CASE REPORT

A novel approach to the management of carotid blowout syndrome: the use of thrombin in a case of failed covered stenting

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SUMMARY

Acute hemorrhage relating to an expanding pseudoaneurysm of the carotid artery is referred to as carotid blowout syndrome (CBS). CBS is associated with a high morbidity and mortality. We describe the case of a patient who presented with dysphagia and a pulsatile mass in the neck. Imaging revealed a pseudoaneurysm originating from the bifurcation of the distal right common carotid artery. On neuroangiography the patient lacked sufficient collaterals to allow for vessel sacrifice. A decision was made to use covered stents to prevent flow into the pseudoaneurysm while maintaining vessel patency. Despite placement of multiple covered stents there was residual slow filling of the pseudoaneurysm. We augmented this therapy with direct percutaneous thrombin injection into the pseudoaneurysm. This resulted in complete thrombosis of the pseudoaneurysm. For recalcitrant lesions in which the usual methods of stopping blood flow to the pseudoaneurysmal sac fail, an adjuvant approach with thrombin should be considered.

Impending CBS presents with an episode of transcervical or transoral hemorrhage that resolves spontaneously or with simple surgical packing. This differs from acute CBS in which patients present with profuse hemorrhage due to complete rupture of the artery that is not self-limiting.^{1–4} The traditional endovascular management of CBS has been occlusion of the bleeding vessel. In patients that cannot tolerate vessel sacrifice, the alternative is to use covered stents.

We describe a case of impending CBS where there was continued filling of the pseudoaneurysm sac (PAS) despite the stepwise use of three covered stents. Eventually we were able to stop flow into the PAS by injecting thrombin percutaneously under fluoroscopic and ultrasound guidance. In recalcitrant lesions where flow into the PAS does not stop despite the use of covered stents, an adjuvant approach with percutaneous injection of thrombin should be considered and has shown success in our case.

BACKGROUND

Acute hemorrhage or symptoms relating to an expanding pseudoaneurysm are referred to as carotid blowout syndrome (CBS), which is a medical emergency with a reported 60% neurologic morbidity and 40% mortality following open surgical intervention.¹ CBS can be subclassified as threatened, impending, acute, or recurrent. Threatened CBS relates to an artery that would invariably rupture if not promptly managed.

CASE PRESENTATION

An elderly patient with a past medical history significant for hypertension, coronary artery disease, abdominal aortic aneurysm, and a right carotid endarterectomy 3 years prior presented with dysphagia for 3 days with an expanding and painful pulsatile mass over the right neck. A CT angiogram of the neck showed a 6.8×6.1×4.5 cm pseudoaneurysm whose neck extended from the distal common carotid artery to the proximal right

Figure 1 (A) Axial CT angiogram of the neck showing contrast extravasation from the right common and internal carotid artery into the pseudoaneurysm. (B) Subtraction angiography showing filling of the pseudoaneurysm from the right common and internal carotid arteries. Clips are from a prior carotid endarterectomy (arrows).

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internal carotid artery (ICA) (figure 1). The decision was made to try and salvage the right ICA and not to occlude it as angiography demonstrated poor collateral circulation from the circle of Willis. Initially, microcoils (six in total) (Penumbra Inc) were placed into the external carotid artery until a good proximal plug was obtained to prevent endoleak into the pseudoaneurysm. A 6×5 cm covered stent (Gore Viabahn) was then placed extending from the mid right ICA to the common carotid artery. An angiogram was performed which showed residual filling of contrast.

Subsequently, a 5 mm balloon catheter was advanced into the covered stent and inflations were performed in the proximal and distal aspects of the stent to better oppose it. A repeat angiogram demonstrated reduced but residual filling into the PAS. At this point, an 8×10 cm stent (Gore Viabahn) was tunneled into the first stent and deployed. A repeat angiogram then demonstrated further reduction of contrast filling into the PAS, yet there was still delayed contrast pooling. An 8 mm balloon was advanced into the stent and inflated. A repeat angiogram then demonstrated very delayed but slow residual filling. The procedure was then terminated with the hope that the PAS would thrombose (figure 2).

Follow-up carotid Doppler revealed persistent flow into the PAS the next day (figure 3). The decision was made to attempt another intervention. On follow-up angiography, the PAS was smaller and measured 2.36×2.9 cm. There was still slow filling into the sac, and therefore another Viabahn stent (5×5 cm) was placed into the ICA overlapping the previously placed stents, extending the coverage distally. Using a 5 mm balloon, angioplasty was preformed to fully oppose the stent against the wall

Figure 2 The angiogram, despite placement of two covered stents, demonstrates residual filling of the pseudoaneurysm sac. Penumbra microcoils (arrow) in the external carotid artery were placed prior to stent graft to prevent endoleak.

Figure 3 Color flow Doppler showing persistent flow into the pseudoaneurysm sac on day 2 post procedure.

of the vessel. Despite these efforts, there was still residual filling of the PAS (figure 4).

