

## Mycotic aneurysm presenting as subdural empyema: illustrative case

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**BACKGROUND** Mycotic aneurysms (MAs) are rare intracranial pathologies. They are associated with spontaneous rupture, which is often the first presenting sign. Subarachnoid hemorrhage and intraparenchymal hemorrhage are the most common sequelae of ruptured MAs, with subdural hematoma being an atypical presentation. The presentation of an MA as a subdural empyema has not yet been reported in the literature.

**OBSERVATIONS** The authors discussed a 68-year-old man who presented with subdural empyema and received surgery for evacuation. He was found to have a ruptured mycotic aneurysm intraoperatively.

**LESSONS** This case demonstrated a rare and atypical presentation of an MA.

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**KEYWORDS** mycotic aneurysm; subdural empyema; subdural hematoma

The term “mycotic aneurysm” (MA) is a misnomer in medicine. Although the term “mycotic” often refers to fungal disease, MA refers to aneurysms that are formed as a result of inflammatory damage to an arterial wall.<sup>1</sup> MAs account for <5% of intracranial aneurysms, with 65% of them being associated with infective endocarditis.<sup>2</sup>

Intracranial MAs may present unruptured as otherwise unexplained neurological deficits in the setting of infective endocarditis. It has been reported that 65% of patients with MAs present with bacterial infective endocarditis, with other common sources of infection including intravenous drug use (6.3%), bacterial meningitis (5.2%), poor dental hygiene (4.2%), and cavernous sinus thrombosis (2.8%).<sup>3</sup> In these settings, characteristic radiological findings in the cerebrovascular system may lead to the diagnosis of an MA. More typically, however, MAs remain clinically silent until spontaneous rupture. Once ruptured, they primarily present as either intracerebral hemorrhage (ICH) or subarachnoid hemorrhage (SAH).<sup>4,5</sup> Rarely do MAs present as subdural hematoma, with only 13 cases reported in the literature (Table 1). The presentation of an MA as subdural empyema, however, has not been reported in the literature.

### Illustrative Case

#### Presentation and Preoperative Course

A 68-year-old man with a history of alcohol abuse, cirrhosis, and morbid obesity presented to his primary care physician with 2 weeks

of worsening headaches, fatigue, fever, chills, back pain, posterior neck stiffness, and neck pain. The patient had multiple tooth extractions several weeks earlier secondary to dental caries that he had neglected because of medical costs. His primary care physician sent blood cultures that tested positive for *Streptococcus sanguinis*. The patient subsequently presented to the emergency department and was admitted to the medicine service for sepsis and presumed meningitis. Computed tomography (CT) of his head on admission did not demonstrate any intracranial pathology. Lumbar puncture performed by the primary service indicated 18 red blood cells, 114 white blood cells, 52 milligrams glucose per deciliter, and 85 milligrams protein per deciliter. Gram stain and culture results were negative. The patient received penicillin per infectious disease recommendations and was monitored in the hospital. During his evaluation, he was also found to have an aortic vegetation with aortic insufficiency.

Despite the initial CT scan of the head demonstrating negative results, the patient’s headache continued to worsen while he remained otherwise neurologically intact. A CT scan repeated on hospital day 6 showed left-sided extraaxial fluid collection with 6 mm of midline shift toward the right. Brain magnetic resonance imaging (MRI) showed the extraaxial collection with diffusion restriction and correlating apparent diffusion coefficient (Fig. 1). There was also an area within the right insular cortex suggestive of stroke.

**ABBREVIATIONS** CT = computed tomography; ICH = intracerebral hemorrhage; MA = mycotic aneurysm; MRI = magnetic resonance imaging; SAH = subarachnoid hemorrhage.

