

Atrioesophageal fistula post atrial fibrillation ablation managed with an esophageal stent followed by surgical repair



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Introduction

Atrioesophageal fistula (AEF) is a rare and serious complication of atrial fibrillation (AF) ablation, defined as a connection between the atrium and the lumen of the esophagus.¹ AEF has an incidence of 0.1% to 0.25%² and a mortality over 50%.³ AEF can occur with any of the available AF ablation modalities, including radiofrequency ablation (RFA), cryoballoon ablation, high-intensity focused ultrasound ablation, and surgical ablation.⁴ The clinical presentation of an AEF is usually seen 2–6 weeks post ablation. The most common symptoms are fever and neurological deficits caused by intracranial embolism of air, saliva, and bacteria.² The most useful diagnostic method is via a chest computed tomography (CT) with intravenous (IV) contrast.

Once AEF is identified, the recommended approach is to consider immediate surgical intervention. The literature shows that, based on the management chosen, the mortality rates are 96% with medical management alone, 100% with stent placement, and 33% with surgical repair.⁵

We present a case of AEF complicated with multiple brain embolic strokes with spontaneous hemorrhagic conversion, which was successfully managed by temporizing with an esophageal stent to avoid further embolization, followed by surgical repair 4 weeks later, once the risk of further neurological compromise during cardiopulmonary bypass on high-dose heparin was deemed to be lower.

Case report

A 44-year-old man with symptomatic paroxysmal AF underwent an AF ablation with pulmonary vein isolation, using a point-by-point ablation strategy with a TactiCath ablation catheter (St. Jude Medical) supported by a deflectable long

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KEY TEACHING POINTS

- Atrioesophageal fistulas can be managed with an esophageal stent as a temporal measure to avoid recurrent embolization, followed by surgical repair.
- Computed tomography scans have limitations as a tool to make a definitive diagnosis.
- Interposing tissue between the esophagus and the atrium at the time of surgical repair is important to avoid surgical complications.

sheath (Agilis NxT Steerable Introducer, St. Jude Medical, St. Paul, MN). A temperature probe in the esophagus was used to assess esophageal temperature rises during each lesion and titrated accordingly to avoid a rise above 1°C from its baseline with each lesion. All the lesions on the posterior wall were limited to 30 watts for 10–15 seconds with an lesion size index of 4.0–4.5. Post procedure he developed signs of pericarditis associated with recurrence of AF and was placed on amiodarone and colchicine, and he was on famotidine daily. His symptoms subsided and he continued with an unremarkable recovery until he presented with fever (38°C), dyspnea, chest pain, and recurrence of AF 35 days after the ablation.

A chest CT was performed and was read as normal, with no air noted in the mediastinum (Figure 1). The following day the patient became delirious and incoherent. Brain magnetic resonance imaging showed numerous acute infarcts in the bilateral cerebral and cerebellar hemispheres with hemorrhagic conversion within some of the larger strokes (Figure 2). In conjunction, the patient developed a septic shock requiring pressor. At this point septic emboli due to an AEF was suspected. Broad-spectrum antibiotics were initiated. Of interest, a repeat CT chest with oral and IV contrast was repeated, showing no evidence of esophageal injury, mediastinum inflammation, or clear

