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Case Report

A rare case of *Streptococcus anginosus* infectious intracranial aneurysm: Proper management of a poor prognosis

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ABSTRACT

Background: Infectious intracranial aneurysms (IIAs), sometimes referred to as cerebral mycotic aneurysms, are an uncommon but feared compilation of bacterial endocarditis, occurring in up to 5% of all bacterial endocarditis cases. While IIAs carry a low risk of rupture, a ruptured mycotic aneurysm carries devastating neurologic consequences with up to an 80% mortality rate secondary to subarachnoid and intracerebral hemorrhage.

Case Description: A 69-year-old man undergoing antibacterial therapy for *Streptococcus anginosus* endocarditis with aortic insufficiency and root abscess presented to the ED with multiple seizures and left-sided weakness. MRI of the head revealed right frontal and temporal abscesses with evidence of scattered septic emboli and subarachnoid hemorrhage. CTA of the head revealed a ruptured 1 mm distal middle cerebral artery mycotic aneurysm. Prior to undergoing surgery, the patient began to decline, becoming lethargic, and failing to respond to commands. The patient underwent endovascular Onyx embolization. After the procedure, the patient remained with partial status epilepticus and was discharged to rehabilitation. Over the following months, the patient made a great recovery and was able to undergo aortic and mitral valve replacement 5 months after neurosurgical intervention.

Conclusion: This favorable outcome is the result of a tremendous deal of long-term coordination and efficient communication between neurosurgery, cardiology, neurology, physical medicine and rehabilitation, and primary care.

Keywords: Endovascular, Infectious intracranial aneurysm, Mycotic aneurysm, Subarachnoid hemorrhage

INTRODUCTION

As with most major cerebrovascular events, rupture of an infectious intracranial aneurysm (IIA) carries a relatively poor prognosis with a high morbidity and mortality rate.[6] This case will explore the long-term management of such an event and highlight the need for longitudinal input of several specialties.

IIAs are an uncommon but feared phenomena most commonly associated with bacterial endocarditis with an incidence of up to 5% in all bacterial endocarditis cases.[13] Although IIAs carry a low risk of rupture, such an event carries devastating neurologic consequences with an

estimated 80% mortality rate secondary to subarachnoid and intracerebral hemorrhage.[4]

Here, we describe a case of bacterial endocarditis complicated by an IIA of rare etiology with a good outcome that required extensive interdisciplinary long-term care and follow-up. The case required months of medical management, two separate surgical interventions, and hours of physical rehabilitation to obtain a favorable outcome. This case highlights the importance of effective interdepartmental communication in achieving optimal care.

CASE REPORT

Presentation

A 69-year-old male with known hypertension and poor dentition presented to the hospital with 5 months of dry cough, night sweats, and weight loss. Physical exam revealed a new 2/6 diastolic murmur. Investigative TEE showed a left aortic perivalvular abscess with vegetations and severe aortic insufficiency. The diagnosis of native valve subacute endocarditis was made, with blood cultures positive for *Streptococcus anginosus*. The patient was discharged on a 1-month course of IV Ceftriaxone and scheduled for a minimally invasive aortic valve replacement with possible aortic root reconstruction on completion of antibiotic therapy. On day 26/28 of antibiotic therapy, the patient presented to his local emergency room after having experienced a first-time seizure and loss of consciousness. He had no personal history or known family history of seizures or seizure disorders.

On admission, physical exam showed elevated blood pressure at 151/61 with a regular heart rate of 68, a faint diastolic murmur at the base, and obvious favoring of the right upper and lower extremities without pronator drift. Physical exam was otherwise unremarkable. The patient was given Levetiracetam 1000 mg and Dexamethasone 6 mg for seizure management.

Investigations

At the patient's local ED, initial contrast CT of the head showed a 1.5 cm rim enhancing lesion with no reported hydrocephalus. He was transferred to our center for further care. On the morning following transfer, the patient suffered a second left-sided seizure at which point an MRI with and without contrast was performed. The MRI revealed a 1.4 × 1.4 × 1.6 cm rim-enhancing lesion in the right middle frontal gyrus with surrounding vasogenic edema suggestive of a pyogenic abscess [Figure 1a]. Further, both cerebral hemispheres showed focal areas of abnormal leptomeningeal and patchy cortical enhancement suggestive of early cerebritis.

