Fusiform aneurysms of the lenticulostriate artery

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ABSTRACT

Lenticulostriate artery aneurysms are rare, can be difficult to diagnose, and when they rupture they are often associated with deep intraparenchymal hemorrhages. In particular, fusiform, dissecting aneurysms of a distal lenticulostriate artery are extremely rare. Typically, they are usually associated with underlying systemic conditions such as systemic lupus erythematosus, moyamoya disease, and substance abuse. Given their usual small size and location, these aneurysms may be difficult to detect with angiography and can be challenging to treat with either endovascular or microsurgical techniques. We provide background information, review the existing treatment experiences reported in the literature, and present a discussion regarding the optimal management using an illustrative clinical vignette. Parent artery obliteration can be a safe and effective treatment in these rare aneurysms.

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1. Introduction

Aneurysms of a lenticulostriate artery (LSA) distal to their take-off are uncommon and are usually associated with underlying systemic conditions including systemic lupus erythematosus, [1] moyamoya disease, [2,3] and substance abuse [4]. Furthermore, given their generally small size and location, these aneurysms may be difficult to detect angiographically and can be challenging to treat. Surgical access to the distal LSA territory and the basal ganglia can be fraught with complications, while selective endovascular access is often not possible due to the sharp angle of take-off of the LSA as well as the often delicate nature of these vessels due to concomitant neurovascular disease.

In particular, fusiform, dissecting aneurysms of the distal LSA are extremely rare. Herein, we review the literature on fusiform aneurysms of the distal LSA. The presentation of these aneurysms can be complex and can pose significant diagnostic challenges given their location and generally small size. Once presented with these lesions, there is significant controversy regarding their management. Endovascular therapy, microsurgery, and radiation have all been proposed. Furthermore, our understanding of the natural history of these lesions is limited and therefore observation is considered a reasonable possibility. In our discussion, we also present the identification and surgical management of a ruptured fusiform aneurysm of a distal LSA in a patient with no underlying vascular disease. This report contributes to the growing body of evidence that these lesions are prone to rupture but can be treated safely via surgery or endovascular therapy with good functional outcome. We aim to examine this specific clinical entity and discuss the critical importance of endovascular diagnosis and the safety of surgical management.

2. Presentation and diagnosis

LSA aneurysms are uncommon, with roughly 30 patients having been reported in the literature to our knowledge [1,3–36]. Ulm et al. have examined 100 middle cerebral artery (MCA) aneurysms and in their series discovered four LSA aneurysms, one of which presented with intracerebral hemorrhage [37]. Of note, however, their study specifically excluded aneurysms of the MCA with a fusiform morphology [37]. The prevalence of fusiform LSA aneurysms has not been studied in detail. These aneurysms are also challenging to diagnose since their presentation can often make vascular imaging more difficult. Reports have shown these lesions to present with tumor [18], intraparenchymal hemorrhage (presented patient and [7,11]), isolated intraventricular hemorrhage [27], and subarachnoid hemorrhage [26]. Imaging with computed tomography angiography (CTA) can miss these lesions. Spontaneous intracerebral hemorrhage, particularly in the basal ganglia or deep brain...
structures, is often associated with hypertension. Deep intracerebral hemorrhage in a young patient or a patient without underlying systemic vascular disease (including hypertension) necessitates a more thorough diagnostic work-up for potential etiologies of hemorrhage including aneurysms and arteriovenous malformations [38,39]. There is debate regarding the sensitivity of digital subtraction angiography (DSA) and CTA techniques in the detection of very small aneurysms [40,41]. Villablanca et al. published a case series demonstrating that CTA detects aneurysms <5 mm as well if not better than DSA, although others have argued that standard DSA remains the gold standard for detecting abnormalities, particularly with initial negative findings [40–42]. There is value in repeat angiography in any patient where there is concern for an underlying lesion with a distribution consistent with aneurysms of the LSA, particularly in those patients with risk factors for aneurysm formation (for example, moyamoya disease, fibromuscular dysplasia, inflammatory vasculopathies, or cocaine use).

To our knowledge, there are only 12 reports of fusiform distal LSA aneurysms in adults in the literature [7,18,26,29,31,33,34,36] (Table 1). We have reviewed the literature to emphasize only those fusiform aneurysms distal on the LSA as a separate clinical entity than all forms of LSA aneurysms. Their presentation and diagnosis are challenging. Their morphology and incorporation with the artery make them difficult to treat with preservation of the parent vessel. Finally, surgical or endovascular obliteration of the vessel seems not only possible, but also safe and effective in treating these lesions without producing significant morbidity.

