Controversy in the Management of Lenticulostriate Artery Dissecting Aneurysm: A Case Report and Review of the Literature

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Key words
- Aneurysm
- Arterial dissection
- Intracerebral hemorrhage
- Lenticulostriate artery

Abbreviations and Aconryms
CT: Computed tomography
IEL: Internal elastic lamina
LSA: Lenticolustriate aneurysm

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BACKGROUND: **Intracranial arterial dissection is an uncommon but well-recognized entity. Treatment remains variable, ranging from observation to intervention via the use of either surgical or endovascular techniques. Aneurysms along the lenticulostriate artery have been reported in only 41 patients. With the current case study we illustrate the effectiveness of observation in the context of a dissecting lenticulostriate aneurysm and discuss other approaches that have been used in the treatment of this particular entity.**

CLINICAL SUMMARY: **An accomplished mountain climber presented, after coitus, with acute headache, mild facial weakness, and forgetfulness. Vascular imaging studies revealed a right putaminal hemorrhage secondary to a 3-mm lenticulostriate artery dissecting aneurysm. Clinically, the patient did well, with marked improvement in presenting symptoms enabling his return to mountain climbing. Follow-up angiography showed spontaneous resolution of the arterial dissecting aneurysm. Among the 41 reported cases, 19 were idiopathic, 5 associated with hypertension, and 17 related to various conditions such as Moyamoya disease, arteriovenous malformation, systemic vasculitis, intraventricular tumor, or substance abuse. Of the 42 cases, including the present case, 28 were surgically or endovascularly managed and 12 observed. Only one of the reported cases, a 33-year-old man with Moyamoya disease, who was managed conservatively, died of rebleeding.**

CONCLUSION: **There is no common consensus in the literature on a single treatment strategy for a lenticulostriate artery aneurysm. The present case illustrates that observation and follow-up vascular imaging can be an important treatment strategy, allowing healing of the vessel wall and disappearance of the dissecting aneurysm.**

INTRODUCTION

Aneurysms arising along the course of the lenticulostriate artery (LSA) are uncommon, with only 41 cases described in medical literature. The majority of these cases are likely dissecting in nature because the LSA has no branches between its origin and the branching arterioles. Among the 41 reported cases, 19 were idiopathic (1, 3, 4, 6, 8, 11, 14, 15, 17, 20, 22-24, 28, 32, 34, 36), 12 associated with Moyamoya disease or similar vascular pattern (1, 9, 10, 12, 19, 21, 27, 31, 33), five with hypertension (7, 9, 23, 30, 35), two with systemic vasculitis (13, 16), and one each with arteriovenous malformation (29), ventricular neurocytoma (38), and substance abuse (9). Of the 24 idiopathic and hypertension cases, 14 were surgically managed, one obliterated with endovascular technique, one received focused radiation, and eight were observed. In the observed cases the vessel spontaneously healed and the aneurysm was no longer evident. This possibility makes a "wait-and-see" approach one option for treatment, thereby avoiding the risks associated with surgery or endovascular intervention. Follow-up angiography may be appropriate before one commits the patient to surgery or endovascular treatment, both of which may require sacrifice of the LSA, which would likely result in cerebral ischemia.

Arterial dissection may be related to mechanical and hemodynamic stress on the internal elastic lamina from turbulent blood flow within the artery, either transient or sustained. This results in a tear in the internal elastic lamina and the creation of a false lumen in the arterial wall, often causing rupture and hemorrhage or stroke in those who present (5, 40). In postmortem studies the dissection has been found to occur either between the internal elastic lamina and tunica media or between the tunica media and tunica adventitia. Formation of a trans-mural hematoma or thrombus between the intima and media may result in lumen occlusion and ischemic infarction. Hemorrhage between the tunica media and adventitia could result in aneurysmal dilatation potentially leading to vessel rupture with subarachnoid and/or intra-cerebral hemorrhage (25).

Recent advances in diagnostic imaging have increased detection of intracranial arterial dissection, including dissections of small penetrating vessels such as the LSA. Here we present an additional case of LSA dissection and review the variability of treatment across the literature. Furthermore, we speculate as to the relationship of this aneurysm to altered cerebrovascular

hemodynamics relative to repeated exposure to high altitude hypoxia.

CASE REPORT

A 50-year-old right-handed man presented with acute headache and left-sided facial weakness. The family noted gradual onset of left-sided facial droop while smiling, as well as mild behavioral change with increasing forgetfulness, over the 3 days before his presentation. During this interval his right-sided headache became increasingly severe. His symptoms started during coitus. The patient was an otherwise-healthy, athletic mountain climber with no known history of hypertension or other medical diseases. There was no history of allergy, smoking, drug use, or recent injury. His family history was unremarkable. His vital signs, including blood pressure, were all within normal limits.

On neurological examination, he was fully oriented with a mild left central facial weakness. The remainder of his neurological examination was normal. Axial brain computed tomography (CT) and CT angiography (Figure 1A), performed at the time of presentation, showed a right 3-cm anterior putaminal hemorrhage with intraventricular extension and a very small LSA aneurysm (arrow). Cerebral catheter angiography revealed a right LSA dissection and associated 3-mm pseudoaneurysm (arrow and inset) localized to the CT defined clot (Figure 1B). The patient was admitted for 10 days and was managed conservatively. Throughout the hospital stay, the patient remained clinically stable, and his facial weakness resolved.