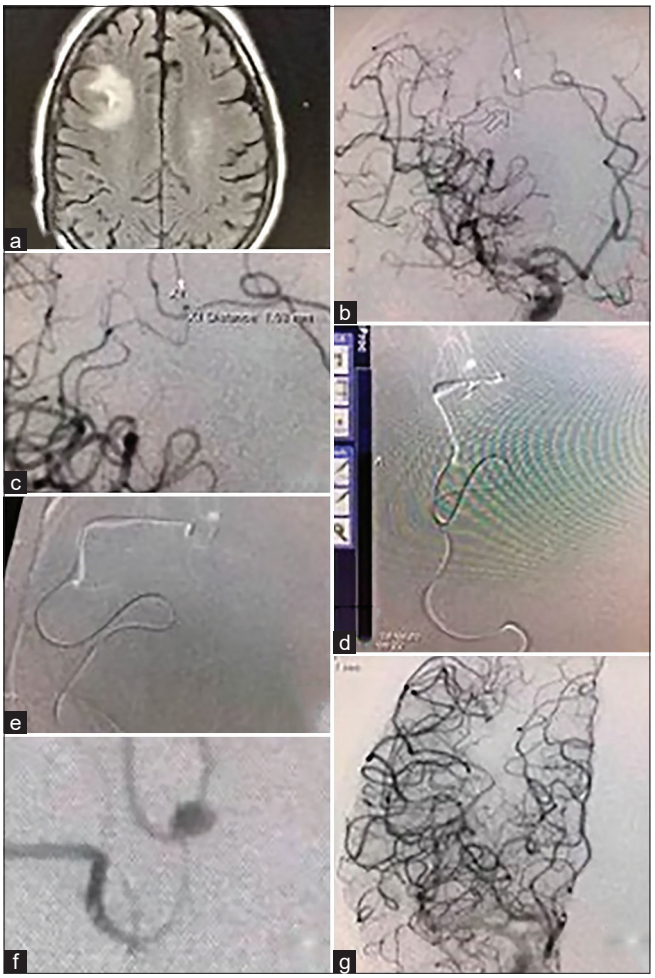

Figure 1: (a) MRI Flair sequence shows right frontal lesion with surrounding edema and subarachnoid hemorrhage consistent with cerebral abscess and ruptured mycotic aneurysm. (b and c) Digital subtraction angiography anterior-posterior and oblique views shows mycotic aneurysm measuring 1 mm. (d and e) show roadmap angiography with microcatheter approach to the aneurysm. (f) Microcatheter injection just proximal to the mycotic aneurysm. (g) Internal carotid artery digital subtraction angiography after onyx embolization of the mycotic aneurysm with no residual filling and no unintended thromboembolic phenomena.

Treatment

After visualization of the suspected abscess on MRI [Figure 1a], a repeat CTA was performed revealing a 1 mm aneurysmal dilation in the M4 frontal branch of the right distal middle cerebral artery (MCA) [Figure 1b and c]. Imaging was highly suggestive of a mycotic aneurysmal rupture with bleed and active inflammation. A cerebral angiogram with embolization was scheduled and performed shortly thereafter. Such cases are typically treated with surgical ligation and removal but due to this patient's poor condition, a minimally invasive endovascular approach was favored.

Vascular access was first gained through the right femoral artery. An Envoy guide catheter was advanced to the right internal carotid artery through roadmap assistance at which point a 3D angiogram was taken to better visualize the right MCA. Due to the tortuosity of the patient's right iliac system, the guide catheter was unable to be advanced any further. Instead, left femoral artery access was gained and the vascular catheter was advanced in a similar fashion. The guide catheter was advanced to the M2 branch of the right MCA where it was exchanged for an intermediate catheter over a microcatheter over a Synchro microwire. A superselective M3 and M4 angiogram were then performed, allowing for visualization of the 1 mm infectious aneurysm in the cortical M4 frontal MCA branch [Figure 1d-f]. On visualizing the feeding vessels, Onyx 18 material was advanced through the Synchro microwire into the M4 pedicle, filling the aneurysm [Figure 1g]. The Envoy catheter was removed and 3-mm J-wires were advanced into the vascular sheaths. The patient remained stable for the duration of the 1-h procedure and was then transported to the neurointensive care unit (NICU) as planned.

One day following surgery, the patient developed seizures with partial status epilepticus. CT of the brain showed a hyperdensity in the subarachnoid space with subtle ventriculomegaly due to rupture of the IIA with subarachnoid hemorrhage (Modified Fisher Scale grade 2/Hunt and Hess grade 3) status post embolization. The patient was closely monitored for deterioration of symptoms. Two days later, follow-up CT of the brain revealed stable subarachnoid and intraventricular hemorrhage with no hydrocephalus. It was ultimately decided no external ventricular drain would be placed given the lack of hydrocephalus and stability of bleed. The patient remained in the NICU for 22 days where he was placed on Levetiracetam, Clobazam, and Lacosamide for seizure control. During this time, the patient was followed closely by neurology with daily electroencephalograms and by pulmonary/critical care as he remained intubated on mechanical ventilation post procedure. Infectious disease also followed the patient for infective endocarditis and later began triple antibiotic treatment with Vancomycin, Cefepime, and Metronidazole for ventilator associated pneumonia and right middle frontal gyrus abscess.

