



A Patient with a Distal Medial Lenticulostriate Artery Aneurysm and Intraventricular Hemorrhage Treated by Endovascular Treatment

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Objective: A patient with a ruptured distal medial lenticulostriate artery (mLSA) aneurysm presenting with intraventricular hemorrhage was successfully treated using endovascular treatment.

Case Presentation: A 60-year-old woman presented with impaired consciousness. Radiological examination revealed intraventricular hemorrhage caused by a rupture of a distal mLSA aneurysm. Using endovascular technique, approaching contralaterally through the anterior communicating artery (ACoM), complete occlusion of the aneurysm was achieved by N-butyl-2-cyanoacrylate (NBCA) injection. The postoperative course was uneventful.

Conclusion: Intraventricular aneurysms at a distal site of the perforating arteries are rare. Although there have been reports on patients with distal mLSA aneurysms treated by open surgery or conservative therapy, endovascular therapy should also be considered as a treatment option.

Keywords ▶ intraventricular hemorrhage, intraventricular aneurysm, medial lenticulostriate artery, endovascular therapy, N-butyl-2-cyanoacrylate

Introduction

Distal perforating artery aneurysms are rare and sometimes exhibit “pure” primary intraventricular hemorrhage (PIVH) not accompanied by intracerebral hemorrhage or subarachnoid hemorrhage.¹⁾

We report a patient with a distal medial lenticulostriate artery (mLSA) aneurysm who presented with intraventricular

hemorrhage and was successfully treated by embolization with N-butyl-2-cyanoacrylate (NBCA).

Case Presentation

Patient: A 60-year-old female

Chief complaint: Impaired consciousness

Past medical history: She had a history of atrial fibrillation and dilated cardiomyopathy, and was receiving oral warfarin after implantation of a biventricular pacing implantable cardioverter defibrillator.

History of present illness: She was found lying at home with her family and transported to our hospital by ambulance. When she arrived at the hospital, the Japan Coma Scale (JCS) score was II-10 and the Glasgow Coma Scale (GCS) score was 14 (E3V5M6), and no clear neurological deficits were observed. Non-contrast head CT revealed hemorrhage in the third and fourth ventricles, and bilateral lateral ventricles with left dominance (**Fig. 1**). Hemorrhage was confined within the ventricles. To find the source of bleeding, brain 3D-CTA was performed. In the arterial phase, the source of bleeding was unclear. In the venous phase, there was spot enhancement on the lateral wall of

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Received: September 22, 2019; Accepted: May 2, 2020

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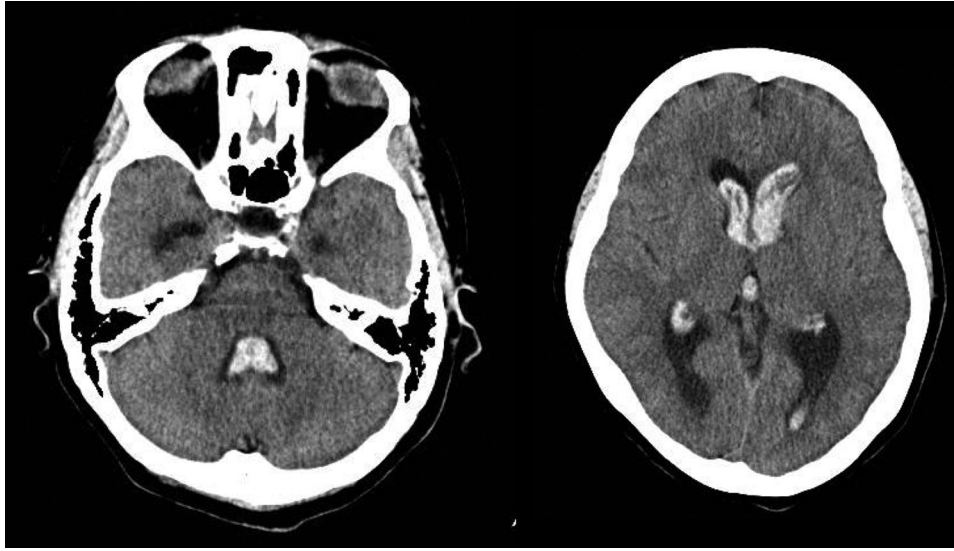


Fig. 1 Plain head CT images at onset. There are high-density hematomas in the bilateral lateral ventricles with left dominance. Hematomas are also present in the third and fourth ventricles, and mild ventricular dilatation can be observed.

