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CASE REPORT | ESOPHAGUS

Acute Stroke After Upper Endoscopy in a Patient With a Suspected Atrioesophageal Fistula

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ABSTRACT

Atrioesophageal fistula (AEF) is a rare complication of atrial fibrillation ablation. We present a man with sepsis and frank hematemesis 3 weeks after atrial fibrillation ablation. Thoracic computed tomography showed no definitive evidence of AEF. He underwent esophagogastroduodenoscopy and subsequently developed an embolic stroke. In the operating room, he was found to have AEF. This case highlights the importance of maintaining a high index of suspicion for AEF because of its nonspecific presentation and difficulty in diagnosing with imaging or endoscopy. Once AEF is suspected, esophagogastroduodenoscopy should be avoided because of the risk of precipitating embolic events.

INTRODUCTION

Atrioesophageal fistula (AEF) is a rare complication of atrial fibrillation (AF) ablation and esophageal malignancy. It presents with variable symptoms. It is a difficult diagnosis that can sometimes be made with thoracic computed tomography (CT). Endoscopic evaluation is contraindicated when there is clinical suspicion for AEF because CO₂ insufflation can lead to acute stroke from CO₂ embolus. The management of AEF is typically urgent surgery because the mortality rate of untreated AEF is 100%.

CASE REPORT

A 40-year-old man presented to the hospital after an episode of hematemesis. Four weeks earlier, he had undergone catheter ablation of the left atrium for medically refractory rapid AF. Three weeks after the procedure, he developed pleuritic chest pain, odynophagia, malaise, headache, and fevers. He presented to the hospital with sepsis and hematemesis with a temperature of 100.8°F, heart rate 96 bpm, and blood pressure 89/50 mm Hg responsive to fluid resuscitation. A thoracic CT with intravenous contrast demonstrated small locules of gas along the esophageal wall abutting the left atrium though no definite esophageal perforation or AEF was seen. Owing to the clinical suspicion for AEF, he was transferred to our cardiothoracic intensive care unit for a higher level of care.

At our hospital, a gated thoracic CT with intravenous contrast was performed, which again showed punctate foci of gas between the left atrium and the esophagus concerning esophageal leak, but without a clear AEF (Figure 1). The patient was too unstable to undergo an esophagram. Gastroenterology was consulted to evaluate the esophagus for mucosal injury. Upper endoscopy was initially performed without CO_2 insufflation to avoid the risk of causing CO_2 emboli through the potential AEF. The esophagus could not be adequately examined. On discussion with the cardiothoracic surgeon, minimal insufflation with CO_2 was used to inspect the esophagus. Immediately after esophageal CO_2 distension, a large amount of fresh blood was seen spurting in the mid esophagus, confirming the presence of the AEF (Figure 2). Approximately 5 minutes after completion of the procedure, the patient developed new-onset left hemiplegia, left gaze deviation, and lethargy. Emergent surgery was recommended. A 3 \times 5 mm defect in the posterior wall of the left atrium with visibility of esophageal fibers through a perforation was observed. A postoperative cranial CT without contrast obtained demonstrated a new right middle cerebral artery territory ischemic stroke. A subsequent cranial CT angiography showed normal vasculature.

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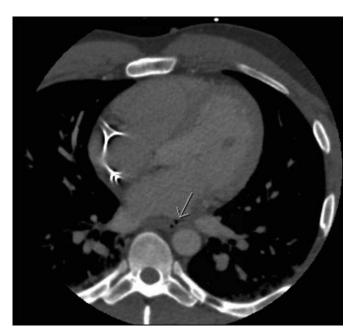


Figure 1. Gated thoracic computed tomography with intravenous contrast showing a punctate focus of gas between the left atrium and esophagus (arrow) concerning for an esophageal leak but without a clear atrioesopahgeal fistula.