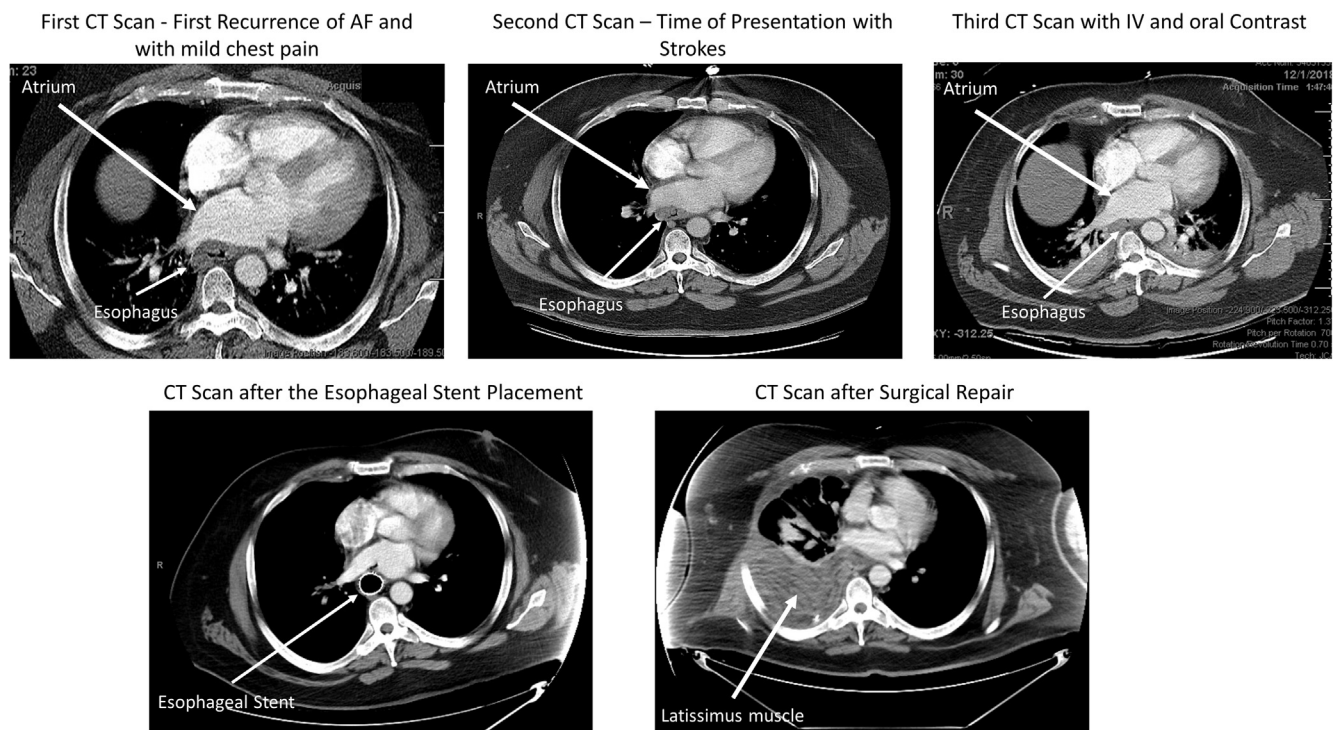


Figure 1 Chest computed tomography (CT) images. The first image represents the first CT scan 2 weeks post ablation and 3 weeks prior to patient's presenting to the emergency department with fever and strokes. Second and third CT are 2 days apart at the time of the atrioesophageal fistula presentation. Fourth CT is post stent placement; fifth CT is after surgical repair and showing the latissimus dorsalis interposed between the atrium and the esophagus.

pneumomediastinum. Given the history of ablation, and the clinical picture, we concluded that an esophageal atrial fistula was the explanation. After a thorough conversation in a multidisciplinary fashion, we felt that attempting a definitive repair with cardiopulmonary bypass and high-dose heparin would be extremely risky, and would likely lead to further intracranial/intraparenchymal bleed, which would result in a poor neurological outcome. Therefore we decided to try an esophageal stent placement to occlude the fistula and avoid further air/saliva embolization until he could get a definitive surgical repair.

Given the risks of further embolization during deployment of the stent, the esophagus was not insufflated. The patient was placed in a reverse Trendelenburg position to force air away from the brain, and a cardiac team was ready for resuscitation and support, as we expected some embolization into his coronary arteries. Venous access was established up front, and a radial arterial line was placed for blood pressure monitoring. During the esophageal stent deployment, air was embolized into the right coronary artery and the patient developed hypotension and ST elevation in the inferior leads, followed by a ventricular

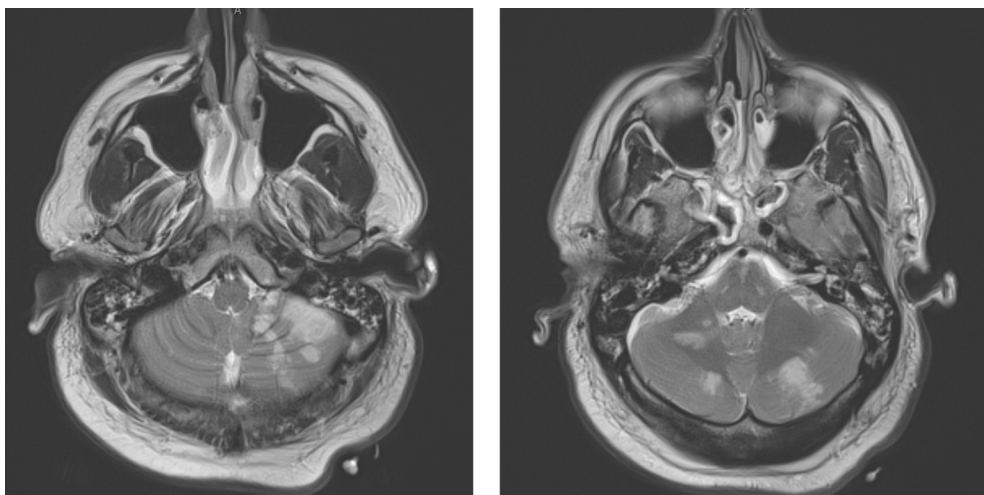


Figure 2 Brain magnetic resonance imaging at presentation showing the multiple cerebellar infarcts with hemorrhagic conversion at the time of presentation.