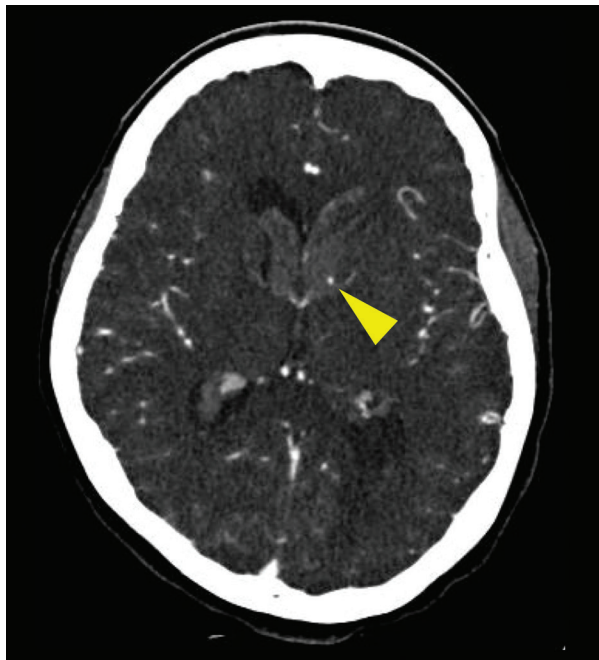


Fig. 2 Cerebral 3D-CTA source image. A lump of contrast is observed as spot enhancement in the anterior horn of the left lateral ventricle (arrowhead).

the anterior horn of the left lateral ventricle, suggesting the source of bleeding (**Fig. 2**). After admission, as there was no progression of impaired consciousness, ventricular drainage was not performed and blood pressure control was initiated in the intensive care unit.

On the third day of admission, cerebral angiograph revealed an aneurysm (3.8 mm in size) at the distal site of

the mLSA arising from the proximal A1 segment of the left anterior cerebral artery (ACA), and was considered to be a ruptured aneurysm (**Figs. 3** and **4**). To prevent re-bleeding, aneurysm embolization was performed.

Endovascular treatment: Under general anesthesia, the right femoral introducer was replaced by a 4Fr 80-cm ASAHI FUBUKI Dilator Kit (Asahi Intecc, Aichi, Japan). After systemic heparinization, a guiding sheath was navigated into the petrous portion of the left internal carotid artery. A 4.2Fr ASAHI FUBUKI (Asahi Intecc) was used as a distal access catheter. Using a Mirage (Medtronic Irvine, CA, USA) guide wire, a Marathon (Medtronic) catheter was advanced to the left A1. However, due to the mLSA arising in a direction reverse to the path of the left A1, catheterization was difficult. Compression study of the right common carotid artery confirmed the anterior communicating artery (ACoM), and a contralateral approach was attempted. The guiding sheath was introduced into the petrous portion of the right internal carotid artery, and the Marathon was advanced through the ACoM into the mLSA origin, but was unable to be advanced to a site near the aneurysm. After changing to a combination of a DeFrictor nano catheter (Medico's Hirata, Osaka, Japan) and TENROU S10 (Kaneka Medix, Osaka, Japan) guide wire, mLSA cannulation was possible. Selective angiography from the DeFrictor confirmed the location of the catheter tip proximal to the aneurysm, and a 33% NBCA-Lipiodol mixture was slowly injected from the DeFrictor. After the perforating artery distal and proximal to the aneurysm were