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**TABLE 1. Subdural hematoma in the setting of ruptured intracranial MAs**

Case No.	Authors & Year	Age (yrs)	Sex	Infective Endocarditis	Involved Cardiac Valve	Associated SAH	Associated ICH	Treatment	Outcome
1	Bamford et al., 1986 <sup>6</sup>	56	M	Yes	Mitral	No	Yes	Craniotomy, removal of aneurysm sac & blood clots	Mild memory disturbance & dysgraphia
2	Bandoh & Sugimura, 1987 <sup>7</sup>	26	F	Yes	Mitral	No	Yes	Craniotomy, removal of SDH & ICH, coagulation & cutting of vessel just distal to aneurysm	Death from cardiac failure 5 days later
3	Barami & Ko, 1994 <sup>8</sup>	36	M	Not reported	Mitral & aortic	No	Yes	Antibiotics, no surgical intervention	Vegetative state
4	Boukobza et al., 2019 <sup>2</sup>	54	M	Yes	Aortic	No	No	Endovascular glue embolization	Full recovery
5	Boukobza et al., 2019 <sup>2</sup>	42	M	Yes	Aortic	No	No	Endovascular glue embolization	Full recovery
6	Kasuya et al., 1985 <sup>9</sup>	13	F	Yes	Mitral	No	No	Unknown; article not in English	Good recovery
7	King, 1960 <sup>10</sup>	23	F	Yes	Mitral	No	No	Craniotomy, removal of aneurysm & hematoma	Good recovery
8	Lee et al., 2013 <sup>4</sup>	72	M	No	Not reported	No	No	Antibiotics, no surgical intervention	Full recovery
9	Matsuda et al., 2002 <sup>11</sup>	22	F	Yes	Mitral	Yes	No	Unknown; article not in English	Good recovery
10	Piastra et al., 2000 <sup>12</sup>	2 mos	M	Not reported	Not reported	Yes	Yes	Surgical clipping & removal	Death
11	Sugino et al., 2002 <sup>13</sup>	65	F	Yes	Mitral	Yes	Yes	Unknown; article not in English	Death
12	Tsuboi & Adachi, 2002 <sup>14</sup>	55	F	Yes	Not Reported	No	No	Unknown; article not in English	Not reported
13	Yamakawa et al., 2003 <sup>15</sup>	21	F	Yes	Mitral	No	Yes	Craniotomy, coagulation of aneurysmal vessel & removal of hematoma	Not reported

SDH = subdural hematoma.

In the clinical context, subdural empyema was presumed. The patient thus received surgery shortly thereafter for evacuation of the subdural empyema.

### Operative Report

The patient was positioned for a standard frontotemporoparietal craniotomy for evacuation of the subdural empyema. After performing the craniotomy and durotomy, we immediately observed that the subdural collection contained both purulent and hemorrhagic components. Furthermore, active bleeding was noted from an arterial vessel. The artery was dilated and irregular, suggesting an MA. The vessel was resected, and the remainder of the subdural collection was evacuated. A subdural drain was placed, duraplasty was completed, and a bone flap was replaced. The incision was closed primarily. The resected vessel was sent for pathology, which confirmed the diagnosis of MA.

### Postoperative Course

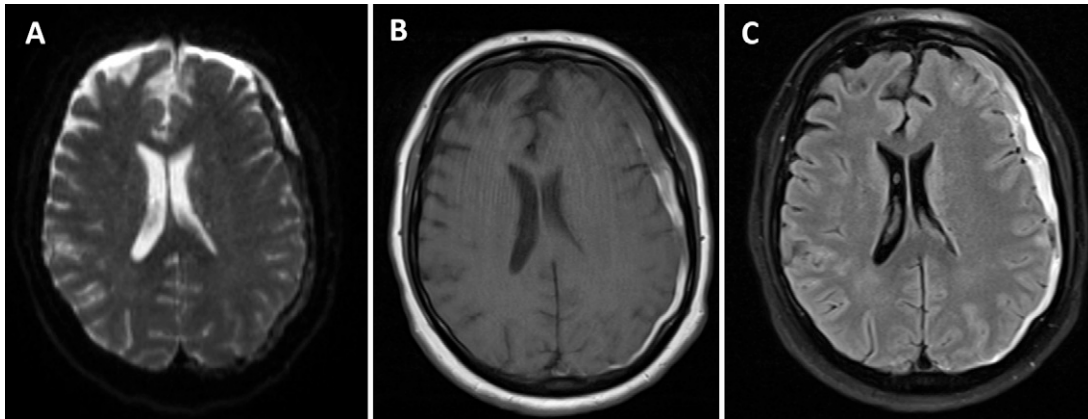
Postoperatively, the patient was monitored in the neurosurgical intensive care unit. Immediate postoperative CT imaging, including

angiography, showed evacuation of the subdural collection and no evidence of aneurysm. The patient improved postoperatively and was neurologically intact with improved headache symptoms. On postoperative day 2, the patient had some intermittent expressive aphasia believed to be secondary to partial seizures. He was maintained on antiepileptic medication at a higher dose. The symptoms were transient and improved with no further issues during the hospital admission. The patient was ultimately discharged on postoperative day 6 to a rehabilitation facility with 6 weeks of antibiotic treatment and close follow-up.

