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Ruptured Distal Lenticulostriate Artery Aneurysm Associated with Ipsilateral Middle Cerebral Artery Occlusion - Case Report -

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ABSTRACT

A 49-year-old man presented with right basal ganglia hemorrhage due to a ruptured distal lenticulostriate artery aneurysm associated with ipsilateral occlusion of M1 segment of middle cerebral artery with moyamoya-type vessels. Conservative treatment was initially selected. However, the patient suddenly became drowsy with left hemiparesis due to recurrent bleeding 14 days after admission, and excision of the aneurysm was urgently performed via right frontotemporal craniotomy. The follow-up angiography showed successful resolution of the aneurysm. This report describes a rare case of a distal lenticulostriate artery aneurysm associated with ipsilateral middle cerebral artery occlusion with moyamoya-type vessels, which resulted in intracerebral hemorrhage on the right basal ganglia and intraventricular hemorrhage. **(Kor J Cerebrovascular Surgery 8:128-31, 2006)**

KEY WORDS : Cerebral aneurysm · Lenticulostriate artery · Moyamoya

Introduction

Spontaneous intracerebral hemorrhage (ICH) accounts for approximately 10 to 20% of all strokes and is a significant cause of devastating condition. Sixty percent of hypertensive hemorrhages occur in the basal ganglia. Although most hemorrhages have been found in elderly and hypertensive patients, a significant proportion occurs in younger patients and structural lesions such as aneurysms, arteriovenous malformation, and moyamoya disease can be identified. Ruptured distal lenticulostriate artery (LSA) aneurysms manifest as basal ganglia hemorrhage, intraventricular hemorrhage and, less commonly, subarachnoid hemorrhage.[2)4)5)8)10)11)13)15)] Cerebral angiography very rarely detects an aneurysm of the distal LSA. We present a case of a patient with ICH on the right basal ganglia and intraventricular hemorrhage due to a ruptured LSA aneurysm associated with ipsilateral MCA occlusion with moyamoya-type vessels.

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Case Report

A 49-year-old man presented with a sudden onset of headache and vomiting. The patient had no history of hypertension, infectious disease, heart disease, head injury, or intravenous drug abuse. On arrival at our hospital, the patient's blood pressure was 130/80 mmHg, pulse was 72 per minute, and respirations were 15 per minute. The patient was alert and had no neurological deficit. Significant laboratory results were not found. Specifically, there was no evidence of hematological disorder or infectious process. Computed tomography (CT) of the head revealed right basal ganglia hemorrhage with extension to whole ventricle (Fig. 1). Because he was relatively young and had no risk factor for ICH, magnetic resonance imaging (MRI) was performed and right MCA M1 occlusion was suspected. Then, cerebral angiography was performed and right carotid angiography

disclosed an occlusion of M1 segment of right MCA and ipsilateral moyamoya-type vessels. The circulation to the distal middle cerebral territory on the right was collateralized from leptomeningeal vessels from the right anterior and posterior cerebral arteries. A small aneurysm was also detected arising from the distal portion of lenticulostriate branch of the right MCA (Fig. 2). Left carotid and bilateral vertebral angiography detected no abnormality. Conservative treatment was initially selected because we thought that this aneurysm was presumed to be a pseudoaneurysm deeply located in the basal ganglia. CT of the head performed at 13 days after admission showed the resolving hemorrhage. On the 14th day of admission, however, the patient suddenly became drowsy and experienced the profound left-sided weakness. The power of the left limbs decreased down to grade II. A CT scan of the head demonstrated recurrent hemorrhage, which was considered to result from rerupture of the aneurysm (Fig. 3). The patient underwent urgent operation via right frontotemporal craniotomy. A small corticotomy on the cortical surface was made and hematoma was gently evacuated. A small aneurysm arising from the distal LSA could be also found. Excision of the aneurysm was successfully achieved without vessel sacrifice. The MCA division and its branch were exposed. Severe stenotic proximal MCA with collateral network was identified. The postoperative course was uneventful and the patient showed progressive neurological recovery. Angiography performed at 4 weeks after surgery showed complete obliteration of the aneurysm (Fig. 4). Microscopic examination of the aneurysm wall revealed a true aneurysm. The patient was discharged at 8 weeks after admission, when the left limb power improved up to grade IV.

Discussion

Distal LSA aneurysms are rare and characterized by deep location and uncertain natural history. Although the etiology was unclear, they have been demonstrated in patients with moyamoya disease, systemic lupus erythematosus, hypertension, arteriovenous malformation, and brain tumor.[5)11)12)15)] It has been reported that aneurysms located in unusual sites are sometimes caused by infection, vasculitis, trauma, or arteriosclerosis. Our patient presented with only unilateral MCA occlusion with development of moyamoya-like vessels associated with aneurysm. Similarly, there have been reports that distal LSA aneurysms have been associated with moyamoya phenomenon, particularly unilateral MCA occlusion with development of moyamoya-like ganglionic

