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Case Report

Ruptured peripheral aneurysms in a collateral pathway associated with stenosis of a major cerebral artery: Report of two cases

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Abstract

Background: While hemodynamic stress can result in aneurysm formation, it rarely contributes to the development of peripheral aneurysms in collateral pathways. We report two patients with ruptured distal aneurysms in a collateral pathway associated with stenosis of a major cerebral artery.

Case Description: A 67-year-old man presented with intracerebral hemorrhage in the right frontal lobe. Digital subtraction angiography (DSA) revealed severe stenosis of the right middle cerebral artery and two aneurysms in the collateral pathway of the right anterior cerebral artery. The ruptured aneurysm was trapped and resected; histologically, it was a true saccular aneurysm. The unruptured aneurysm was clipped and the patient was discharged without additional neurological deficits. The second patient was a 73-year-old woman with subarachnoid hemorrhage. DSA revealed three arterial dilations. On the 7th day of hospitalization, one of the aneurysms in a posterior inferior cerebellar artery–anterior inferior cerebellar artery anastomosis that functioned as a collateral pathway in the presence of severe basilar artery stenosis was found to be enlarged. It was treated by selective aneurysmal coil embolization with parent artery preservation. Her postoperative course was uneventful and she was discharged without any neurological deficits.

Conclusion: We document the successful treatment of two patients with ruptured aneurysms in the peripheral portion of a collateral pathway. We discuss the histology of peripheral aneurysms and present a review of the literature.

Key Words: Coil embolization, collateral pathway, histology, peripheral aneurysm, posterior inferior cerebellar artery–anterior inferior cerebellar artery anastomosis, true aneurysm

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INTRODUCTION

Cerebral aneurysms tend to arise around the circle of Willis; aneurysms in the peripheral circulation are rare.[6,21] The differential diagnosis of peripheral aneurysms includes mycotic-, traumatic-, and dissecting

aneurysms.[2,5,9,24] In rare cases, hemodynamic stress is involved in the development of aneurysms, and peripheral aneurysms attributable to hemodynamic stress have been reported to be associated with arteriovenous malformation (AVM).[16,19] A few are associated with anatomical variations,[3,7,13] Moyamoya disease,[26] major cerebral artery stenosis or occlusion,[17,20,26] parental artery occlusion after surgery,[4,22] and superior temporal artery–middle cerebral artery (MCA) bypass.[14,25]

We encountered two patients with ruptured aneurysms located in the peripheral portion of collateral pathways, associated with stenosis of a major cerebral artery. One emerged in the distal right anterior cerebral artery (ACA) that functioned as a collateral pathway due to severe right MCA stenosis and the other involved a posterior inferior cerebellar artery (PICA)–anterior inferior cerebellar artery (AICA) anastomosis that acted as a collateral pathway due to severe basilar artery (BA) stenosis.

CASE REPORTS

Case 1

A 67-year-old man presented with sudden-onset headache, dysarthria, and slight left hemiparesis. Computed tomography (CT) performed at admission revealed intracerebral hemorrhage in the right frontal lobe [Figure 1a]. Digital subtraction angiography (DSA) disclosed severe stenosis in the M1 portion of the right MCA and two aneurysms in the posterior internal frontal artery and the paracentral artery of the ipsilateral ACA [Figure 1b and c]. Although an echocardiogram and blood test did not indicate infective endocarditis, due to their peripheral locations, we suspected the aneurysms to be mycotic pseudo-aneurysms and we started conservative antibiotic treatment. Follow-up DSA on day 5 demonstrated an enlarged aneurysm in the posterior internal frontal artery; based on the distribution of a hematoma, we thought it to have ruptured. The aneurysm in the paracentral artery was not enlarged [Figure 2a].

On day 7, he underwent right frontal craniotomy under intraoperative DSA. After evacuation of the intracerebral hematoma, we found that the ruptured aneurysm in the posterior internal frontal artery did not involve any arterial branches and so it was surgically trapped and resected [Figure 3a]. The neck of the other unruptured aneurysm in the paracentral artery was clipped [Figure 3b]. Intraoperative DSA confirmed the obliteration of both aneurysms [Figure 2b]. Postoperative CT demonstrated no newly developed low-density areas due to the trapping procedure and he was discharged on foot with improved left hemiparesis.

