

DISTAL LENTICULOSTRIATE ARTERY ANEURYSM RUPTURE PRESENTING AS INTRAPARENCHYMAL HEMORRHAGE: CASE REPORT

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OBJECTIVE AND IMPORTANCE: Aneurysms involving the distal lenticulostriate artery branches are a rare cause of spontaneous intracerebral hemorrhage. We report a case of ruptured lenticulostriate aneurysm in an otherwise healthy patient and review the literature on this topic.

CLINICAL PRESENTATION: Computed tomography showed a right basal ganglia hemorrhage in a 44-year-old Native American woman with acute left hemiparesis. Cerebral angiography showed a 2-mm aneurysm in a distal lenticulostriate artery branch.

TECHNIQUE: A pterional craniotomy was performed on a delayed basis using computed tomographic angiography and frameless stereotactic guidance. The basal ganglia hematoma was evacuated, and the aneurysm was identified and clipped using microsurgical technique. Because the base of the aneurysm involved a portion of the parent vessel wall, it was wrapped with cotton and reinforced with cyanoacrylate glue. The patient did well after surgery and was discharged to home with outpatient rehabilitation on the third postoperative day.

CONCLUSION: Although intraparenchymal hemorrhages are fairly common, the underlying vascular abnormality is rarely identified. Most are related to hypertensive vascular degeneration, rupture of a Charcot-Bouchard aneurysm, or both. When intracerebral hemorrhage occurs in young patients, however, aggressive investigation is warranted to rule out a structural vascular abnormality.

KEY WORDS: Angiography, Cerebral hemorrhage, Intracerebral hematoma, Stroke

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Intracerebral hemorrhage (ICH) is reported as the cause of stroke in 10 to 20% of all strokes and is a significant cause of disability. The reported incidence varies with ethnicity. The rate among Caucasians is 7 to 12/100,000 persons per year, and the rate among Asians is as high as 60/100,000 persons per year (6, 19). Although most hemorrhages occur in the elderly, a significant proportion occur in the young. Many of these hemorrhages are attributed to microaneurysms formed in hypertensive patients. These aneurysms are rarely discovered with conventional imaging techniques either before or after a hemorrhage. We report a basal ganglia hemorrhage caused by rupture of a distal lenticulostriate artery aneurysm in an otherwise healthy individual.

CASE REPORT

A 44-year-old Native American woman sought treatment in the emergency department for the acute onset of left-sided weakness and headache. She had no history of headache, hypertension, or diabetes and took no prescription medicines. She had no cognitive abnormalities. Her pupils were equal and reactive, and her speech was normal. She had a dense facial droop on the left, no motor function in her left upper extremity, and anti-gravity strength in her left leg. Motor function on her right side was normal.

Computed tomography of the head at admission showed a right basal ganglia hemorrhage compromising the putamen and globus pallidus (*Fig. 1*). Because she was relatively

young and had no risk factor for ICH (e.g., hypertension, substance abuse), cerebral angiography was performed; it showed a 1- to 2-mm aneurysm arising from a distal lenticulostriate artery (Fig. 2). The remainder of the examination disclosed nothing abnormal.

The patient was initially referred for endovascular treatment; however, on the basis of the results of the diagnostic angiogram, she was considered a poor candidate. It was judged that cannulation of this small branch would be difficult and that endovascular options would be limited to vessel sacrifice with associated risks of additional deficits. She was transferred to an inpatient rehabilitation facility on posthemorrhage Day 3, with plans for follow-up angiography in 7 to 10 days. She was discharged to home after 1 week with near-normal function in her left arm and walking with a cane.

A repeat angiogram performed on posthemorrhage Day 10 demonstrated persistent filling of the aneurysm, and the patient was referred for surgical treatment. A stereotactic image-guided pterional craniotomy (StealthStation; Medtronic Surgical Navigation Technologies, Louisville, CO) was performed. The sylvian fissure was split, and the hematoma cavity and aneurysm were localized using computed tomographic angiography (Fig. 3). The hematoma was evacuated, and the aneurysm was identified and clipped using microsurgical technique (Fig. 4). A small residual at the base, which could not be clipped because it involved the parent vessel, was

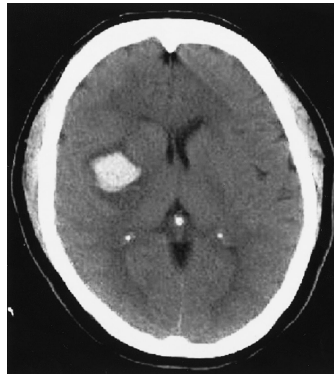


FIGURE 1. Admission axial computed tomographic scan of the patient, who experienced the acute onset of right hemiparesis.