3. Treatment

Non-surgical treatments for LSA aneurysms are varied from medical management [8,20] to endovascular strategies [11]. There has even been radiosurgical treatment of aneurysms such as by Lan et al. [34] Surgical options include direct (clip) obliteration of the aneurysm or parent vessel sacrifice to exclude the aneurysm from circulation. Distal fusiform aneurysms of the LSA, however, present a unique challenge. Endovascular access can be very challenging and surgical management often cannot provide a clear aneurysm neck to occlude. In both treatment paradigms, sacrifice of parent vessel has been the practice, not the exception. Three out of 12 patients underwent embolization [11,29,36] while only seven patients underwent proximal clipping of a LSA in the treatment of an aneurysm (Table 1). The majority of patients underwent microsurgical clip obliteration with proximal occlusion of the parent vessel. Narayan et al. reported success using intra-operative three dimensional angiography to clip a parent vessel in the treatment of a distal LSA saccular aneurysm in an otherwise healthy patient who displayed minimal residual deficits 6 weeks post-operatively [13]. Endo et al. clipped a parent vessel in a similar procedure on a child with a distal aneurysm of the LSA while Gandhi et al. described a series of three fusiform aneurysms in patients with underlying conditions that were treated with proximal parent vessel clipping [6,7]. Proximal fusiform aneurysms of the LSA were treated with proximal clipping and sacrifice of the parent vessel by Eddelman and colleagues in an adult and by Binning and colleagues in a child [4,22]. Kocher et al. used superselective angiography to delineate and attempt endovascular treatment of a distal fusiform LSA aneurysm. Because this lesion was not amenable to endovascular treatment based on anatomy, surgical exclusion by clipping was performed [26]. Kalani et al. also attempted an aggressive endovascular course but performed clip obliteration of the fusiform aneurysm due to vessel tortuosity [31].

Although Krieger et al. have reported that occlusion of perforating arteries can cause permanent infarction, Jabre and Simon have suggested an occlusion time of 5–10 minutes is acceptable and Eddelman et al. successfully occluded the parent vessel of a LSA aneurysm without neurological deficit [4,43,44]. In the future, intra-operative angiography prior to vessel clipping would be

<table>
<thead>
<tr>
<th>Study</th>
<th>Age/ Sex</th>
<th>Associated condition</th>
<th>Presentation</th>
<th>Location/ type</th>
<th>Size (mm)</th>
<th>Treatment</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Larrazabal et al. [11]</td>
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<td>4</td>
<td>n-BCA embolization</td>
<td>Persistent left hemiparesis</td>
</tr>
<tr>
<td>Gandhi et al. [7]</td>
<td>59/M</td>
<td>Moyamoya</td>
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<td>Right distal LSA</td>
<td>4</td>
<td>Proximal clipping</td>
<td>No residual deficit</td>
</tr>
<tr>
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<td>41/M</td>
<td>Cocaine use</td>
<td>Right insular IPH</td>
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<td>4</td>
<td>Proximal clipping</td>
<td>Mild headache</td>
</tr>
<tr>
<td>Gandhi et al. [7]</td>
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<td>Moyamoya</td>
<td>SAH</td>
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<td>Slight disability</td>
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<td>Kocher et al. [26]</td>
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<td>Diffuse SAH</td>
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<td>2</td>
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<td>Transient left facial weakness</td>
</tr>
<tr>
<td>Ellis et al. [27]</td>
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<td>Fibromuscular dysplasia</td>
<td>Isolated IVH</td>
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<td>4</td>
<td>Observation</td>
<td>No residual deficit</td>
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<tr>
<td>Tsai et al. [36]</td>
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<td>Left temporal IPI/VIH</td>
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<td>4</td>
<td>n-BCA embolization</td>
<td>No residual deficit</td>
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<tr>
<td>Lan et al. [34]</td>
<td>21/F</td>
<td>None</td>
<td>Right basal ganglia</td>
<td>Right distal LSA</td>
<td>5</td>
<td>Gamma Knife radiosurgerya</td>
<td>No residual deficit</td>
</tr>
<tr>
<td>Chalouhi et al. [29]</td>
<td>49/M</td>
<td>Moyamoya</td>
<td>Left basal ganglia IPH</td>
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<td>3</td>
<td>Onyx embolizationb</td>
<td>Persistent right hemiparesis No residual deficit</td>
</tr>
<tr>
<td>Kalani et al. [31]</td>
<td>66/F</td>
<td>Moyamoya</td>
<td>Incidental</td>
<td>Right distal LSA</td>
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<td>Proximal clipping</td>
<td>No residual deficit</td>
</tr>
<tr>
<td>Lama et al. [33]</td>
<td>50/M</td>
<td>High altitude exposure/ coitus</td>
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<td>Right basal ganglia IPI/VIH</td>
<td>3</td>
<td>Observation</td>
<td>No residual deficit</td>
</tr>
<tr>
<td>Present patient</td>
<td>41/F</td>
<td></td>
<td>Right basal ganglia IPI</td>
<td>Right distal LSA</td>
<td>3</td>
<td>Proximal clipping</td>
<td>Persistent left hemiparesis</td>
</tr>
</tbody>
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F = female, IPI = intraparenchymal hemorrhage, IVH = intraventricular hemorrhage, LSA = lenticulostriate artery, M = male, n-BCA = n-buty1 cyanocrylate, SAH = subarachnoid hemorrhage.