Histological study of specimens from the resected aneurysm yielded no findings of infection, bacteria, or inflammation. Portions of the aneurysmal wall demonstrated three layers (tunica intima, -media, and -adventitia) and there were no elastic fibers at the point of rupture. These findings were indicative of a true ruptured saccular aneurysm [Figure 3c and d].

Case 2

A 73-year-old woman had undergone pancreatoduodenectomy for cholangiocarcinoma 5 months earlier. She suffered sudden-onset severe headache. CT obtained at admission revealed subarachnoid hemorrhage (SAH) mainly distributed in the posterior fossa [Figure 4a]. DSA demonstrated three arterial dilations: one was located just distal to an area of severe BA stenosis and the other two were in a right PICA–AICA anastomosis that functioned as a collateral

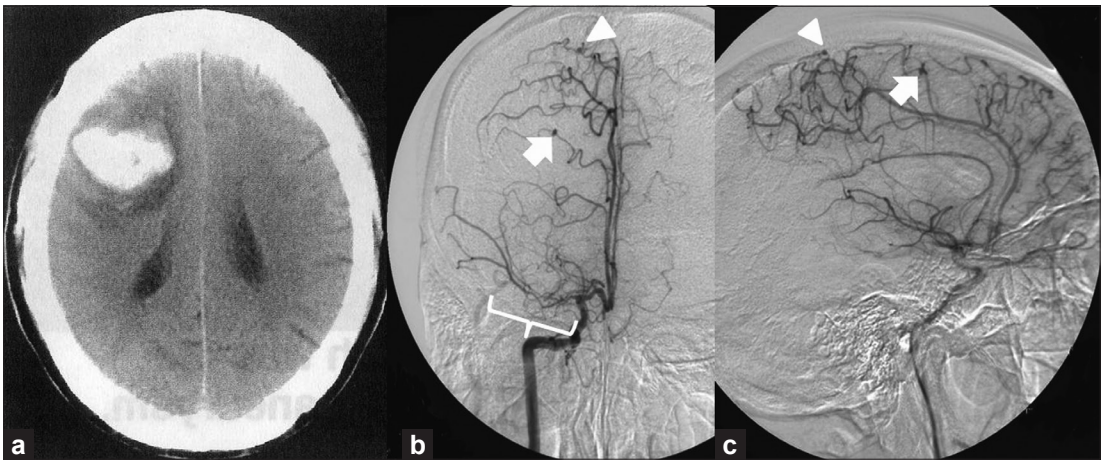

Figure 1: Case 1: neuroimaging findings on admission. (a) Axial computed tomograph (CT) showing an intracerebral hemorrhage in the right frontal lobe. (b, c) Digital subtraction angiograph (DSA) showing severe right middle cerebral artery stenosis (parenthesis) and two aneurysms in the posterior internal frontal artery and paracentral artery of the ACA (thick arrow and arrowhead, respectively) that functioned as collateral pathways

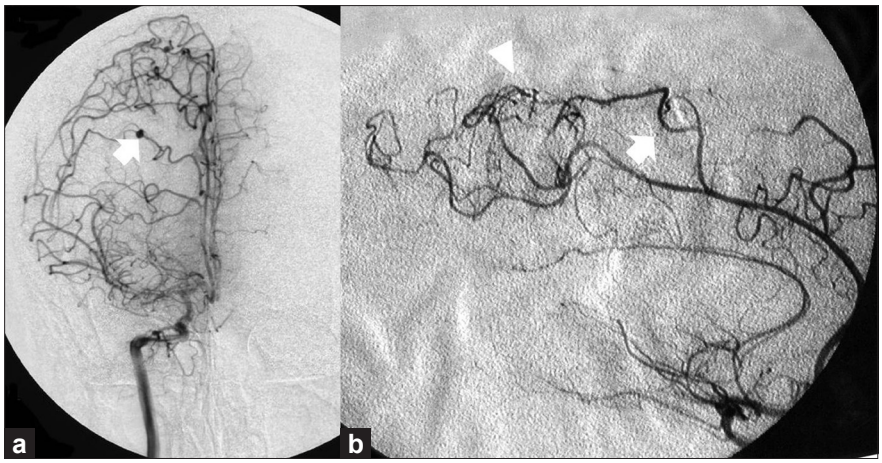

Figure 2: Case 1. (a) Follow-up DSA obtained on day 5. The aneurysm in the posterior internal frontal artery of the ACA showed enlargement (thick arrow). (b) Intraoperative DSA showing obliteration of the aneurysms in the posterior internal frontal artery and paracentral artery (thick arrow and arrowhead, respectively)

