Clipping of a Mycotic Basilar Trunk Aneurysm under Cardiac Arrest in a Pregnant AIDS Patient

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ABSTRACT

We present the first case of a coccidioidomycosis mycotic anterior inferior cerebellar artery (AICA) aneurysm that was clipped under hypothermic cardiac standstill in a pregnant acquired immunodeficiency syndrome (AIDS) patient. A 24-year-old pregnant AIDS patient presented with intraventricular hemorrhage and hydrocephalus. Angiography revealed an 8-mm basilar trunk aneurysm with the right AICA protruding from the side wall of the aneurysm. The patient underwent a retrosigmoid craniotomy and direct clipping of the aneurysm under hypothermic cardiac standstill. At presentation, the patient had a poor grade due to subarachnoid and intraventricular hemorrhage. Despite her large posterior circulation aneurysm in the setting of AIDS with extensive coccidioidomycosis meningitis, the lesion was clipped successfully. To do so required the full range of neurosurgical repertoire, including a skull base approach and hypothermic cardiac standstill.

KEYWORDS: Coccidioidomycosis meningitis, hypothermic cardiac standstill, mycotic aneurysm

We report the first surgical clipping of a large mycotic aneurysm related to coccidioidomycosis meningitis in a patient with autoimmune deficiency syndrome (AIDS). The aneurysm, which involved the basilar trunk and anterior inferior cerebellar artery (AICA), required hypothermic cardiac standstill to clip. Coccidioidomycosis infection is caused by the fungus *Coccidioides immitis*. Patients acquire coccidioidomycosis by inhalation of the spores in areas such as Arizona, where this patient lived. Infections can be serious in patients with an immunocompromised state such as our patient. We discuss the complexities of therapeutic planning for this patient.

CASE REPORT

A 24-year-old woman was taken to an outside hospital by emergency medical personnel after she was found unconscious on the bathroom floor by her husband. On arrival at the hospital, the patient was somnolent but able to move all extremities. Computed tomography (CT) scan of the head showed intraventricular hemorrhage associated with mild ventriculomegaly. A ventriculostomy was performed before cerebral angiography. Angiographic injection of the left vertebral artery showed an ~8-mm anteriorly directed basilar trunk aneurysm at the level of the AICAs (Fig. 1A and B). The right AICA originated from the inferolateral aspect of the aneurysmal wall. A

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Figure 1 (A) Anteroposterior and (B) lateral angiographic views of the head. The left vertebral artery injection shows an 8-mm, anteriorly directed basilar trunk aneurysm at the level of the AICA. (Used with permission from Barrow Neurological Institute.)

focal area of stenosis was present in the basilar artery just proximal to the aneurysm.

The patient had a complicated medical history. Seven years before this event at the age of 16, she was diagnosed with human immunodeficiency virus (HIV). At presentation, the patient's CD4 count was $17\mu^3$. She was on lopinavir/ritonavir and lamivudine/zidovudine. She was also on empiric antibiotic prophylaxis with trimethoprim/sulfamethoxazole and aztreonam. Four years later she was diagnosed with AIDS. Her opportunistic infection, coccidioidomycosis meningitis, proved to be refractory to treatment. She failed treatment with fluconazole and voriconazole. At the time of her intracranial hemorrhage, she was on salvage therapy with posaconazole and amphotericin B.

Her hematocrit was 23 and creatinine was 1.5 mg/dL. The anemia was attributed to the AIDS and lamivudine/zidovudine therapy. Renal insufficiency was thought to be related to amphotericin B therapy. Serum human chorionic gonadotropin was positive for pregnancy. Transvaginal ultrasonography showed a viable 13-week fetus.

At cerebral angiography, the basilar trunk aneurysm was interpreted as a dissecting mycotic aneurysm. The right AICA, which arose from the side wall of the aneurysm, appeared to be in significant jeopardy if coiling were attempted. The decision was thus made to treat the aneurysm surgically.

On posthemorrhage day 2, the patient underwent a right retrosigmoid craniotomy. Once the thickened arachnoid was opened, we encountered numerous nodules of thickened purulent material. The avenue of approach was between the seventh and eighth cranial nerve complex and the lower cranial nerves. A large thin-walled aneurysm was visualized at the basilar

trunk. During dissection of the aneurysm at the neck, we caused a small tear that was easily controlled with a piece of Surgicel (Ethicon, Somerville, NJ) and mild compression. At this point, because of the friable appearance of this mycotic aneurysm and because of our intraoperative assessment that we lacked adequate proximal control, we decided to perform the remainder of the aneurysm dissection under hypothermic cardiac standstill.