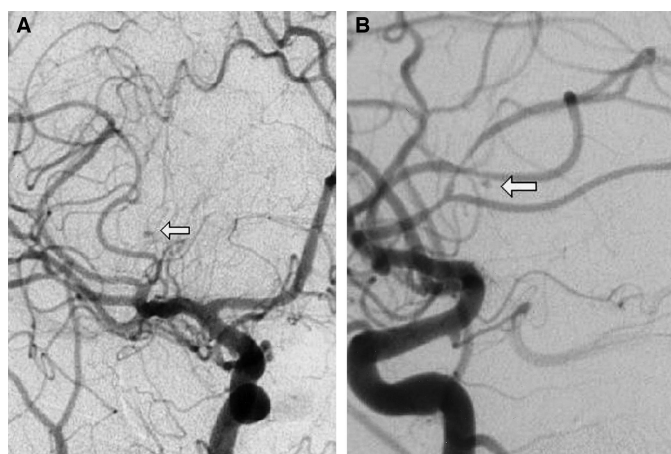


FIGURE 2. Digital subtraction angiograms in the oblique (A) and lateral (B) views showing a 1- to 2-mm aneurysm on the distal lenticulostriate artery (arrows).

reinforced with cotton and cyanoacrylate glue. The follow-up angiogram demonstrated filling of the parent vessel and partial residual filling of the aneurysm (Fig. 5). The patient did well and was discharged to home on postoperative Day 3 with outpatient therapy. At a 4-month follow-up examination, she had recovered complete motor and sensory function in her affected arm and leg, and her facial droop had resolved.

DISCUSSION

Lenticulostriate artery aneurysms are rare. When present, they most often arise secondarily to an underlying vascular abnormality such as an arteriovenous malformation, moyamoya disease, or vasculitis (1, 7, 12, 13, 20). The first case of an aneurysm involving the distal lenticulostriate artery was described more than 10 years ago (8). Since then, five other reports have demonstrated lenticulostriate aneurysms in seven patients (Table 1) with no other concomitant vascular disease (5, 11, 14, 16, 17).

In the 19th century, Charcot and Bouchard (3) first hypothesized that ICH originated from microaneurysms (<1 mm) arising from the perforating arteries (14). Cole and Yates (4) strengthened this hypothesis by using microangiographic techniques to demonstrate a significantly higher incidence of microaneurysms in patients with hypertension. Since then, however, the validity of the original Charcot-Bouchard hypothesis has been debated.

Other investigators have found no true microaneurysms in patients with ICH. They have observed pseudoaneurysms, dissecting aneurysms, and lipohyalinosis in pathological specimens from evacuated hematomas (2, 4, 18). Atherosclerosis is thought to play a role in weakening the vessel wall of the lenticulostriate arteries, leading to degeneration of the media and rupture (18). The hemorrhage is considered to be the direct result of rupture or dissection caused by degeneration of the vessel and hemodynamic stress (15).

The low incidence of microaneurysms in patients with ICH may reflect the low rate of angiographic evaluation performed in these patients, particularly those with typical risk factors such as advanced age and hypertension. The increased risks of angiography in this latter population tend to limit evaluation (10); however, a more aggressive approach seems warranted in younger patients and those without hypertension. In a prospective study of 80 patients with ICH, Halpin et al. (9) reported that angiography identified structural lesions in 74% of patients less than 50 years of age. This number increased to 83% in

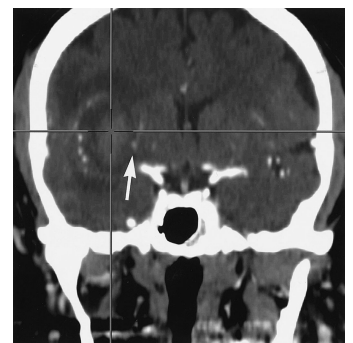


FIGURE 3. Intraoperative stereotactic computed tomographic angiogram. The small aneurysm (arrow) arising from the lenticulostriate artery is seen adjacent to the hematoma.