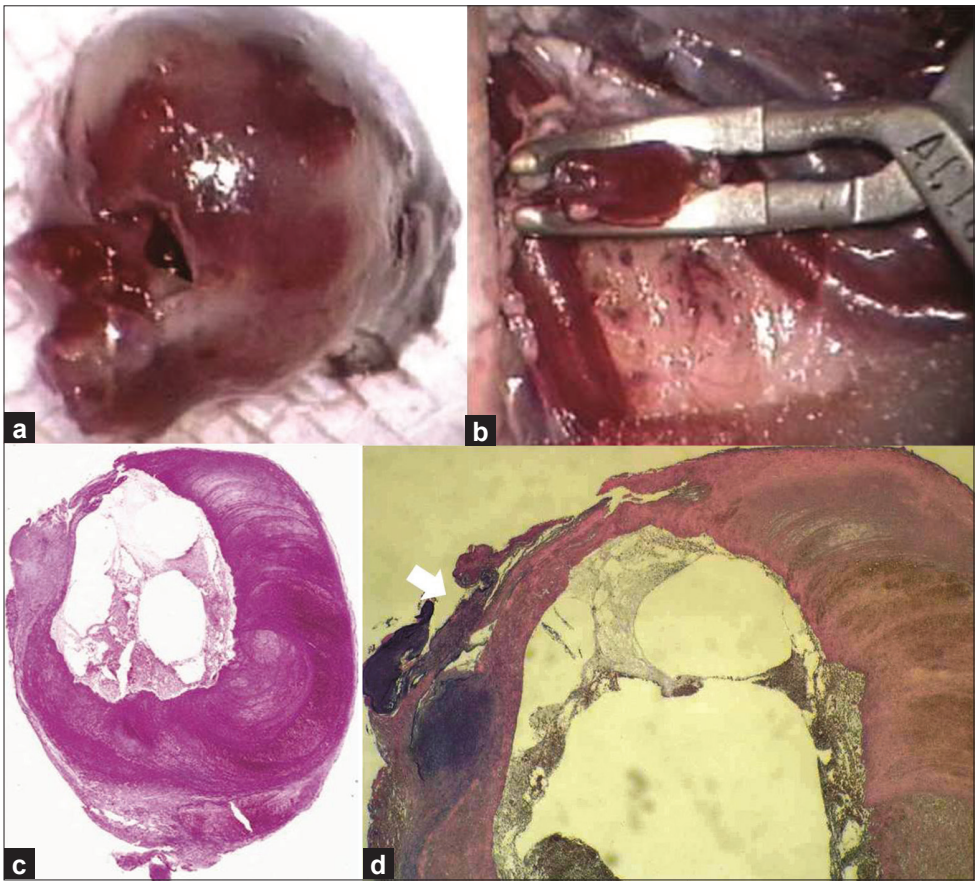

Figure 3: Case 1. (a) Photograph of the trapped and resected aneurysm in the posterior internal frontal artery. (b) The clipped aneurysm in the paracentral artery. (c) Hematoxylin and eosin staining of the trapped and resected aneurysm. Parts of the aneurysmal wall consisted of three layers. (d) Weigert staining of the resected aneurysm. Elastic fibers were stained blue/purple; the point of rupture was devoid of elastic fibers (thick arrow)

pathway due to the BA stenosis. None of the aneurysms manifested branch involvement [Figure 4b].