On leaving the NICU, the patient was evaluated by physical medicine and rehabilitation who initiated aggressive physical therapy, occupational therapy, and speech therapy. The patient was discharged to a hospital-adjacent rehabilitation center 1 week later where he continued to undergo physical, occupational, and speech therapy for 1 month.

Four months after the patient's initial hospital discharge, he was deemed a suitable candidate to undergo cardiothoracic surgery. The patient underwent an aortic and mitral valve replacement without complication and was discharged home after 4 days of hospital care.

Outcome and follow-up

For 6 months, the patient remained out of the hospital with mild lifestyle modifications and changes to mood (including some confusion) which improved on cessation of steroid therapy.

The patient was readmitted to the hospital 6 months after cardiac surgery when he suffered a left sided seizure with associated Todd's paralysis attributed to possible nonadherence. During the admission, he suffered a pulmonary embolism for which an IVC filter was placed. He was discharged home again with daily speech, occupational, and physical therapy. Since the episode, the patient had no major hospital admission and has been adherent to his antiseizure regimen of Levetiracetam, Lacosamide, Topiramate, and Clobazam.

At present, the patient is 1½ years status post initial rupture of the IIA. The patient is currently alive, and his mood has significantly improved per both his family and healthcare team. He has suffered several minor seizures in the interim due to medication titrations. However, he has reached the point where he is again confident enough and possibly eligible to begin driving school.

DISCUSSION

IIAs, sometimes referred to as cerebral mycotic aneurysms, are an uncommon but feared compilation of bacterial endocarditis. While bacterial endocarditis is the underlying cause of IIAs in nearly all modern cases, valve surgery is typically postponed until resolution of the aneurysm due to use of perioperative anticoagulation. IIAs have been found to occur in up to 5% of bacterial endocarditis cases, and typically present with multiple lesions.[12,13] Many IIAs present silently and resolve with just antibiotic therapy. While cerebral mycotic aneurysms are rare and carry a low risk of rupture, a ruptured mycotic aneurysm carries devastating neurologic consequences with up to 80% mortality.[3,4] Knowing this, management and surveillance of ruptured IIAs are essential in optimizing long-term outcomes. Asymptomatic cerebral mycotic aneurysms are typically monitored under treatment with just antibiotic therapy. Growing, symptomatic, or ruptured IIAs are typically treated endovascularly or surgically, with recent studies favoring endovascular treatment.[1] Chun *et al*. proposed an algorithm in which endovascular management be pursued in ruptured IIAs in non-eloquent areas without evidence of hematoma.[5] Similarly, Ando *et al*. proposed an algorithm in which endovascular treatment is preferred for management of small, distal IIAs with or without concurrent cardiovascular instability.[1] In this current case, endovascular treatment was chosen due to the distal location and small vessel size, as well as the patient's poor clinical condition.

While the aforementioned algorithms exist, it is important to note that no randomized control trials exist to compare management of IIAs due to their low prevalence.[7,18]

With regard to the etiologic organism, mycotic aneurysms due to *S. anginosus* are exceedingly rare, with just one case documented in literature between the years of 1990 and 2020.[17] This case represents the first complete report of *S. anginosus* IIA. Members of the *S. anginosus* group, namely *Streptococcus constellatus*, are equally rare with the first well documented case of *S. constellatus* having been described by Yen *et al*. in 2007.[16] IIAs are most commonly due to *Viridans streptococci* and *Staphylococcus aureus* which together account

Table 1: Rare etiologies of infectious intracranial aneurysm, corresponding treatment, and outcome.