The patient was fully heparinized. The cardiothoracic surgery team cannulated the femoral artery and vein to prepare for cardiopulmonary bypass. The patient's core temperature was decreased to 18°C . Once the patient's body temperature reached 18°C , the cardiopulmonary bypass was stopped and the blood was drained from the venous side into the reservoir. Immediately, the intravascular space, including in the basilar artery, was decompressed. We then resumed dissection of the aneurysm wall, which was significantly deflated. The aneurysm was clipped successfully with a 7-mm slightly curved clip and a 9-mm straight clip. On visual inspection the right AICA appeared to be kinked but patent. However there was inadequate space to confirm patency by Doppler ultrasonography or indocyanine green angiography.

The patient was under cardiac standstill for 6 minutes and 23 seconds. Blood flow was then gradually increased to full capacity while the patient was rewarmed simultaneously. Once the mixed venous temperature reached 36°C , the cardiopulmonary bypass was slowly weaned. Once the cannulas were removed, the patient was transfused with fresh-frozen plasma and platelets. The heparin was reversed with 30 g of intravenous protamine. After meticulous hemostasis was obtained, the craniotomy was closed. Per infectious

Figure 2 (A) Anteroposterior and (B) lateral angiographic views the head. The left vertebral artery injection shows no residual aneurysm. The basilar trunk is mildly narrowed, but the right AICA is occluded completely. (Used with permission from Barrow Neurological Institute.)

disease recommendation, an Ommaya reservoir was left in the posterior fossa with the tip of the catheter in the cisternal magna before closure.

Postoperative cerebral angiography showed no residual aneurysm. The basilar trunk was mildly narrowed, but the right AICA was occluded completely (Fig. 2A and B).

After surgery, the patient moved all extremities spontaneously but did not follow commands. Immediately after surgery, transvaginal ultrasonography confirmed viability of the fetus. The patient, however, underwent a spontaneous abortion on postoperative day 6. She spent 10 days in the intensive care unit. During her hospitalization, the patient's neurological status improved progressively.

On postoperative day 19, the patient was discharged home under 24-hour family supervision with home physical and occupation therapy. At her last follow-up 7 months after surgery, the patient was neurologically intact. Her only complaint was of intermittent headaches. She was on intrathecal amphotericin B twice weekly as well as voriconazole, efavirenz, and lamivudine/zidovudine. Despite this regimen, the patient's coccidioidomycosis meningitis has proven extremely refractory to treatment. Per infectious disease recommendations, she is currently scheduled to undergo placement of an intrathecal pump for continuous infusion of intrathecal amphotericin B.

DISCUSSION

We report the case of a large mycotic AICA aneurysm treated via a retrosigmoid approach. Previously, we have reported our cumulative experience with the surgical

treatment of AICA aneurysms.¹ Thirty were proximally located. Twenty-one aneurysms had ruptured. Surgical approaches included the retrosigmoid, far-lateral, and orbitozygomatic. Eighteen patients (56%) had neurological complications. A mean of 31 months of follow-up was available in slightly more than half of the patients. Patients' clinical outcome at discharge was not significantly different from their status at their initial assessment. Intraoperative hypothermic cardiac standstill was used to clip eight giant aneurysms.

In this case, we made preoperative arrangements for possible hypothermic cardiac arrest. However, the decision to proceed with this modality was made intraoperatively once we determined that proximal control was inadequate and that clip reconstruction would require more manipulation than the tense, friable aneurysm could withstand. Under cardiac standstill, it was much easier to manipulate the aneurysm without fear of rupture. The collapsed aneurysm significantly improved visualization of the surrounding anatomy and increased the working space.

Hypothermic cardiac arrest allows collapse of the aneurysm, increases working space, and improves visualization of the anatomy.^{2,3} These advantages provide a bloodless surgical field and cerebral protection, both of which make precise clipping of giant or large aneurysms possible. The disadvantages of this technique include relatively inefficient and uneven cooling rewarming, cardiac congestion and arrhythmia, severe physiological changes, and coagulopathy.⁴ Even after the heparin has been reversed, oozing related to coagulopathy can be significant, thereby increasing the chance of bleeding complications. Even with deep hypothermia and barbiturate coma, there is always a chance of strokes from

thromboembolism or hypoperfusion. Furthermore, hypothermic cardiac arrest increases the risk of spontaneous abortion and preterm delivery in pregnant patients.⁵ Indeed, our patient did have a miscarriage, several days after surgery.