We initially delivered conservative therapy because we were unable to identify the lesion responsible for the SAH. Follow-up DSA performed on day 7 demonstrated that the arterial dilation in the PICA–AICA anastomosis had progressed to a saccular aneurysm with a 2-mm

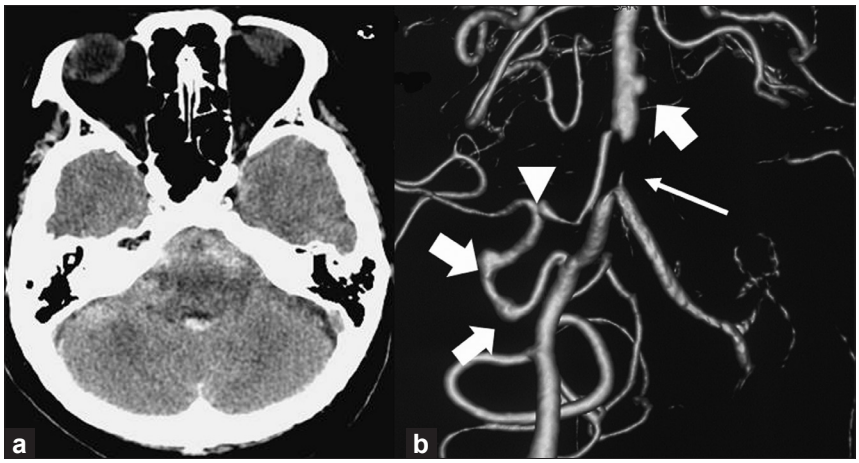

Figure 4: Case 2. (a) CT performed at the time of admission showed diffuse subarachnoid hemorrhage and hemorrhages in the fourth ventricle. (b) 3D-DSA also acquired at admission revealed severe basilar artery (BA) stenosis (long arrow) just distal to the vertebral artery union.There were three arterial dilations (thick arrows): two were in a posterior inferior cerebellar artery (PICA)–anterior inferior cerebellar artery (AICA) anastomosis (arrowhead) and one was in the BA trunk distal to a stenotic site

neck size; it was 2.7 mm wide and of 2.9 mm in height [Figure 5a]. Aneurysmal trapping carried a high risk for ischemic complications because the PICA–AICA anastomosis constituted an important collateral pathway in this patient. Considering her general condition, we chose less invasive coil embolization of the enlarged aneurysm, and on day 15, she underwent selective aneurysmal coil embolization with parent artery preservation under local anesthesia. At this point, the target aneurysm had further increased in size [Figure 5b]. Post-treatment DSA showed a small aneurysmal neck remnant; blood flow in the PICA–AICA anastomosis was preserved [Figure 5c and d]. The remaining two aneurysms showed no change in size on three subsequent DSA studies and were left untreated and followed up radiographically. Her postoperative course was uneventful and she returned to the referring hospital without additional neurological deficits.

DISCUSSION

We reported two patients with ruptured aneurysms who presented with severe major cerebral artery stenosis and multiple aneurysms in peripheral portions of collateral pathways of the stenoses. Cases similar to our case 1[12,20,26] and a patient with a distal PICA aneurysm combined with BA stenosis, similar to our case 2, have been reported.[1] To the best of our knowledge, our case 2 that involved multiple aneurysms in a PICA–AICA anastomosis associated with severe stenosis of the BA is the first of its kind reported in the literature.

There is no consensus on whether peripheral aneurysms attributable to hemodynamic stress are true aneurysms or pseudo-aneurysms and few reports have included histological diagnoses of resected aneurysms in the peripheral cerebral artery. Kim *et al*.,[13] who documented a true aneurysm involving a ruptured anterior thalamoperforating artery aneurysm with internal carotid artery occlusion, suggested that it developed as a result of hemodynamic stress. The aneurysm was composed of thickened fibrous tissue and devoid of elastic fibers. Others reported peripheral aneurysms that were found to be pseudo-aneurysms. Marks *et al*.[17] studied the histology of intranidal aneurysms that were hemodynamically associated with AVM. They observed that they were thin-walled vascular structures, but did not identify them as true or pseudo-aneurysms. Yuasa *et al*.[27] encountered a ruptured distal aneurysm associated with Moyamoya disease; it was histologically confirmed as a pseudo-aneurysm. Study of the surgical specimen from the aneurysm in the distal ACA (case 1) showed that it was subjected to hemodynamic stress attributable to altered blood flow due to severe right MCA stenosis. Histological study revealed no inflammatory cells but the presence of thick elastic fibers and the absence of fibers at the site of rupture, indicating a true saccular aneurysm. Based on our observations we conclude that even if they are located in the distal portion of the cerebral artery, aneurysms in collateral pathways can be true aneurysms except in cases with underlying vascular anomalies such as Moyamoya disease or AVM.