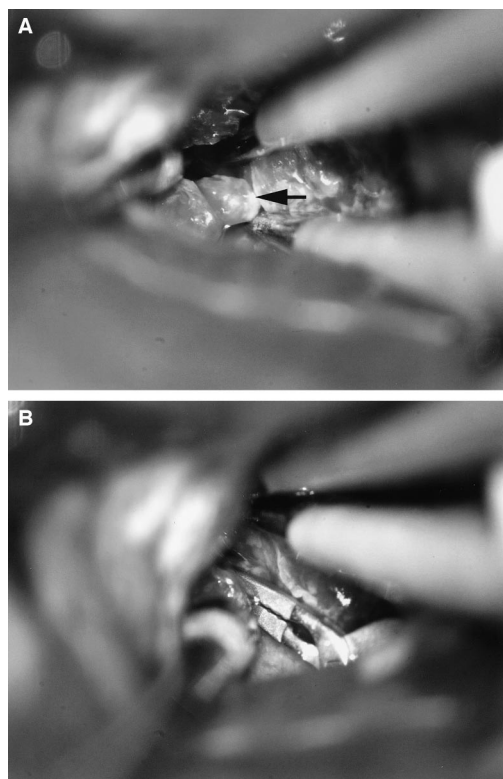


FIGURE 4. Intraoperative photographs showing the aneurysm (arrow) arising from the lenticulostriate artery before (A) and after (B) clip application.

normotensive patients in this age group. Zhu et al. (20) examined the effect of age, hypertension, and site of hemorrhage on the possibility of finding an underlying vascular abnormality by cerebral angiography in a prospective study of 206 patients with ICH. They found a significantly higher incidence of vascular lesions in patients less than 45 years of age (50% versus 18%, $P <$

0.001) and in those without preexisting hypertension (44% versus 9%, $P <$ 0.001).

In the present case, cerebral angiography revealed a lenticulostriate artery aneurysm arising in the region of the hemorrhage. Little is known about the natural history of these lesions. The initial thought was that the lesion might spontaneously thrombose; however, follow-up angiography demonstrated persistent filling of the aneurysm 10 days after the initial hemorrhage.

Endovascular treatment was considered but ruled out because of the difficulty

posed by cannulating the lenticulostriate branches and the desire to preserve flow in the involved artery. In considering a surgical approach to this lesion, we noted that computed tomographic angiography demonstrated the aneurysm and clearly showed its relationship to the hematoma. Combining frameless stereotactic guidance with computed tomographic angiography allowed the aneurysm to be readily localized during surgery.

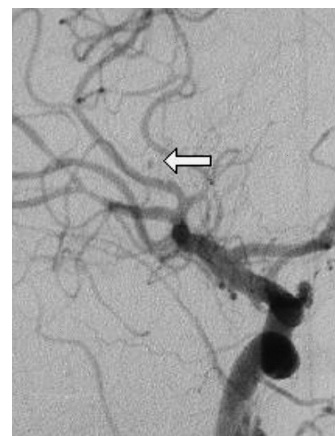


FIGURE 5. Postoperative angiogram demonstrating residual filling of the wrapped aneurysm (arrow). Note the patency of the parent vessel.

CONCLUSION

Spontaneous ICH accounts for approximately 10 to 20% of all strokes. Hypertensive vasculopathy is the most common cause of hemorrhage in the elderly; however, structural pathological findings are frequently identified in younger patients.

TABLE 1. Reported cases of angiographically observed lenticulostriate artery aneurysms in patients with no concomitant neurovascular pathological findings^a

Series (ref. no.)	Age/sex	Presentation	Treatment	Outcome
Gupta et al., 1989 (8)	36 yr/F	Hemiplegia/hematoma	Conservative	Residual hemiparesis
Oka, et al., 1991 (16)	59 yr/F	Incidental (SAH from separate aneurysm)	Surgical wrapping	No deficit
	59 yr/F	Incidental (SAH from separate aneurysm)	Surgical clipping	No deficit
	44 yr/F	Hemiparesis/hematoma	Hematoma evacuation	No rebleeding at 8 yr
Petrela, et al., 1992 (17)	27 yr/M	Hemiparesis/hematoma	Hematoma evacuation/clipping	Residual hemiparesis
Endo, et al., 1996 (5)	12 yr/F	Hematoma	Surgical clipping	No deficit
Kaptain, et al., 2001 (11)	2 mo/M	Hemiparesis/hematoma	Surgical excision	Residual hemiparesis
Maeda, et al., 2001 (14)	62 yr/F	Hemiplegia/hematoma	Surgical excision	Residual hemiparesis

^a SAH, subarachnoid hemorrhage.