The decision was then made to attempt a novel approach of treatment. Under ultrasound guidance, a 22 gauge spinal needle was advanced into the pseudoaneurysmal sac. We removed the inner stylette of the spinal needle and observed return of pulsatile blood flow. We then injected a small amount of contrast under fluoroscopic guidance through the spinal needle, and we opacified the PAS (figure 5A). A balloon was then inflated within the right ICA. Pooled human thrombin (250 units) was directly injected into the PAS under ultrasound visualization (figure 5B). The balloon was deflated after 2 min and a repeat angiogram showed significantly reduced flow into the PAS. Yet, there was still a small amount of residual filling at the pseudoaneurysm neck. The needle was then redirected near the neck of the PAS and the above technique was repeated using another

Figure 4 Image showing placement of the third stent and balloon inflation.

Figure 5 (A) A spinal needle is inserted into the pseudoaneurysm sac and placement is confirmed with contrast opacification (arrow). (B) A spinal needle is advanced into the pseudoaneurysm sac under fluoroscopic and ultrasound guidance (probe on the left), with a balloon inflated into the internal carotid artery to prevent recirculation of thrombin.

250 units of thrombin. Repeat angiogram showed complete occlusion of the PAS with no filling in the early or late phase noted (figure 6). The patient had no neurological deficits post-operatively and a follow-up Doppler demonstrated no flow in the PAS.

OUTCOME AND FOLLOW-UP

Although the procedure was a technical success and the patient had no neurological deficits post procedure, the patient ultimately developed complications of septic shock. The patient had a prolonged stay in the intensive care unit and died from infectious causes not related to his CBS.

DISCUSSION

CBS can be a catastrophic event and is associated with high morbidity and mortality. Surgical intervention has fallen out of favor because of its high associated mortality. Endovascular management for CBS allows for both deconstructive and reconstructive techniques.^{4 5} The addition of balloon occlusion testing and angiographic evaluation of distal collaterals can lead to better outcomes.

The use of deconstructive techniques (ie, vessel sacrifice) such as balloons, coils, liquid embolic agents, or occluder plug devices has been fairly well described. Yet these techniques should not be used in patients with limited distal collaterals.

Figure 6 After graded stepwise percutaneous injection of thrombin into the pseudoaneurysm sac (PAS), no flow into the PAS is noted on carotid Doppler (A) and CT angiogram (B). ICA, internal carotid artery.

Even in those patients with good collaterals, deconstructive techniques have a 3–5% risk of stroke.⁶ Reconstructive endovascular methods, with the use of endovascular stent grafts, have been described. The benefits of stent grafts are obvious in that they allow continued perfusion to the brain while excluding the pseudoaneurysm. Complications from stent grafts include infection and spontaneous thrombosis which can both lead to stroke. Vessel re-rupture proximal or distal to the stent may still occur.

In the case of our patient, given the poor collateral circulation pattern on angiography, the patient would not have tolerated vessel sacrifice. We therefore decided on the use of covered stents to bypass flow from the neck of the pseudoaneurysm. Despite the use of multiple covered stents, flow persisted into the pseudoaneurysm. Given that the covered stents failed, we decided on an adjunctive approach of direct thrombin injection into the sac of the pseudoaneurysm.

Thrombin is commonly used in the endovascular management of peripheral pseudoaneurysms. Generally, the pseudoaneurysms treated with thrombin have a narrow neck. Potential complications include arterial thrombosis, hypotension, and bradycardia from acquired coagulation factor inhibitors. However, these complications can be minimized by injecting thrombin under ultrasound guidance directly into the pseudoaneurysm cavity while compressing the tract. Furthermore, cross reactivity is generally attributed to bovine thrombin, and can be minimized if thrombin pooled from human plasma is used.⁷

We found one prior case documenting the use of percutaneous thrombin injection for the treatment of a carotid artery pseudoaneurysm. Holder *et al*⁸ treated the aneurysm solely by percutaneous injection of thrombin, without the prior use of covered stent grafts. In their case, this approach was successful. However, the risk of thrombin recirculating into the cerebral circulation can be catastrophic.⁹ Only after the failed use of three covered stents did we consider the use of percutaneous thrombin. In order to reduce the possibility of recirculating thrombin we used low dose thrombin. It was injected after inflating a balloon across the neck of the pseudoaneurysm within the stent graft to further reduce any chance of thrombin recirculation.

With appropriate caution, success can be achieved in difficult cases with the use of thrombin and simultaneous balloon occlusion, and should be considered as an adjuvant approach in the management of CBS.

Learning points

- ▶ Acute hemorrhage or symptoms related to an expanding pseudoaneurysm are referred to as carotid blowout syndrome.
- ▶ Surgical approaches were previously the gold standard of therapy; however, endovascular approaches with covered stents are now the norm in the treatment of aneurysms.
- ▶ We demonstrated through our report that in the case of failed stenting for recalcitrant lesions, adjuvant approaches must be sought, especially in cases where the aneurysm is at high risk of rupture.
- ▶ We were successfully able to inject thrombin percutaneously into the pseudoaneurysm sac to occlude flow into the sac, without any evidence of leakage of thrombin into the cerebral or systemic circulation.

Contributors All authors contributed equally in writing the manuscript. KA and JF were the physicians that managed the case and carried out the procedure.

Competing interests None declared.

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