The appropriate treatment to address a distal aneurysm in a collateral pathway depends on whether it is a true aneurysm or a pseudo-aneurysm. In our case 2, we did not consider embolization because many PICA aneurysms are dissecting- or pseudo-aneurysms,[11,15,23] and thus are prone to perforation during coil embolization.[8,10,18] However, our findings in case 1 suggest that the aneurysm in case 2 was a true aneurysm; therefore, endovascular treatment was an appropriate treatment option. We performed

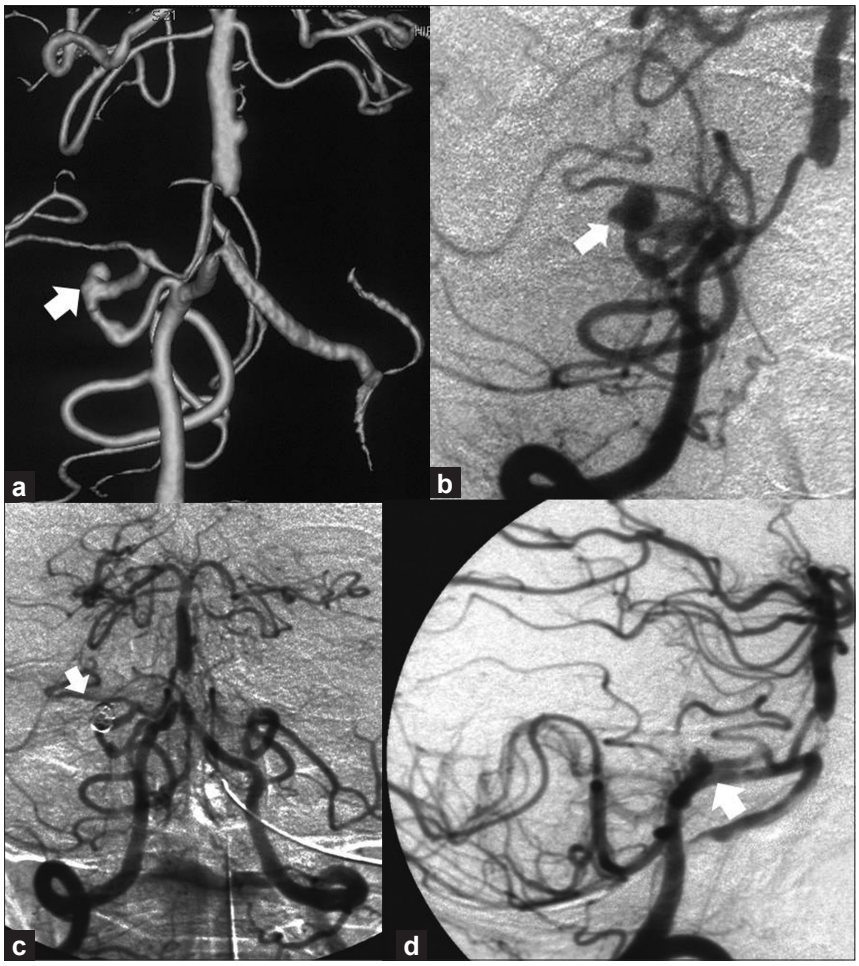

Figure 5: Case 2. (a) 3D-DSA acquired on day 7. The thick arrow indicates the enlarged aneurysm in the PICA–AICA anastomosis. (b) DSA obtained on day 15 pre-embolization. The thick arrow points to an aneurysm in the PICA–AICA anastomosis. There is aneurysmal enlargement. (c, d) DSA performed after selective aneurysmal coil embolization with parent artery preservation. The thick arrow in (c) indicates the preserved PICA–AICA anastomosis. The thick arrow in (d) points to a small neck remnant left over from the embolized aneurysm

selective aneurysmal coil embolization and completed the procedure successfully without perforation.

Of our two patients with ruptured aneurysms in collateral pathways associated with major cerebral artery stenoses, one had a histologically proven true saccular aneurysm. In patients with peripheral aneurysms in a collateral pathway, the arterial structure must be studied carefully. If the major artery stenosis is associated with vascular variations only, the presence of a true distal aneurysm must be considered.

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