Embolization of an Unruptured Distal Lenticulostriate Aneurysm Associated with Moyamoya Disease

SUMMARY: Lenticulostriate aneurysms are rare and usually present with intracranial hemorrhage, limiting understanding of their natural course. We describe an unusual case of an unruptured rapidly growing distal LSA aneurysm in the setting of Moyamoya disease, successfully embolized with n-BCA following functional neurologic testing with amobarbital.

A 35-year-old woman with Moyamoya disease and a history of 2 ruptured aneurysms was referred for follow-up. Physical examination demonstrated mildly decreased strength on the left, decreased bilateral upper quadrant peripheral vision and right-sided hearing loss, and slightly unsteady gait. Reflexes and sensory examination findings were normal. Mental status examination demonstrated normal higher cortical and language function. The findings of the remainder of her physical examination and medical history were unremarkable.

CT angiography demonstrated a 3-mm right distal LSA aneurysm and a Moyamoya vascular pattern, with numerous small-vessel collaterals supplying the MCA and ACA territories bilaterally. A right frontal pericallosal artery branch aneurysm clip was present. Conventional angiography 1 month later again demonstrated a right LSA aneurysm, now measuring \(4.2 \times 3.9 \times 3.8\) mm with a 1.8-mm neck, and collateral supply to a posterior frontal MCA branch. She was referred for endovascular treatment 10 days later.

A 6F Guider catheter (Boston Scientific, Fremont, California) was positioned in the distal right internal carotid artery, through which an Excelsior SL-10 microcatheter (Target Therapeutics/Boston Scientific, Fremont, California) was advanced over a Synchro-10 microguidewire (Boston Scientific) more distally in the same vessel and another 25 mg of Amytal was administered. No resultant deficits were observed. The lenticulostriate vessel and aneurysm were then successfully embolized by using n-BCA, eliminating both antegrade and retrograde filling of the aneurysm (Fig 1, 2).

Following improvement of left facial palsy, a Spinnaker Elite 0.15 microcatheter (Boston Scientific) was advanced over a Mirage microguidewire (ev3, Irvine, California) more distally in the same vessel and an additional 25 mg of Amytal was administered. No resultant deficits were observed. The lenticulostriate vessel and aneurysm were then successfully embolized by using n-BCA, eliminating both antegrade and retrograde filling of the aneurysm (Fig 1B).

The patient’s postoperative course was uneventful, and she was at preprocedural baseline neurologic status at discharge. At 1-month follow-up, she had no new neurologic symptoms and had begun running for exercise.

Discussion

A significant proportion of reported cases of LSA aneurysm are in patients with Moyamoya disease or Moyamoya-like vascular changes. These patients are significantly more susceptible to aneurysm formation compared with a baseline risk of 1%–6% in the general population; in a series of 81 patients with Moyamoya disease, 15% had cerebral aneurysms. Although most aneurysms in Moyamoya disease are in the circle of Willis (56%), a significant minority are found in the basal ganglia (18%) and collateral vessels (22%). Aneurysms are thought to arise in unusual branch vessels and collaterals in Moyamoya disease due to altered hemodynamics and the inherent vascular fragility. CT angiographic surveillance should, therefore, include not only the circle of Willis but the entire intracranial circulation. Although it has been suggested that most peripheral or collateral artery aneurysms in Moyamoya disease are pseudoaneurysms due to prior rupture of friable vessels, most reported cases of LSA aneurysms have undergone surgical treatment and have been confirmed as true aneurysms.

Intracranial hemorrhage in Moyamoya disease, a common manifestation in adults, is associated with a high incidence of rebleeding and significant morbidity. In addition, the outcome of aneurysm rupture is poor in patients with Moyamoya disease, with 39% of ruptured basal ganglia aneurysms and...
32% of collateral vessel aneurysms resulting in death or severe disability in 1 series of 111 patients. The average size of ruptured LSA aneurysms was 3.2 mm in 1 series of 6 patients and ranged from 2 to 5 mm in most reported cases in which size was reported. We observed approximately 40% growth of an unruptured LSA aneurysm during the course of 1 month, with a size within this range. Such rapid growth and historically small size at rupture underscores the need for urgent treatment of these aneurysms on diagnosis.

Most reported cases of LSA aneurysms have been treated surgically, often because anatomic considerations have rendered endovascular access difficult or impossible. However, improving microvascular techniques and technology, such as formable and torquable microcatheters and microguidewires, may enable more frequent endovascular treatment in the future. Although endovascular treatment may result in sacrifice of the parent LSA, a surgical approach may have the same end result, with the additional risks of damage to valuable collaterals and delayed injury to the parent LSA due to aneurysm clipping. In addition, the inherent difficulty in surgical treatment of these aneurysms is compounded in Moyamoya disease by vessel friability and hemodynamic fragility. An endovascular approach not only spares the patient a craniotomy and its attendant risks but also allows functional evaluation of the vascular territory via neurologic examination following amobarbital injection or test occlusion by catheter. This can be particularly useful in cases in which parent vessel occlusion might otherwise result in potentially devastating neurologic deficits.

In summary, distal LSA aneurysms can rapidly enlarge, commonly present with rupture, and are associated with significant morbidity and mortality. Vascular and hemodynamic fragility in Moyamoya disease complicates already difficult surgical treatment. Endovascular techniques can circumvent some inherent surgical risks and allow functional evaluation of the intended target vasculature via amobarbital or test occlusion before embolization. This is particularly useful in cases in which parent vessel occlusion might otherwise result in potentially devastating neurologic deficits.

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