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useful in equivocal cases, as would newer techniques being used in aneurysm surgery such as angiographic roadmapping, near-infrared indocyanine green videoangiography, and frameless stereotaxy [45–48]. In patients with hematoma, there is also consideration of clot evacuation after clip obliteration. While Güresir et al. showed the benefit of clot evacuation after aneurysmal rupture and subarachnoid/intracerebral hemorrhage, there are limited data in delayed clot evacuation, particularly in this rare situation [49].

Fusiform aneurysms of the LSA are rare and are almost always associated with underlying condition predisposing one to develop vascular disease (Table 1). Vates et al. described a patient with a distal fusiform aneurysm secondary to a neighboring neurocytoma which was being fed by the parent LSA. This patient was treated with tumor resection, hematoma evacuation, and artery excision with persistent post-operative left hemiparesis [18]. Gandhi et al. reported three patients with fusiform aneurysms of LSA – two associated with a moyamoya-like vasculitis and the other associated with cocaine use [7]. All three patients tolerated their surgeries well and had mild to moderate post-operative disability (modified Rankin scale scores 1–2). While Eddleman et al. have demonstrated a proximal fusiform aneurysm of the LSA in an otherwise healthy adult, only Kochar et al. have reported a true distal fusiform aneurysm in an adult without any underlying predisposing factor who was treated with surgical clipping [4,26]. Binning et al. reported a 1 cm proximal fusiform aneurysm in an otherwise healthy child which had presented as an ischemic stroke, had thrombosed, and was treated with surgical clipping with good post-operative recovery of her hemiparesis [22].

4. Clinical vignette

A 41-year-old woman with no history of hypertension presented with acute onset of left hemiparesis primarily involving her left leg, along with acute mental status changes. Her medical history was notable for residual pain status post-lumbar laminectomy and fusion approximately 6 years prior. For her chronic pain syndrome after her laminectomy and fusion, she underwent placement of both a spinal cord stimulator and an intrathecal pump delivering analgesia consisting of the opioid sufentanil, the anesthetic bupivicaine, and the central α2-agonist clonidine. On the day of her presentation, she was seen for management of her intrathecal medications. Her neurological examination was notable for agitation, complaint of severe left-sided pain, and left-sided visual and tactile neglect. Cranial nerve exam demonstrated right supra-nuclear gaze deviation, left facial droop, and mild dysarthria. She had minimal antigravity movements of her left lower extremity and no movement of her left upper extremity. Her sensory examination was notable for diminished sensation to light touch of left side. Her National Institutes of Health Stroke Scale score was 16.

CT scans of the head without contrast demonstrated an acute 4.0 × 3.6 × 2.5 cm intracerebral hemorrhage involving the right insula, centrum semiovale, and superior right temporal lobe with evidence of mild right-to-left midline shift (Fig. 1A). Of note, on the day of admission, a CTA was also performed that demonstrated a focal outpouching of a right lenticulostriate artery adjacent to the hemorrhage (Fig. 2A, B). To clarify the nature of this outpouching, an initial cerebral angiogram was performed that did not identify a vascular etiology for the hemorrhage (Fig. 2C, D). Based on her CT scan evidence of a deep intracerebral hemorrhage, negative cerebral angiogram, and her neurologic condition, she was admitted to the intensive care unit.