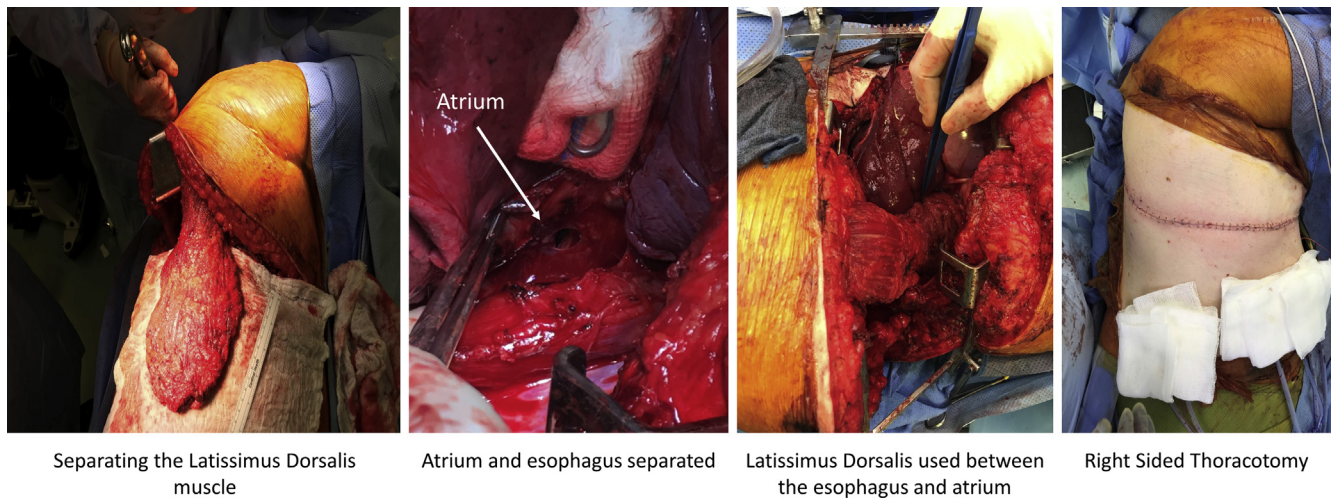


Figure 3 Surgical procedure. From left to right: harvesting of the latissimus dorsalis muscle; the atrioesophageal fistula after the inflamed atrial myocardium was debrided; the latissimus dorsalis placed between the esophagus and left atrium after the primary repair of both structures; right thoracotomy used for the surgical repair.

fibrillation arrest. Cardiopulmonary resuscitation was initiated while a balloon pump was placed, epinephrine and amiodarone were used, and sinus rhythm was restored after a 360 joule shock. The ST changes were transient and disappeared after about 15–20 minutes. A coronary angiogram was performed and found no evidence of obstructive coronary disease or thrombus to suspect an acute coronary syndrome due to plaque rupture.

Post esophageal stenting, the patient started to improve clinically. The pressors were weaned off and the balloon pump was removed. CT chest/abdomen/pelvis with IV contrast showed stable esophageal stent placement (Figure 1). Another brain magnetic resonance imaging showed evolving multifocal supratentorial and infratentorial ischemic infarcts with new punctate focus of ischemia and multiple areas of hemorrhagic conversion (Figure 2). The blood cultures came back positive for *Candida albicans*, *Streptococcus mitis/loralis* (*S. viridans* group), and *Rothia mucilaginosa* (*Stomatococcus mucilaginosus*). All the central lines were removed and replaced. IV antibiotics were adjusted accordingly.

The patient continued to improve clinically. On day 10 of admission (day 7 post stenting), he was extubated. On day 20 of admission (day 17 post stenting), he was discharged to an inpatient rehabilitation center with IV antibiotics.

Five days prior to the scheduled surgery for atrioesophageal repair, the patient became febrile and the antibiotics were switched to meropenem and micafungin. At that point and given the improved neurological status, another brain CT was repeated, showing evolving bilateral infarcts, and no intraparenchymal blood was noted, at which point he underwent full surgical repair.

During the surgery a right lateral thoracotomy was performed, the latissimus dorsi muscle flap was harvested (Figure 3), the fifth rib was removed, and the right chest was entered.

Once exposed and after reaching an activated clotting time of 450 seconds, the aorta and inferior/superior vena cava were cannulated and the patient was placed on cardiopulmonary bypass. Then the ascending aorta was cross clamped to avoid further embolization. At that point an endoscopy was done to remove the esophageal stent. The left atrium was then opened and the AEF was transected with the cautery. A small hall with surrounding inflamed tissue was debrided and the atrium was repaired with a pericardial patch. The patient was taken off cardiopulmonary bypass and the esophagus was repaired with primary closure. The latissimus dorsi flap was placed between the esophagus and the atrium (Figure 3).