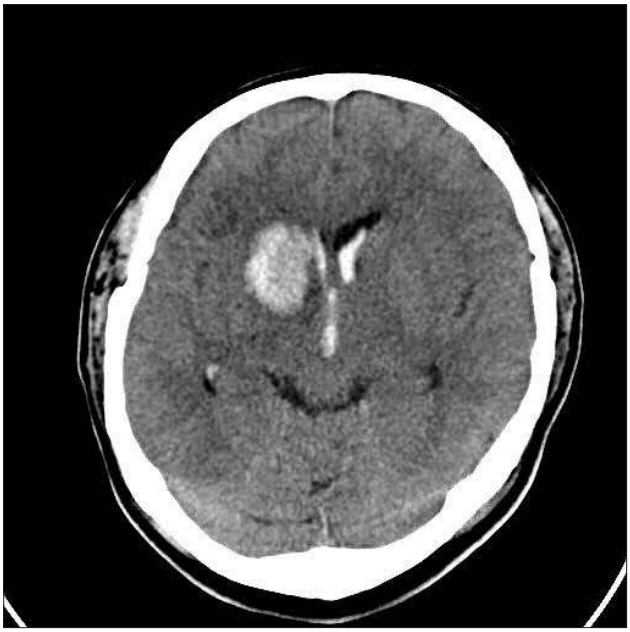

Fig. 1. A: Computed tomography on the day of admission shows intracerebral hemorrhage on the right basal ganglia and intraventricular hemorrhage.

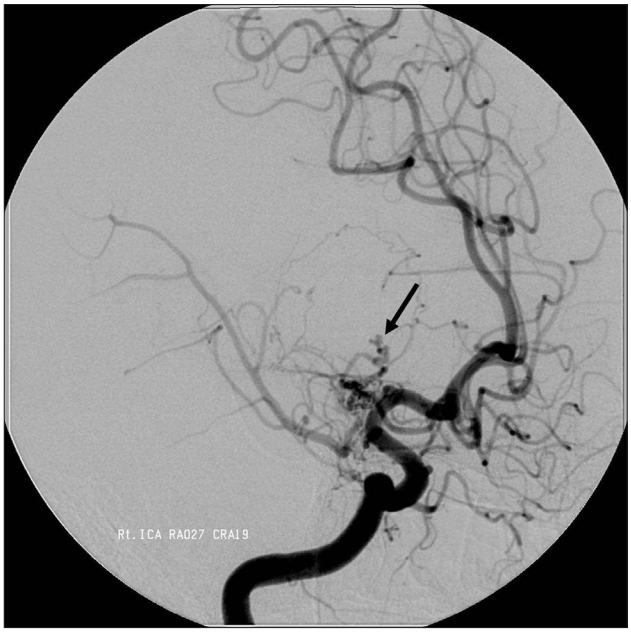

Fig. 2. Right carotid angiogram shows occlusion in the proximal middle cerebral artery, moyamoya-like vessels, and a small aneurysm (arrow) arising from the distal lenticulostriate artery.

collateral vessels, as seen in moyamoya disease.[3)8)9)13)]

It is well known that moyamoya disease is accompanied by cerebral aneurysms, with an incidence of 3 to 14%.[1)6)] Aneurysms associated with moyamoya disease are usually located in the basal ganglia or periventricular white matter.[7)] Moyamoya disease has presented with hemorrhage caused by either a rupture of a true aneurysm or pseudoaneurysms secondary to rupture of the fragile collateral vessels. The pseudoaneurysm spontaneously regresses on serial angiography. Although aneurysms associated with moyamoya disease are often considered to be a pseudoaneurysm, in particular, in case of peripheral artery aneurysms, they can be reruptured with an approximately 20~30% incidence and the clinical outcomes are very poor.[6)] Therefore, surgical intervention should be performed in patients with the increasing size of the aneurysm for prevention of rebleeding. It has been suggested that factors related to rebleeding and poorer outcome are sex (with women being more susceptible), massive intracranial hemorrhage and early recurrence.[8)] Cerebral aneurysms can develop in patients with MCA occlusion with development of ganglionic collateral vessels as the result of hemodynamic stress secondary to unilateral MCA occlusion, as seen in moyamoya disease. Theses aneurysms have been usually identified within the distal branches of the anterior and posterior choroidal arteries and LSA, less commonly, circle of Willis. MCA occlusion associated with moyamoya phenomenon is characterized by unilateral occlusion or stenosis restricted to the MCA M1 segment and no evidence of atheromatous plaques or stenosis in other cerebral arteries.[14)] Exact pathogenesis of MCA occlusion with moyamoya phenomenon is still under discussion. Although a congenital origin of the moyamoya phenomenon has been usually suggested, moyamoya phenomenon has been also reported in patients with neurofibromatosis, tuberculous meningitis, leptospirosis, previous radiation therapy, and arteriosclerosis.[3)] In this case, there was no evidence to support any of the aforementioned causes.