Four days after presentation, the patient was noted to have an acute worsening of her headache and worsening left-sided hemineglect and weakness. Based on this, repeat imaging was obtained including a head CT scan that demonstrated slightly increased deep
intracerebral hemorrhage with new intraventricular hemorrhage and worsening midline shift (Fig. 1B). A repeat angiogram was performed and demonstrated a dissecting 3 mm fusiform aneurysm of a right lateral LSA located distally to the branch origin (Fig. 3). Given the newly identified vascular lesion, it was deemed that an intervention was required. Endovascular options were considered, such as embolization of the aneurysm with liquid embolic agents, but were not pursued due to the very small caliber of the affected LSA and the acute angle of take-off from the MCA. Therefore, the patient was taken to the operating room for surgical clipping of the proximal origin of the dissecting artery using intra-operative angiographic guidance. Initially, a left frontal ventriculostomy was placed. The opening pressure was normal. Cerebrospinal fluid was sent for appropriate laboratory analyses, and then the patient was placed in standard position for a right pterional craniotomy. The Sylvian fissure was widely split and the M1 and distal internal carotid were visualized to obtain proximal control. A number of normal appearing lenticulostriate perforating arteries were identified. Given the lack of a clear abnormal vessel and the inability to visualize the aneurysm, it was decided to occlude the vessel based on anatomic considerations. A temporary clip was used and an intra-operative angiogram was immediately obtained, which revealed continued filling of the lesion (Fig. 4A). The temporary clip was removed. A second more distal and adjacent LSA was then clipped with a temporary clip, and at this point there was no further angiographic filling of the lesion (Fig. 4B). The temporary clip was then replaced with a permanent mini clip. Due to the location of the bleed and the normal intracranial pressure, it was decided not to evacuate the hemorrhage. The patient tolerated the procedure well without complication.

On post-operative examination after extubation, the patient was awake, alert, and oriented to person, place, and time. She was following commands fully on her right side, but had minimal movement on her left side. She had continued dysarthria and dysphagia. Her neurological examination was unchanged from her pre-operative examination. Repeat angiogram was performed 3 days post-operatively that demonstrated mild right greater than left A1 and mid-basilar spasm, which was successfully treated during angiography. Work-up for vasculitis was negative. The patient was discharged to rehabilitation. On follow-up 1 year after presentation, the patient was able to ambulate with a cane and had moderate residual left-sided hemiparesis. The patient also had occasional seizures and was on anti-epileptic medications. Given the unusual nature of the vascular lesion, a follow-up angiogram showed the hematoma had been completely reabsorbed and demonstrated no residual aneurysm or any other vascular abnormality (Fig. 4C).

5. Discussion

Distal fusiform aneurysms of the LSA in an adult with no underlying or associated vascular condition are rare. While the patient presented had significant medical history for chronic pain after lumbar laminectomy, she was managed well with a spinal cord stimulator and intrathecal administration of analgesia. To our knowledge there has been no association between either the stimulator or the intrathecal medications and increased risk for vascular disease or aneurysm formation. The combination of clonidine, sufentanil, and bupivacaine are commonly used together for the treatment of severe neuraxial pain without significant adverse medication interactions, although mild side effects such as mild hypotension are common [45]. Clonidine acts as a central agonist at the α2-receptor in dorsal horn neurons, which results in pain control and a decrease in blood pressure. It serves as a potentiation agent for nociceptive pain when used in combination with fentanyl-like opioids [46] and is believed to be safe in a recent meta-analysis that showed improvement in pain control, including neuropathic pain, with only a slightly higher complication rate of mild hypotension [52–54]. It should be noted that another rare side effect of α2-receptor antagonists when combined specifically with morphine is a transient hyperalgesia, but this has not been shown to alter central nervous system hemodynamics or be associated with aneurysm formation [47,48]. Both sufentanil and bupivacaine are commonly used for epidural and intrathecal analgesia with no known predisposition to aneurysm formation or hypertension, which might lead to basal ganglia hemorrhage. On the day of presentation, our patient had received an increased dose of her intrathecal analgesia, but based on the present literature, there is no reason to associate her treatment for chronic neuropathic pain with the development of a dissecting fusiform aneurysm. In fact, the common side effect of mild hypotension would argue against a transient hypertension as even contributing to the aneurysm formation and rupture.

6. Conclusion

Aneurysms of the LSA are uncommon while fusiform aneurysms in this region are extremely rare. Here we present a report...
of a distal fusiform aneurysm in a patient with no associated vascular conditions. Repeated angiography is useful and necessary in patients who present with our patient's type of intracerebral hemorrhage and have no predisposing risk factors such as increased age or hypertension. Finally, while other treatment options are available, there are good operative outcomes with proximal clipping of parent vessels in LSA fusiform aneurysms.

Conflict of interest/disclosure

The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication.

References