The patient had an uncomplicated postsurgical recovery. An esophagram showed an intact esophagus without leaks and the patient was discharged on postoperation day 22.

Post discharge, the patient continued to improve and was left with no neurological deficits, and was able to go back to work 9 months after the event.

Discussion

AEF can occur 1–6 weeks after the index ablation,² typically at 20 ± 12 days post-RFA.³ The median age of presentation is 55 ± 13 years, more frequently in male than in female subjects.⁴ It can happen after any mode of ablation, including surgical ablation, percutaneous RFA, percutaneous cryoballoon ablation, and high-intensity focused ultrasound.⁶ The esophageal damage after ablation can happen right after the procedure, and it is a slow progression from there until the fistula is formed. One recent study showed that within 7 days after ablation, esophageal injuries happened in 15% of the cases. Among these injuries, 36% were esophageal erythema, 39% were superficial ulcers, and 25% were deep ulcers. Furthermore, 4.2% of the deep ulcers progressed to esophageal perforation or AEF.⁷

Our patient presented at 35 days after the ablation. He initially had such nonspecific symptoms as fever, chest

pain, and dyspnea, and his chest CT was initially unremarkable. On the day after admission, the patient developed symptoms of stroke. Although further CT of the chest with IV and oral contrast were not clearly suggesting an AEF, the clinical presentation pointed to the diagnosis. Han and colleagues⁶ had reported that in 7% of the AEF cases, the initial CT was normal, suggesting progression of the AEF over time, and a repeat CT 4–12 days later was required to demonstrate the abnormal findings by imaging. It is important to note that obvious findings like mediastinal air or extravasation of IV or oral contrast were never noted in our case and the diagnosis was established mainly owing to the clinical presentation (Figure 1).

Traditionally, treatment of an AEF is a medical emergency that requires urgent surgical repair. Previous case series and systemic reviews have reported mortality rates of 83%–100% with nonsurgical management as compared to 34%–41% with surgical repair.^{1,4} Mohanty and colleagues⁸ have reported a case series of 9 patients; 5 patients receiving stents died within 1 week of the procedure, while the other 4 patients who received surgical repair survived at median follow-up of 2.1 years. There are only small numbers of case reports that have reported successful esophageal stenting therapy. Bunch and colleagues⁹ have described the first case where esophageal stenting combined with IV antibiotics and nasogastric tube feeding was used to manage an esophageal perforation without a fistula into the atrium. The patient was followed up for 18 months and had been returned to physical activities and remained free from AF on antiarrhythmic drugs. Eitel and colleagues¹⁰ reported 3 cases of esophagopericardial fistulas successfully treated with stenting.

We believe the surgical intervention still remains the mainstay of AEF treatment. Our case explored temporizing the patient with an esophageal stent, followed by surgical repair, in a patient who was not suitable for immediate surgical intervention owing to strokes with hemorrhagic conversion. This case adds to the literature that supports this intervention, as very few reported attempts have been described. A review article by Zhou and colleagues¹¹ presented a favorable outcome of combined therapy with stenting and surgery. The article showed that mortality was 100% in patients treated with stenting only or conservative medical management and 71% in patients who had surgery; 2 out of 3 patients who were implanted with esophageal stent after performing emergency atrial wall repair thoracotomy survived; and 2 patients who received esophageal stenting and, subsequently, surgery both survived.¹¹

In our particular case the latissimus dorsi was used and placed between the esophagus and the atrium after the esophagus was surgically repaired. Other techniques described employ the omentum, a pericardial patch, or an intercostal muscle to wrap the esophagus or interpose tissue between

the structures and build a barrier to wall off the inflammatory process and avoid recurrence of the fistula, esophageal leakage, or mediastinitis. Cases where tissue has been interposed between the esophagus and the atrium have been associated with fewer postoperative complications compared to those where primary repair alone was performed.⁴

The present case adds to the body of literature and specifically addresses the measures taken at the time of stent deployment, as well as the thought process and sequence of events that resulted in a very favorable outcome. The rationale would be that as the esophageal ulceration penetrated into the left atrium, a 1-way valve is formed with leakage into but not out of the left atrium.² Thus, occluding the esophageal perforation will effectively stop the ongoing embolization, but does not lead to healing of the atrium; therefore, surgical repair remains the cornerstone of the definitive treatment.

Conclusion

For those patients with AEF who cannot tolerate surgery or have contraindication for immediate surgery, esophageal stenting could be a temporary bridge to prevent further embolization until the patient is capable of undergoing definitive surgical fistula repair.

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