Repeat digital subtraction angiography 10 days after his admission was performed with intention to treat but instead revealed spontaneous disappearance of the LSA pseudoaneurysm. He was discharged after 10 days. On his follow-up CT brain imaging 6 weeks after the onset of symptoms, the putaminal blood clot was resolved (Figure 1C). To confirm the permanent occlusion of the pseudoaneurysm, and to confirm fitness for mountain climbing, he underwent another digital subtraction angiography 6 months after initial presentation, which demonstrated complete resolution of the pseudoaneurysm consistent with arterial healing (Figure 1D). At follow-up, the patient noted only mild cognitive impairment. He was able to return to both work and mountain climbing, including a hike to Mt. Everest's base camp.

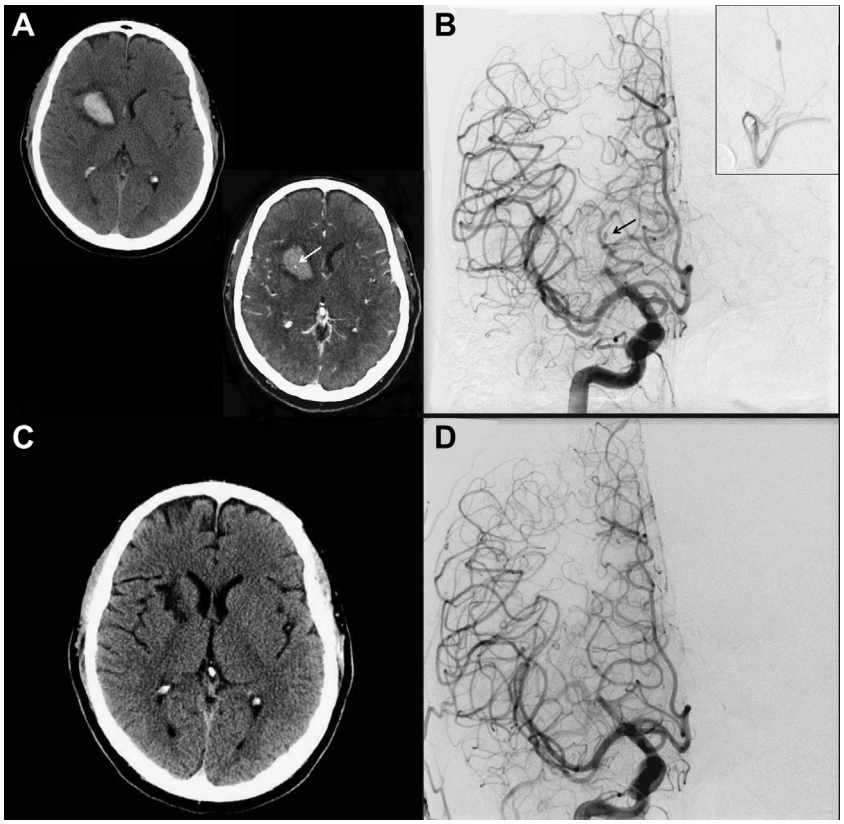

Figure 1. Axial CT and CT angiography (**A**) showing a right anterior putaminal hemorrhage with intraventricular extension and lenticulostriate pseudoaneurysm (*arrow*). Cerebral angiography revealed a right lenticulostriate arterial dissection and associated 3-mm pseudoaneurysm (*arrow and inset*) localized to the CT defined clot (**B**). Follow-up CT imaging at 6 weeks after the onset of symptoms (**C**) confirmed clot resolution. Repeat angiography at 10 days and at 6 months (**D**) demonstrated complete resolution of the pseudoaneurysm.

DISCUSSION

Lenticulostriate artery aneurysms are uncommon. Since 1960, only 41 cases of ruptured LSA aneurysm have been reported in the literature (Table 1). In 19 cases no identifiable underlying etiology was found, which include all five pediatric cases. The average age was 40 years (range, <1-71 years). This literature review showed a younger mean age of presentation, no female predominance, and greater rates of underlying vascular lesions in patients with LSA aneurysms compared with berry aneurysms. One third of reported cases were managed with observation only, 66% with either direct surgery or endovascular approaches, one with bypass surgery, and the remaining (5%) with various medical approaches (Table 2).

Turbulent blood flow is a natural part of the circulatory system and repetitive vessel injury and repair are normal (18, 25). Various extrinsic factors that have been attributed to intracranial arterial dissection include hypertension, disorders that increase hemodynamic stress such as arteriovenous malformation or Moyamoya disease, inflammation, autoimmune, and trauma. Intrinsic mechanisms such as a dysplastic vessel wall or altered repair mechanisms secondary to systemic disease also have been attributed to the etiology. Regardless of the type of dissection with formation of transmural and/or mural hematoma, spontaneous healing or recurrent dissection may occur. Controversy over treatment exists because while