Cerebral angiography and endovascular or surgical management of vascular lesions should be considered in all patients less than 50 years of age who present with spontaneous ICH.

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COMMENTS

This case report by Horn et al. illustrated the presentation (acute putaminal hematoma) and microsurgical treatment of a 2-mm Charcot-Bouchard aneurysm in a distal lenticulostriate artery.

Two issues require in-depth discussion. First, spontaneous putaminal intracerebral hemorrhage is commonly associated with preexisting hypertension in the elderly population. The majority of these patients do well without further investigation and treatment (1). In younger patients (<45 yr of age), it is recommended that angiography should be performed to exclude intracranial aneurysms, arteriovenous malformation, and moyamoya disease (2). The natural history of these Charcot-Bouchard aneurysms and microaneurysms is not consistently demonstrated by modern imaging, although we are all familiar with the occurrence of rebleeding in this group of patients. Second, when a sizable aneurysm of 2 mm is demonstrated, as was described in this case report, intervention is reasonable. This case report has demonstrated a very important point: the treatment of choice for this lesion is not endovascular coiling but rather microsurgical clipping and wrapping of the abnormal base of the Charcot-Bouchard aneurysm.

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The authors have reported an important case that emphasizes the need to fully investigate patients with spontaneous intracerebral hemorrhage when the pathogenesis is unclear. When patients present with deep cerebral or brainstem hemorrhages, it is often assumed that hypertension is the cause. Although hypertension accounts for a significant number of spontaneous hemorrhages in the chronically hypertensive and elderly population, younger patients without a clear history of hypertension should be fully evaluated with cerebral angiography before the hemorrhage is attributed to hypertension.

This case also illustrates an innovative surgical approach to this challenging lesion. The use of frameless stereotactic guidance was critical in identifying the aneurysm. We recently treated a very similar case in which we used intraoperative angiography for intraoperative localization of the aneurysm. Distal lenticulostriate aneurysms are very difficult to identify through a standard frontotemporal approach with sylvian fissure splitting, and some form of intraoperative guidance is useful in minimizing dissection. The authors have presented a thorough evaluation and a creative surgical solution.

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Although the need for angiography to rule out structural lesions in young patients with deep intracerebral hemorrhages is well established, it cannot be emphasized enough,

especially for those without risk factors. Although perforating artery aneurysms are rare discoveries, arteriovenous malformations, cavernous malformations, and tumors are not uncommonly revealed during aggressive radiological workup. The evaluation should include not only angiography but also magnetic resonance imaging, and consideration should be given to early and delayed imaging, because the clot may temporarily obscure small lesions.

What makes the present case so impressive is the authors' ability to preserve the vessel while repairing the lesion. To the best of our knowledge, most historical efforts to deal with these lesions have resulted in vessel sacrifice, especially when endovascular techniques were used. This is primarily because these lesions tend to involve the entire circumference of the vessel rather than a part of a vessel bifurcation. Matters are only made more challenging by the small caliber of the parent arteries, which predispose them to catheter spasm, near flow arrest, and thrombosis.

In this case, the authors were able to spare the vessel by approaching it directly, clipping the dome and wrapping the residual dysplasia. One can only hope that this results in

long-term stability and patency of the segment, because it is not entirely clear that trapping or occluding such a vessel always results in clinical deterioration, especially in cases with associated hematomas and marked baseline deficits. Nonetheless, given this patient's incomplete hemiparesis on presentation, the relatively small size of the clot, and the location of the clot lateral to the internal capsule, one might argue strongly for a vessel-preserving approach using computed tomographic angiographic guidance. Her remarkable recovery certainly attests to this and underscores the ability of patients, particularly young ones, to show dramatic functional recoveries after seemingly devastating deep-seated bleeds.

With longer follow-up, this case may encourage surgeons to manage those patients capable of recovering function with open surgical techniques first, while retaining endovascular approaches, especially those destined to obliterate the vessel, for recurrences and those in whom the clot has wreaked irrevocable damage.

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