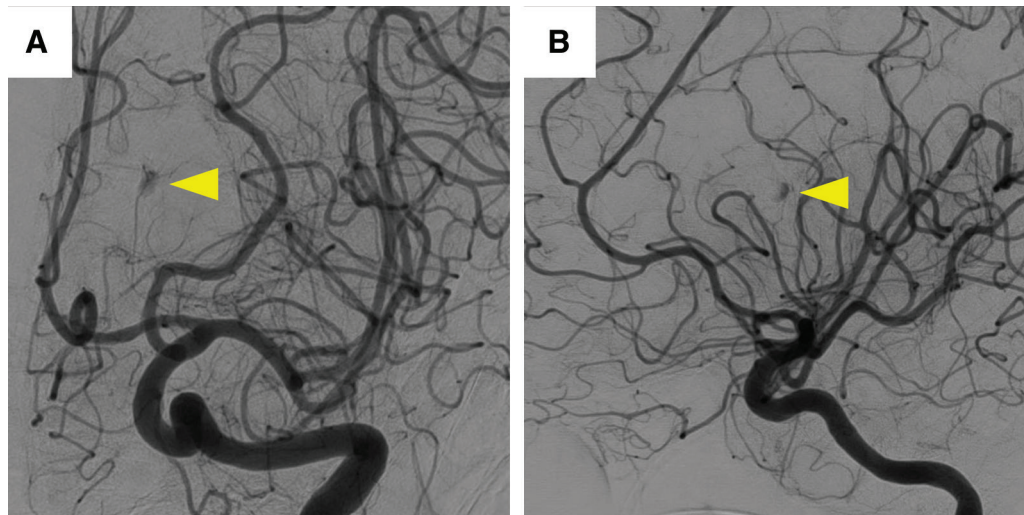


Fig. 3 Cerebral angiograms of the left internal carotid artery (a, frontal image; b, lateral image). A saccular aneurysm is observed adjacent to the lateral ventricle (arrowheads).

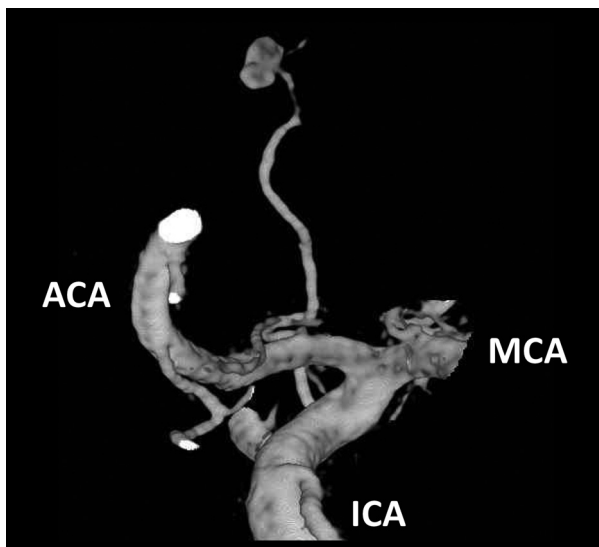


Fig. 4 3D rotation angiography of the left internal carotid artery. An aneurysm (3.8 mm in size) is present at a distal site of the mLSA arising from the A1 segment of the left ACA. ACA: anterior cerebral artery; ICA: internal carotid artery; MCA: middle cerebral artery; mLSA: medial lenticulostriate artery

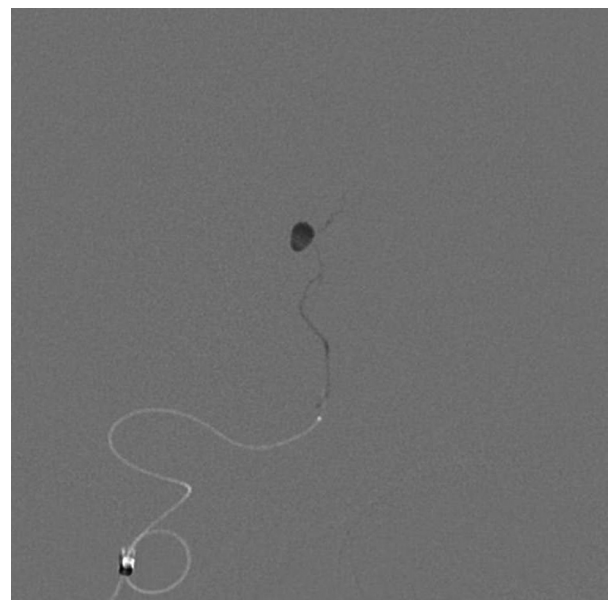


Fig. 5 Imaging after NBCA injection. The inside of the aneurysm was filled with NBCA, showing complete embolization. Slight reflux of NBCA was observed in the proximal artery. NBCA: N-butyl-2-cyanoacrylate

confirmed to be filled with NBCA, the DeFrictor was removed (**Fig. 5**). Angiography after NBCA injection demonstrated disappearance of the aneurysm.

