CASE REPORT

Importance of repeat angiography in the diagnosis of iatrogenic anterior cerebral artery territory pseudoaneurysm following endoscopic sinus surgery

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SUMMARY

Iatrogenic intracranial pseudoaneurysm formation and rupture are rare complications following endoscopic sinus surgery. Given the propensity for devastating neurologic injury after a relatively routine procedure, swift diagnosis and treatment is essential. The authors present a patient who experienced bifrontal intracranial hemorrhage and subarachnoid hemorrhage due to a ruptured iatrogenic frontopolar artery pseudoaneurysm caused during routine endoscopic sinus surgery. The pseudoaneurysm was not present on initial angiograms but became apparent radiographically 19 days after the endoscopic procedure. Endovascular treatment consisted of coil and Onyx embolization for sacrifice of the parent vessel proximal to the lesion. Early recognition of iatrogenic intracranial vascular injury is important to allow for rapid treatment. Initial radiographic studies may be unreliable in excluding the presence of a pseudoaneurysm so delayed repeat angiographic assessment is necessary, particularly in the presence of a high index of clinical suspicion. Endovascular techniques may provide an effective and safe option for the treatment of iatrogenic anterior cerebral artery distribution pseudoaneurysms.

BACKGROUND

Iatrogenic arterial injury is a rare but potentially catastrophic complication of functional endoscopic sinus surgery. The most common and most devastating of these injuries is violation of the internal carotid artery.^{1,2} Rarely, cases of anterior cerebral artery (ACA) injury have been reported and are often associated with intracranial hemorrhage.³⁻⁵ We present a case of delayed diagnosis of a frontopolar artery pseudoaneurysm following endoscopic sinus surgery due to its obscurity by severe cerebral vasospasm and/or pseudoaneurysm thrombosis and recanalization. This case emphasizes the importance of persistent diagnostic investigation for a possible pseudoaneurysm in the setting of high clinical suspicion and vasospasm.

CASE PRESENTATION

A patient aged in the mid 60s was transferred to our institution with bifrontal intraparenchymal hemorrhage and basal frontal subarachnoid hemorrhage following endoscopic endonasal sinus surgery 10 days earlier (figure 1). Immediately after surgery at an outside institution the patient was noted to be abulic, lethargic, and to have left lower extremity weakness. A head CT scan and CT angiogram

obtained before transfer were concerning for the presence of a frontopolar artery pseudoaneurysm. The patient had undergone bilateral ethmoidectomies and middle meatal antrostomies for the treatment of chronic sinusitis refractory to medical therapy. According to operative notes from the outside institution, cerebrospinal fluid egress and pulsatile bleeding were noted high in the ethmoid cavity. This skull base violation was reportedly repaired easily using AlloDerm tissue matrix (LifeCell Corporation, Bridgewater, New Jersey, USA) and Gelfoam, followed by placement of Rhino Rocket nasal packing (Shippert Medical Technologies, Centennial, Colorado, USA).

Upon transfer to our facility, diagnostic cerebral angiography demonstrated severe cerebral vasospasm in the ACA distribution but failed to demonstrate any evidence of pseudoaneurysm (figure 2). Diagnostic cerebral angiography was performed daily and an intra-arterial verapamil infusion (dose range 20–40 mg into each internal carotid artery) was administered daily, with transient and gradual improvement of the vasospasm. On postoperative day 19, diagnostic angiography showed significant improvement in the degree of cerebral vasospasm and the presence of a small, inferiorly projecting frontopolar artery pseudoaneurysm (figure 3).

INTERVENTION

Because there was significant collateral blood flow to the distal frontopolar artery, we decided to sacrifice the artery proximally to prevent pseudoaneurysm filling. A Synchro-2 microwire (Stryker, Fremont, California, USA) and SL-10 2.4 F microcatheter (Penumbra, Alameda, California, USA) were advanced into the frontopolar artery. Galaxy coils (1.5 cm×2 mm; Codman, Raynham, Massachusetts, USA) were placed into the proximal part of the vessel after which Onyx 34 (ev3-Covidien, Irvine, California, USA) was injected to further occlude the artery.

OUTCOME

A post-treatment angiogram demonstrated occlusion of the proximal frontopolar artery with no filling of the pseudoaneurysm and robust retrograde filling of the distal segments of the frontopolar artery (figures 4 and 5). The patient tolerated all diagnostic procedures, intra-arterial vasospasm treatments, and vessel sacrifice without untoward events. Unfortunately, the patient had experienced

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Figure 1 Non-contrast CT images, axial views demonstrating subarachnoid hemorrhage filling the basal cisterns and bifrontal intraparenchymal hemorrhage.

only minimal improvement in language and motor functions at our last evaluation, 6 months after the procedure. Imaging studies performed at that time demonstrated expected evolution of bifrontal hemorrhagic infarcts.

DISCUSSION

Intracranial vascular injury is a rare but potentially devastating complication of endoscopic sinus surgery. The most dangerous and unfortunately most common of these injuries involves the internal carotid artery. Few reports in the literature have described cases of injury to vessels of the ACA distribution.^{3–5} However, as in our patient, most cases in the literature have resulted in frontal intraparenchymal hemorrhage. This characteristic pattern of hemorrhage should prompt suspicion for an ACA distribution pseudoaneurysm, even in the presence of negative initial radiographic studies.

Valle *et al*⁶ highlighted the importance of delayed angiographic evaluation, describing two patients with subarachnoid hemorrhage who initially had false-negative angiograms yet delayed angiograms demonstrated aneurysms. The authors attributed this phenomenon to aneurysm thrombosis and recanalization. Although this is one possibility for the initial false-negative angiograms in our patient, we also noted significant cerebral vasospasm, particularly in the ACA distribution.

