosis at 6 months, resolving of the stenosis at 1 year, and growing aneurysm at 2 years; the course is unusual. In the vertebrobasilar artery for the most part of the dissection, the natural history of the intracranial vertebral artery dissection presenting with ischemia seems relatively benign. The probability of spontaneous angiographic cure, namely, resolution of the stenosis or progression to complete occlusion, is high; however, the incidence of the persistent aneurysmal dilatation is 9% to 20% [7,11,17]. As to the other sites, there were no reports in which ACA dissection presenting with ischemia changed in the aneurysmal dilatation on the follow-up examinations as presented in the present case. The initial angiography showed an intimal flap, and the angiography 2 years from onset revealed a saccular-like aneurysm. A persistent arterial dissection usually forms in a fusiform shape, and it is extremely rare to present a saccular-shaped aneurysm as in the present case. Serial angiography and/or MRA are necessary because of the possibility of dynamic change of dissecting aneurysms.

A number of the A1 dissecting aneurysms were reported to cause subarachnoid hemorrhage. This is the first case report of A1 dissecting aneurysm presented with an ischemic event. When double lumen or intimal flap in the A1 dissection is confirmed at the initial angiography, we should consider the possibility of growing aneurysm and the subsequent bleeding. It is necessary for A1 dissection to observe the sequential change with neuroimaging in long period even after disappearance of dissection.

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Commentary

The authors present an interesting case of a dissecting aneurysm, which developed to become a saccular-like shape over 2 years follow-up. The importance is not that this is the first reported case of A1 dissecting aneurysm, but rather the unusual development of aneurysmal shape is a warning to neurosurgeons that follow-up radiological studies are important.

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This is an interesting case report of a 53-year-old woman who presented with a dissection of the right A1 segment and an ischemic deficit in the appropriate distribution. The dissection was documented by angiogram and MRA. Two years after the event, an aneurysm developed in this segment, which had recanalized. It was repaired surgically. This case report is helpful in the sense that it describes a dissection and aneurysmal degeneration at a rare site, namely, the A1 segment. It also emphasizes the need for follow-up angiography in this situation.

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