Course: There were no signs of hemorrhagic complications or cerebral infarction on the postoperative head CT. Neither impaired consciousness nor paralysis developed. Lipiodol remained as a high-density spot on the lateral ventricular wall of the anterior horn of the left lateral ventricle. Subsequently, sepsis triggered by pneumonia

developed and a decrease in muscle strength associated with disuse remained. However, after 6-month rehabilitation, she was discharged to home with a modified Rankin Scale (mRS) of 1.

Discussion

The majority of intraventricular hemorrhage is secondary to intracerebral or subarachnoid hemorrhage. PIVH

Table 1 Previous reports on distal mLSA aneurysms

Author, year	Age/sex	Location	Associated diseases	Treatment	Pathology	GOS
Hagihara et al., 2006	55/F	Caudate nucleus Intraventricular	None	Craniotomy	Not performed	GR
Matushita et al., 2007	5/M	Intraventricular	None	Craniotomy	True aneurysm	GR
Ellis et al., 2011	71/F	Intraventricular	FD	Conservation	Not performed	GR
Kim et al., 2013	28/M	Intraventricular	None	Craniotomy	Pseudo aneurysm	GR
Srivastava et al., 2013	45/F	Intraventricular	None	Conservation	Not performed	GR
Nomura et al., 2018	81/F	Caudate nucleus Intraventricular	None	Conservation	Not performed	GR
Present case	60/F	Intraventricular	None	Endovascular therapy	Not performed	GR

FD: fibromuscular dysplasia; GOS: Glasgow outcome scale; GR: good recovery; mLSA: medial lenticulostriate artery

accounts for 2%–3.3% of all intracranial hemorrhage.^{2–4} The causes of PIVH include hypertension, arteriovenous malformation (AVM), aneurysms, trauma, Moyamoya disease, cavernous malformation, and coagulation abnormalities.^{5,6} Intraventricular aneurysms can also cause PIVH.^{1,2,5,6} Intraventricular aneurysms occurring at the distal site of the perforating arteries are rare, and there has been no established consensus on their natural history or treatment principles.

The present case had an intraventricular aneurysm developing at a distal site of the mLSA arising from the A1 segment of the ACA. Spot enhancement inside of an intracerebral hematoma (or “spot sign”) is a sign of intracerebral hemorrhage indicating extravasation of the contrast medium into the hematoma, and is a risk factor for an enlargement of the hematoma. The “spot sign” reflects bleeding in the cerebral parenchyma, and whether it indicates primary or secondary vascular injury is unknown, and whether or not related to aneurysms is also unclear.⁷ In intraventricular hemorrhage, unlike intracerebral hemorrhage, there is no brain parenchyma or vascular structures that can be secondarily injured around the hematoma. The bleeding is considered to be due to primary vascular injury, and secondary injuries of the blood vessels are unlikely. Therefore, when spot enhancement is observed on the lateral side of PIVH, a distal perforating artery aneurysm should be considered, although differentiation between a true aneurysm and pseudoaneurysm is difficult. This case was a case of PIVH not accompanied by cerebral parenchymal hemorrhage. Considering the possibility of an intraventricular aneurysm based on a previous study,⁸ cerebral angiography was performed, which led to the diagnosis. When similar enhancement is observed within the hematoma of PIVH, cerebral angiography should be considered.

To our knowledge, seven patients with distal mLSA aneurysms, including the present patient, have been reported (**Table 1**).^{8–13} Their ages varied from 5 to 81 years. Of the seven patients, five presented with PIVH, and most of them had idiopathic aneurysms without underlying disease. In patients on whom pathological examination was performed, a true aneurysm and pseudoaneurysm were observed in one each. Conservative treatment was performed for three patients and radical treatment by direct surgery for three patients. In the three patients who underwent conservative treatment, the duration from onset to treatment varied from 3 days to around 1 month. All reported patients had favorable outcomes, and the present patient was the first to undergo endovascular treatment. In this patient, no pathological diagnosis was made due to the endovascular treatment. Whether the aneurysm is a true aneurysm or pseudoaneurysm is difficult to determine only by morphological evaluation on angiography.