The presence of cerebral vasospasm has been hypothesized to be a reason for initial false-negative cerebral angiograms. Kaim *et al*⁷ described eight patients with initial false-negative angiograms, four of whom were noted to have vasospasm. It is possible that the decreased caliber of the spastic parent vessel prevents filling of the aneurysm or pseudoaneurysm by causing apposition of the walls of the aneurysm neck. Therefore, with resolution of vasospasm, the ostium of the aneurysm may again open, resulting in filling.

The optimal time to perform repeat angiography when initial investigations are negative remains unclear. There is little evidence to support one time over another. Our approach has been very case-specific. In cases with low clinical suspicion, we perform angiography weekly until confident that vasospasm has resolved. When clinical suspicion is high, we often perform diagnostic angiography several times weekly, especially when cerebral vasospasm is noted. We feel that one cannot rest assured in the setting of a negative angiogram while vasospasm is present. In these cases, a patient diligent pursuit for underlying pathology is obligatory.

Maniglia⁵ reported one case of ACA laceration during endoscopic sinus surgery that resulted in death. Grigorian *et al*⁸ reported two cases in which pseudoaneurysms developed on the A2 segments of the ACA following endoscopic sinus surgery.

Figure 2 Digital subtraction angiogram performed upon arrival at our institution (postoperative day 10). Anteroposterior (left) and lateral (right) views demonstrate significant vasospasm of the anterior cerebral artery (arrows). No pseudoaneurysm is visualized.

Figure 3 Digital subtraction angiogram performed on postoperative day 19. Oblique anteroposterior (left) and oblique lateral (center) views demonstrate the presence of an inferiorly projecting pseudoaneurysm arising from the proximal frontopolar artery. The three-dimensional angiogram (right) confirmed the presence of a pseudoaneurysm. The arrow in each images shows the pseudoaneurysm.

Figure 4 Digital subtraction angiogram performed on postoperative day 19 immediately following treatment. Oblique anteroposterior (left) and oblique lateral (right) views demonstrate occlusion of the proximal frontopolar artery with no filling of the pseudoaneurysm.

However, unlike in our patient, massive arterial bleeding was encountered during these procedures. These patients, as well as our patient, were lethargic in the postoperative period. Therefore, an unexpected delay in emergence from anesthesia and/or lethargy in the postoperative period following endoscopic sinus surgery should prompt evaluation for intracranial

processes, including ACA pseudoaneurysm formation or rupture. It is important to note, as evidenced by the patients described in the report by Grigorian *et al*,⁸ that focal deficits may be absent, especially if the injury is isolated to the basal frontal lobe(s). We believe that our patient's focal deficit was the result of hemorrhagic infarction due to vasospasm rather than the primary intracranial insult.

Previous cases of iatrogenic ACA pseudoaneurysms reported in the literature were repaired with microsurgical clipping and vessel reconstruction. These modalities were considered and discussed among our team, as well as with the patient's family. Given the patient's compromised neurological status, a craniotomy with requisite administration of general anesthesia was better avoided. To our knowledge, this is the first case of an ACA territory pseudoaneurysm associated with endoscopic sinus surgery that was treated using endovascular techniques. Given the significant amount of vasospasm and previous iatrogenic vessel injury, we were concerned about the integrity of the vasculature and aneurysm wall to support traditional endovascular aneurysm occlusion (eg, primary coiling or stent-assisted coiling). Although it is possible that such treatment could have been successful, in this case we elected to sacrifice the frontopolar artery using coils and Onyx. Paramount to our decision to proceed with vessel sacrifice was our assessment of the collateral circulation, which was robust distal to the site of the lesion. Post-treatment angiographic injections demonstrated preservation of blood flow to the distal segments of the frontopolar artery through these collaterals.

ACA distribution pseudoaneurysm is a rare complication of endoscopic sinus surgery often resulting in basal frontal intraparenchymal and subarachnoid hemorrhage. This characteristic pattern of hemorrhage should prompt an unrelenting search for an ACA distribution pseudoaneurysm. In this setting,

Figure 5 Digital subtraction angiogram performed on postoperative day 19 immediately following treatment. Lateral view in the capillary phase demonstrates retrograde filling of the distal frontopolar artery branches.

a pseudoaneurysm should not be considered excluded on the basis of a single cerebral angiogram or in the presence of cerebral vasospasm. With adequate collateral circulation, endovascular parent vessel sacrifice is an effective and minimally invasive treatment option.

Learning points

- ▶ To our knowledge, this is the first report of an anterior cerebral artery territory pseudoaneurysm following endoscopic sinus surgery that was treated using endovascular techniques.
- ▶ The angiographic diagnosis of pseudoaneurysm was delayed because of obscurity by severe cerebral vasospasm and/or pseudoaneurysm thrombosis and recanalization.
- ▶ This case emphasizes the importance of persistent diagnostic investigation for a possible pseudoaneurysm in the setting of high clinical suspicion and vasospasm.

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the speakers' bureaus of Codman & Shurtleff and Genentech; serves on National Steering Committees for the following company-sponsored trials: 3D Separator (Penumbra), FRED (MicroVention), and SWIFT PRIME (Covidien); serves on advisory boards for Codman & Shurtleff and Covidien Neurovascular; and has received honoraria from Abbott Vascular and Codman & Shurtleff for training other neurointerventionists in carotid stenting and endovascular stenting for aneurysms. KVS receives research and consultant support from Boston Scientific, Cordis, EndoTex, EPI, Guidant, Medtronic, Micrus, Kerberos, Zimmer, Abbott Vascular, ev3, and Toshiba. He has financial/shareholder interests in EndoTex, Micrus, BSC EPI, Access Closure, Primus, Cordis, Boston Scientific, and Niagara Gorge Medical.

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