It has been reported that ICH is associated with higher incidence of vascular lesions in patients less than 45 years of age and in those without preexisting hypertension.[16)] Therefore, the clinical workup in patients presenting with ICH should include an MRI and cerebral angiography, especially in those patients who are young or have no known risk factors such as advanced age or hypertension. Our patient had no known risk factors for ICH. The initial CT showed basal ganglia hemorrhage that was characteristic of a hypertensive hemorrhage. However, the patient had no

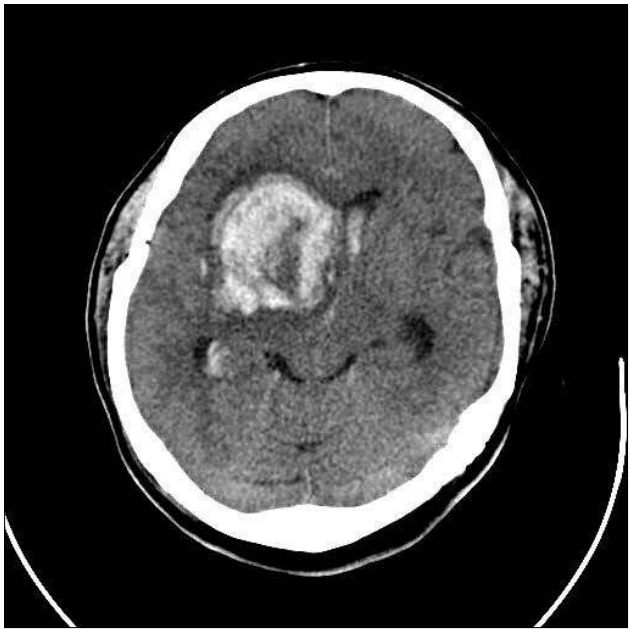

Fig. 3. Computed tomography on the 14th day of admission shows recurrent hemorrhage.

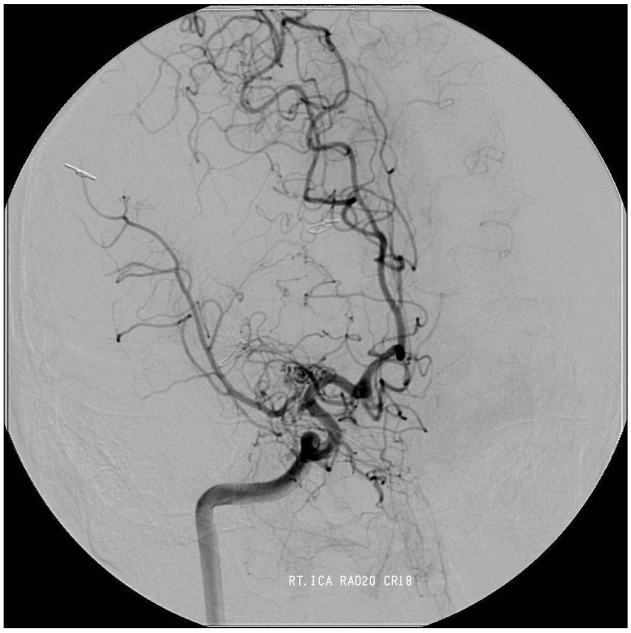

Fig. 4. Right carotid angiogram performed at 4 weeks after surgery demonstrates complete obliteration of the aneurysm seen on the preoperative study.

history of hypertension and the patient was normotensive on admission. In this case, there was a propensity for aneurysms to develop. Distal LSA aneurysm associated with unilateral MCA occlusion with moyamoya-like vessels could be found. We believed that the risk of rebleeding in our case was low because the patient was male and the amount of hemorrhage was small. In addition, deep location of the aneurysm made us consider conservative treatment. However, the aneurysm reruptured after 14 days after initial bleeding episode and the patient's neurological status suddenly deteriorated. Hematoma evacuation and surgical excision of the aneurysm were urgently performed and we could preserve the vessel while repairing the lesion.

Aneurysms arising from distal branches of the LSA are challenging lesions to treat, due to their fragility, the risk of damaging important collateral vessels and deep location and no clear guidelines exist regarding the best treatment. While conservative treatment and careful follow-up angiography seem appropriate for lesions consistence with pseudoaneurysm, surgical or endovascular means have been described in selected cases.[2)4)5)8)10)11)13)15)] Surgical or endovascular means of the LSA aneurysm usually carries a risk of damage to the eloquent neuronal pathways due to deep location in the basal ganglia and they also carry the risk of parent artery occlusion leading to cerebral infarction of the perforating artery and further clinical deterioration. However, these aneurysms should be treated surgically to prevent rebleeding in case of typical saccular aneurysms.

Conclusion

Spontaneous basal ganglia hemorrhage is commonly associated with preexisting hypertension in the elderly. However, vascular lesions are frequently identified in young patients without a history of hypertension. They should be fully evaluated with MRI and cerebral angiography. Furthermore, we should pay attention to detect aneurysms in patients with hemorrhage associated with moyamoya phenomenon. While conservative treatment and careful follow-up angiography seem appropriate for lesions consistence with pseudoaneurysm, surgical intervention is recommended for prevention of rebleeding in selected cases.

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