The patient's hospital course was complicated by *Streptococcus mitis* bacteremia with no evidence of infective endocarditis on transthoracic echocardiogram. After 20 days of hospitalization, the patient was discharged home on intravenous antibiotics for a six-week treatment and continued outpatient rehabilitation. On a 3-month outpatient follow-up, the patient had significant clinical improvement overall, with mild left-sided neurological deficits.

DISCUSSION

AEF is a rare complication that occurs in 0.1%-0.25% of AF ablation procedures with a higher prevalence among men



Figure 2. Esophagogastroduodenoscopy showing a large amount of fresh blood in the mid esophagus.

compared with women.^{1,2} Symptom onset for AF ablation complications has been reported to occur around 19–20 days postprocedure (although it can vary between 1 and 6 weeks), with delayed diagnosis typically suggestive of AEF carrying high mortality.^{3,4} This time frame was similar for our patient who presented approximately 3 weeks after the ablation procedure. The close proximity of the anterior esophagus and the posterior left atrial wall allows for this fistula formation, typically serving as a one-way valve from the esophagus into the atrium.¹ There are various proposed mechanisms of injury, including direct thermal injury, exacerbation of or new-onset gastroesophageal reflux disease caused by ablation, infection from the lumen, and ischemic injury due to thermal occlusion of end arterioles.⁵

Patients with AEF present with nonspecific signs and symptoms of fever, fatigue, chest discomfort, dyspnea, dysphagia, nausea, vomiting, odynophagia, hematemesis, and melenasimilar to our patient's presentation. In one systematic review of 53 case reports of AEF, fever was the most common (n = 44), with second being neurological symptoms (n = 27), and finally hematemesis (n = 19).⁶ The one-way valve mechanism from the esophagus into the left atrium explains why neurologic events, strokes caused by esophageal debris, and air traveling to the brain are more commonly seen than hematemesis. Patients undergoing upper endoscopy have a significantly higher risk of embolic events even if CO2 is used for insufflation. This is because the force of insufflation opens the valve closing the fistula, allowing a rush of air and esophageal contents to travel into the left atrium. This is the most likely reason why in our patient we only saw blood in the esophagus after insufflating the esophagus. Although CO₂ is an inert, noncombustible gas that is rapidly absorbed and is highly soluble in water and blood, there have been case reports of fatal embolic events occurring with CO2 during endoscopy. For this reason, endoscopy should be avoided if there is any concern for AEF.1 Patients presenting with sepsis due to AEF also tend to grow Gram-positive organisms in their blood culture, most likely attributed to oral flora, very similar to our patient who had bacteremia with S. mitis.

Diagnosis of AEF is best made with magnetic resonance imaging or CT of the esophagus with oral contrast, which can be seen extravasating from the esophagus into the left atrium.¹ In one review of 120 AEF cases, thoracic CT was the most common modality of diagnosis, accounting for 68%, whereas another systematic review of 53 cases identified 51% of diagnosis performed by the same modality. 4,6 Similarly, another systematic review found a clear diagnosis of AEF was made in 35% of cases with thoracic CT modality.8 As such, in cases with high suspicion, surgeons sometimes may have to commit to a surgery without a confirmed diagnosis of fistula, especially since early surgical repair is crucial, and the mortality without treatment is 100%. There have been reported cases of endoscopic esophageal stent placement for the management of AEF; however, most of these cases have shown much worse outcome than compared with surgical repair. 10

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In summary, AEF is a rare complication of AF ablation that carries high morbidity and mortality. It is important to be aware of the delayed time frame of patient presentation, presenting signs and symptoms, diagnostic modality, and the best management strategy. If there is any suspicion of AEF, upper endoscopy should be avoided because of the high risk of causing systemic embolization of air, CO₂, and oral/gut bacteria, which can lead to serious neurologic deficits and systemic sepsis. Similarly, endoscopic management of AEF leads to significantly poor outcomes and thus should not be attempted.

DISCLOSURES

Author contributions: N. Thapa and Y. Ando wrote the manuscript and reviewed the literature. SW de Melo Jr wrote the manuscript, reviewed the literature, approved the final version, and is the article guarantor.

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