## Discussion

### Observations

MAs are rare intracranial pathologies that typically present after rupture. The literature has indicated that MAs are clinically recognized in 3%–10% of patients with infective endocarditis and account for <5% of all intracranial aneurysms.<sup>3</sup> The presence of SAH or ICH has been reported to be the most common sequela of MAs rupture. The formation of a subdural hematoma as a consequence



**FIG. 1.** MRI sequences showing subdural fluid collection. **A:** Diffusion-weighted imaging. **B:** T1-weighted imaging. **C:** Fluid-attenuated inversion recovery imaging.

of MA rupture is far more atypical but has also been mentioned in the literature. We conducted a PubMed literature review to assess the frequency of subdural hematoma and subdural empyema formation as sequelae of MA rupture. In our literature review, we identified only 13 cases since 1960 of subdural hematoma formation in the setting of ruptured MAs and no reports of subdural empyema as a sequela of MA rupture (Table 1).

The mean age of the 13 patients in our literature review was 37.3 years (range: 2 months to 72 years). Six (46%) of the patients were male and 7 (54%) were female. Ten (77%) of the 13 patients in our literature review presented with associated infective endocarditis, 1 patient did not, and information regarding this condition was missing for 2 patients. The most common cardiac valve involved in patients with infective endocarditis was the mitral valve (70%), with the aortic valve being involved in 2 cases (20%) and the mitral and aortic valves involved in 1 case (10%). In addition to subdural hematoma in these patients, 3 (23%) of them had additional associated SAH, and 6 (46%) of them had associated ICH. In terms of outcomes, 6 (46%) of the 13 patients recovered, whereas 1 patient (8%) had mild memory disturbances and agraphia, 1 patient (8%) was left in a vegetative state, and 3 patients (23%) died. The outcomes of the remaining 2 patients in our literature review were not reported. The management strategy was reported for 9 patients. Of these patients, 5 (56%) received craniotomy for removal of the aneurysm and blood, 2 (22%) received endovascular glue embolization, and 2 (22%) received antibiotic therapy without any surgical intervention.

Our case provides the first account of a patient with a subdural empyema in the setting of a ruptured mycotic cerebral aneurysm. Before the patient received surgery for evacuation, there was no clinical suspicion of mycotic cerebral aneurysm. During the operation, amid the mixture of purulent material and hematoma, the ruptured MA was identified and confirmed through pathology. When we retrospectively reviewed the patient's preoperative MRI, there was evidence of acute hematoma on the susceptibility-weighted sequence present within the subdural collection. Although this finding is most commonly associated with an acute or chronic subdural hematoma, in the setting of this patient's presentation with meningismus and positive blood culture results, an underlying MA as the cause of the subdural collection may have been considered.

However, the lack of more typical types of cerebral hemorrhages associated with MAs and lack of evidence on vascular imaging still makes that an unlikely connection, especially without any literature precedence. Fortunately, in this clinical situation, the MA was small and originated from a superficial cortical vessel that was managed intraoperatively without difficulty. However, a higher index of suspicion in cases with similar presentation and imaging findings is absolutely necessary to ensure that the appropriate team members and surgical equipment are available for management.

### Lessons

Our patient presented with signs of meningeal irritation, infection, and infective endocarditis, all of which may have raised suspicion for an MA rupture. However, the presence of a subdural empyema without SAH or ICH placed this differential lower on the list of possibilities. Although this case report is limited by its nature of being a single case, it demonstrates an atypical presentation of MA rupture that has yet to be reported in the literature and thus contributes to an informed clinical judgment in the assessment of subdural empyema.

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#### Author Contributions

Conception and design: Elhamdani, Jeong, Yu. Acquisition of data: Lucas, Elhamdani, Yu. Analysis and interpretation of data: all authors. Drafting the article: Lucas, Elhamdani, Jeong. Critically revising the article: Elhamdani, Jeong, Yu. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Lucas. Statistical analysis: Lucas. Study supervision: Elhamdani, Jeong.

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