Endovascular coiling with or without stent placement is definitely an option for the treatment of such proximal AICA aneurysms. There are several reports of good outcomes after endovascular occlusion of such lesions.^{6–9} Pandey et al reported their extensive, 10-year experience with endovascular treatment of posterior circulation aneurysms.¹⁰ Of their 275 aneurysms, 23 were at the intersection of the basilar trunk and AICA. Overall, they successfully embolized 88% of their cases. Their mean angiographic follow-up was 31 months for procedures performed before 2004 and 14 months for procedures performed during 2004 and 2005. During this latter period, 24.5% of aneurysms recanalized at least 5%. Retreatment was required in 4.9%, and rehemorrhage occurred in 1.1% of patients.

Our case was particularly challenging because not only was the lesion a large basilar trunk/AICA aneurysm, which is associated with less than optimal visualization and proximal control during surgery, but it was also mycotic. Patients with AIDS are prone to opportunistic infections. The presence of a cerebral aneurysm in an AIDS patient naturally raises the possibility of its being mycotic. Moreover, the HIV itself can cause arteriopathy with subsequent aneurysm formation in the absence of opportunistic infections.^{11,12} A handful of AIDS-related mycotic cerebral aneurysms have been reported. Most were secondary to aspergillosis, a known angioinvasive fungal pathogen with a propensity for involving the central nervous system.^{13–18} This is the first report of a mycotic aneurysm related to coccidioidomycosis meningitis. The aneurysm was proximal, as is often the case in the setting of meningitis. In contrast, aneurysms associated with endocarditis are usually in the distal distribution of the internal carotid artery.¹⁹ Aneurysms related to meningitis are associated with a relatively worse prognosis. The aneurysm in our patient involved the vertebrobasilar artery and was associated with a fungal infection, which is also a predictor of a poor outcome.^{14,19–21}

Although surgery has been the mainstay of treatment for ruptured mycotic aneurysms, endovascular therapy is a viable option. Most patients with ruptured mycotic aneurysms are in moribund condition, making them attractive candidates for endovascular therapy. Early, there were concerns about the safety and efficacy of endovascular treatment of such thin-walled and friable aneurysms. With the rapid evolution of endovascular technology and techniques, however, there has been a flurry of recent reports of successful coiling of mycotic aneurysms.^{22–28} Neuro-

form stents have been placed alone successfully in mycotic proximal aneurysms that enlarged after treatment with intravenous antibiotics.²⁹ The mechanism of action is presumably flow redirection. There are also reports of successful endovascular vessel sacrifice for the treatment of mycotic aneurysms in patients who passed a balloon occlusion test.³⁰ In our patient, however, we believed that the risk profile favored surgery and that surgery would give us the best chance at clip reconstruction of the basilar trunk while sparing the right AICA.

Our patient's postoperative angiogram showed that the right AICA was actually occluded. It is possible that endovascular coil embolization of the aneurysm would have produced the same outcome: occlusion of the aneurysm and the right AICA, which originated from its wall. Fortunately, the patient did not suffer a stroke, presumably because her collateral flow was adequate.

Hypothermic cardiac arrest has enjoyed utility in the past for the clipping of giant and otherwise untreatable aneurysms. Given the very poor prognosis of untreated giant aneurysms, the high morbidity and mortality rates associated with hypothermic cardiac arrest seemed acceptable. In current neurosurgical practice, however, we have at our disposal a new array of state-of-the-art minimally invasive treatments associated with very low morbidity rates. Advances in endovascular neurosurgery have allowed us to perform successful embolization of giant aneurysms, from stent-assisted coiling to balloon-assisted Onyx HD500 (eV3 Corporation, Irvine, CA) embolization. Because these new options can be exercised with few complications, the use of hypothermic cardiac arrest has been in decline and will continue to do so.

CONCLUSION

Ruptured mycotic aneurysms in the posterior circulation present a formidable challenge for vascular and endovascular neurosurgeons. Our patient presented with a poor grade from subarachnoid and intraventricular hemorrhage. She had a large posterior circulation mycotic aneurysm in the setting of AIDS and extensive coccidioidomycosis meningitis. Despite these negative prognosticators, the patient's aneurysm was clipped successfully. To do so required the full range of neurosurgical repertoire, including a skull base approach and hypothermic cardiac standstill.

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