Etiologic Organism	Author and Year	Country	Age	Gender	Immunosupression	Location	Treatment	Outcome
Bacterial								
Enterococci	Kannoth, 2007[7]	India	12	Female	No	Distal MCA	Clipping	Complete clinical recovery
	Monsuez, 1989[10]	France	38	Male	No	MCA	Clipping and ICHD	Non-resolving severe spastic hemiparesis
Mycobacterium tuberculosis	Saraf, 2013[15]	India	24	Male	No	Left insular MCA	Endovascular	Complete clinical recovery
Cardiobacterium hominis	Okomurua, 2019[11]	Japan	62	Male	No	Distal MCA	ABX and ICHD	Near-complete clinical recovery
Streptococcus constellatus	Yen, 2007[16]	Taiwan	46	Male	No	Bl IICA	Endovascular and ABX	Severe neurological defects
Haemophilus parainfluensa	Barrow, 1990[3]	United Kingdom	33	Male	No	PCA/PICA	Clipping and ABX	Complete clinical recovery
Streptococcus morbidiformis	Barrow, 1990[3]	United Kingdom	53	Female	No	ACA	ABX and ICHD	Complete clinical recovery
Pseudomonis aeruginosa	Barrows, 1993[3]	United Kingdom	12	Male	Burkitt's Lymphoma	Basliar ICA	Clipping and ABX	Perioperative mortality
Streptococcus milleri	Oohara, 1998[12]	Japan	32	Female	No; Postpartum	PCA	Clipping	Perioperative coma followed by mortality
Fungal								
Candia albicans	Barrow, 1990[3]	United Kingdom	18	Female	SLE	Basilar artery	ABX	Perioperative mortality
	Chun, 2001[5]	United States	9	Male	No	MCA M1	ABX	Perioperative mortality
Aspergillus	Yen, 2007[16]	Taiwan	73	Male	ITP	Rt IICA	Endovascular and ABX	Perioperative mortality
	Yen, 2007[16]	Taiwan	79	Female	No	Lt IICA	ABX	Perioperative mortality
	Barrow, 1990[3]	United Kingdom	4	Male	No	BVA	ABX	Perioperative mortality
Petriellidium boydii and Pseudallescheria boydii	Barrow, 1990[3]	United Kingdom	7	Male	No	PCA/PICA	Clipping and ABX	Perioperative mortality
Cocciodes	Chun, 2001[6]	United States	30	Female	HIV	ACA, ACOM	Clipping and wrapping	Complete clinical recovery
Toxoplasma	Chun, 2001[6]	United States	33	Male	HIV	Distal MCA	Clipping and wrapping	Complete clinical recovery

ACA: Anterior cerebral artery, ABX: Antibiotics, ACOM: Anterior communicating artery, Bl Bilateral, BVA: Basilar vertebral artery, HIV: Human immunodeficiency virus, ICHD: Intracerebral hemorrhage decompression, IICA: Intracavernous internal carotid artery, ITP: Immune thrombocytopenia purpura, Lt: Left, MCA: Middle cerebral artery, PCA: Posterior cerebral artery, PICA: Posterior inferior cerebellar artery, Rt: Right, SLE: Systemic lupus erythematosus

for roughly 90% of all IIA cases. Other Gram-positive bacteria including *Enterococci* as well as a few Gram-negative pathogens such as *Pseudomonas aeruginosa* and *Salmonella* make up the remainder of cases.[2,9,11,15] In addition, few rare cases of IIA have been reported with unique viral (e.g., HIV-1 and VZV) and fungal (e.g., *Aspergillus* and *Candida*) etiologies.[6] In this report, we have included a brief summary of well documented IIA cases of rare and unusual causative organisms [Table 1].

As for treatment of the underlying cardiac pathology, valve surgery for infective endocarditis in IIA patients is typically postponed. Current American Association of Thoracic Surgeon guidelines suggest valve surgery be delayed by 1–2 weeks in the setting of non-hemorrhagic stroke and 3–4 weeks in patients with hemorrhagic strokes. In patients with severe neurologic damage such as the patient in this case, it is recommended cardiovascular surgery be delayed until potential for neurological recovery is established.[14] Because no definitive timeline exists for valve surgery in patients with severe neurologic damage, exceptional communication within the patient's care team must be established given the uncertain clinical course.

Due to the rarity of ruptured mycotic aneurysms, little data are available regarding long-term care and outcomes of ruptured IIAs. Unruptured IIA carry a better prognosis than ruptured IIA however the degree of favorability is uncertain.[8] One review of 287 IIA cases over the years 1950–2009 revealed further neurological decline in 20% of surviving ruptured IIA patients on follow-up.[6] Older reviews showed absence of neurological deficit in 40% of ruptured IIA patients at follow up between 1 and 4 years.[10] In conclusion, ruptured IIAs exhibit highly variable prognoses that are confounded by comorbidities, time to intervention, and natural progression of disease.

CONCLUSION

IIAs, sometimes referred to as cerebral mycotic aneurysms, are an uncommon but feared complication of bacterial endocarditis. Rupture of an IIA carries devastating neurologic consequences with up to 80% mortality. Even with timely repair of ruptured IIA, there is still a 20–60% likelihood of progressive or permanent neurological deficits. A well-coordinated, multidisciplinary approach to care for a patient with ruptured IIA resulting from infectious endocarditis can yield favorable long-term results.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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