Intraventricular aneurysms are treated by conservative treatment, direct surgery, or endovascular treatment. Conservative treatment for cerebral aneurysms is performed when their spontaneous regression is expected. Previous reports described some patients with distal perforating artery aneurysms associated with Moyamoya disease in whom spontaneous aneurysm regression was observed on cerebral angiograms.^{14,15} Radical surgery indicated only for patients not exhibiting spontaneous regression was also reported. However, there have been no reported patients with AVM-associated or idiopathic aneurysms who exhibited spontaneous regression. In such patients, treatment to prevent re-rupture may be necessary. Some AVM-associated or idiopathic aneurysms were reported to be true aneurysms.^{16,17} Thus, regarding pathology, there is not only pseudoaneurysm formation accompanied by hemorrhage but also intracerebral hemorrhage associated with

rupture of a cerebral aneurysm that had been present before the onset. In addition, although spontaneous regression of cerebral aneurysms is considered to occur due to spontaneous thrombosis in the aneurysm, aneurysm regression on angiography can also be transient and does not always mean re-rupture was prevented.¹⁸⁾ Pathological evaluation of distal mLSA aneurysms in previous studies was insufficient; however, six (85%) of the seven reported patients had idiopathic intraventricular aneurysms. The prognosis of intraventricular aneurysms after re-rupture may be markedly poor, and early radical surgery can be a treatment option.

In direct surgery, the ventricle must be reached via the cerebral parenchyma using a trans-cortical or trans-callosal approach. In either approach, when the approach site is appropriate, surgical invasion can be minimized. However, when surgery for the dominant hemisphere is necessary, there is a risk of cortical injury or postoperative development of epilepsy. When only part of the aneurysm is exposed to the intraventricular space, the parent artery should be confirmed by sacrificing a part of the parenchyma in the ventricular wall for complete aneurysm occlusion.

There are several recent studies on endovascular treatment for distal perforating artery aneurysms^{19–22)} in which endovascular treatment was performed for aneurysms developing in the distal lateral lenticulostriate artery (ILSA) as the perforating artery of the middle cerebral artery (MCA). However, there have been no studies on endovascular treatment for aneurysms at a distal site of the mLSA arising from the ACA. This may be partly due to the small number of cases, but there have been cases, such as that reported by Matushita et al.,¹⁰⁾ in which endovascular treatment was abandoned due to difficulty in approach. Although the mLSA arose from the ACA in this patient, it arises in an acute angle to the course of the ACA and the ipsilateral approach was difficult. In patients such as ours demonstrating well-developed AComA, the contralateral approach may enable catheter insertion into the mLSA.

In endovascular treatment, choices of embolization using coils or liquid embolic material such as NBCA. To insert the catheter to the distal site of the perforating artery, embolization with a liquid embolic material using a flow directed catheter is rational.^{19–21)} Although the mLSA territory differs among individuals, cerebral infarction in the perfusion area of a well-developed mLSA has a risk of apathy and changes in personality.^{23,24)} As this patient had undergone implantation of a biventricular pacing implantable cardioverter defibrillator, MRI

evaluation was not possible, but no development of clear cerebral infarction was noted on postoperative CT. In patients such as ours in whom catheterization close to the aneurysm is impossible, proximal perforating artery embolization is performed, which increases the risk of cerebral infarction in the perforating artery perfusion area; therefore, careful selection between embolization and direct surgery is necessary.

Conclusion

Embolization with NBCA was performed in a patient with a distal mLSA aneurysm presenting with PIVH, and a favorable outcome was obtained. When CTA demonstrates spot enhancement on the lateral side of PIVH, there is a possibility of a distal perforating artery aneurysm, and cerebral angiography and radical surgery to prevent re-bleeding should be considered. In the future, additional cases are necessary, but endovascular treatment for this condition may be a safe and effective treatment option.

Acknowledgment

The authors thank Dr Alexander Zaboronok, Department of Neurosurgery, University of Tsukuba, for manuscript revision.

Disclosure Statement

We declare no conflict of interest.

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