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Left Atrial to Esophageal Fistula: A Case Report and Literature Review

Authors' Contribution:
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Statistical Analysis C
Data Interpretation D
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Patient: Male, 57
Final Diagnosis: Left atrial to esophageal fistula
Symptoms: Chest pain • syncope
Medication: —
Clinical Procedure: —
Specialty: Cardiology





Objective: Unusual clinical course
Background: Left atrial to esophageal fistula (LAEF) is a rare fatal complication of radiofrequency ablation (RFA) for atrial fibrillation and is associated with high mortality. Clinical features can be nonspecific and include fever, dysphagia, upper gastrointestinal (GI) bleeding, sepsis, and embolic stroke a after recent history of RFA for atrial fibrillation.

Case Report: A 57-year-old Caucasian male was brought to the emergency department (ED) by his family because of an altered mental status. He had undergone a radiofrequency ablation for paroxysmal atrial fibrillation three weeks earlier. Several hours after admission to the ED, the patient transiently became unresponsive and had a right sided hemiplegia. A brain MRI revealed multiple cerebral infarcts. On the following day, the patient had an episode of melena, and an esophagogastroduodenoscopy (EGD) was performed which did not reveal any source of bleeding. While the patient was being monitored in the intensive care unit (ICU), he had an episode of hematemesis and went into cardiac arrest from which he was successfully resuscitated and transferred to another facility. He had another EGD, which uncovered a flap of mucosa covering the lower third of his esophagus and a 1 cm fistulous opening was seen with fresh blood oozing out of it. The patient had another cardiac arrest during the endoscopy and died despite all measures.

Conclusions: We present this case to stress the importance of early diagnosis of LAEF. LAEF can be fatal if diagnosis is delayed or missed. Early surgical intervention can reduce LAEF morbidity and mortality. Newer diagnostic modalities such as endoscopic ultrasound (EUS) can be helpful in cases where conventional imaging is unclear.

MeSH Keywords: Atrial Fibrillation • Catheter Ablation • Endosonography • Esophageal Fistula • Heart Atria

Full-text PDF: <http://www.amjcaserep.com/abstract/index/idArt/899878>

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Background

Left atrial to esophageal fistula (LAEF) is a rare often fatal complication of radiofrequency ablation (RFA) for atrial fibrillation and is associated with high mortality [1]. It usually develops between three and 60 days post RFA [2]. The incidence of LAEF after atrial fibrillation ablation is 0.3–0.4% [3]. RFA is a common procedure that is performed in medical treatment-resistant patients who have persistent atrial fibrillation. In one study of catheter ablation of atrial fibrillation, LAEF was reported to be the second most common cause of death with a mortality rate of 71% [4]. The presenting clinical features of LAEF include fever, dysphagia, upper gastrointestinal (GI) bleeding, sepsis, and embolic stroke after a recent history of RFA or cryoablation for atrial fibrillation [5]. There are no current guidelines regarding the diagnosis and management of this lethal complication. Morbidity and mortality in LAEF cases can be secondary to stroke after septic or air embolus to the brain, septicemia is usually from a GI source organism or GI bleed [6]. Blood cultures from patients with sepsis secondary to LAEF often grow Gram-positive organisms [2]. Herein, we report a rare case of a LAEF, together with a review of the current literature, to aid in a better understanding of this critical condition.

Case Report

A 57-year-old Caucasian male with a past medical history of hypertension, diabetes mellitus, and paroxysmal atrial fibrillation presented to our emergency department (ED) post radiofrequency ablation. Approximately three weeks prior to his ED visit, he had pulmonary vein isolation of all four pulmonary veins without any immediate post-procedure complications (no further details of his treatment were available). The patient was brought to the ED by emergency medical services (EMS) for altered mental status. He had been experiencing recurrent sharp chest pain after the ablation procedure and was on colchicine for presumed pericarditis. According to his family, the patient suddenly became unresponsive while having dinner, without any response to verbal commands and without any seizure-like activity, or urinary or bowel incontinence. The EMS cardiac monitor revealed an episode of supraventricular tachycardia (SVT), which resolved spontaneously. Shortly thereafter, he was more alert; he stated that he had experienced sweating with palpitations and a funny sensation in his chest just prior to the unresponsive event. His home medications included warfarin, tamsulosin, omeprazole, colchicine, and multivitamins.

On admission, his vitals included temperature of 98.1°F (36.7°C), blood pressure of 149/88 mm Hg, heart rate of 66 beats/minute, respiratory rate of 27 breaths/minute, and oxygen saturation

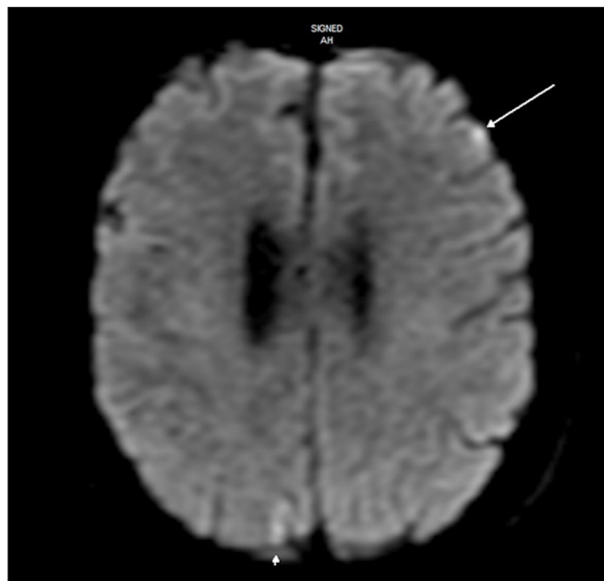


Figure 1. MRI of the brain showing multiple bilateral infarcts (white arrows).

of 100% on two liters via nasal cannula. On cardiac examination, the patient had normal first and second heart sounds with no audible murmurs, rubs, or gallops. The patient's lung fields were clear to auscultation bilaterally. Neurological examination revealed the patient to be alert, awake, and oriented with a normal tone and strength in upper and lower extremities with preserved reflexes. His laboratory workup showed first troponin I of <0.30 ng/L, sodium of 136 mEq/L, potassium of 3.9 mEq/L, chloride of 98 mEq/L, bicarbonate of 22 mEq/L, creatinine of 0.62 mg/dL, hemoglobin of 10.9 g/dL and an INR of 3.4. The radiograph of his chest was normal with no evidence of cardiomegaly, pulmonary infiltrates, or edema.

A few hours after the admission, the patient had another episode of unresponsiveness during which he did not respond to verbal or painful stimuli. Two minutes later, he regained consciousness but was unable to move his right upper and lower extremities. Neurological examination at this time showed no cranial nerve deficits but 2/5 strength in right upper and lower extremities with preserved reflexes in all limbs and a positive Babinski reflex on the right side. A 12-lead ECG showed sinus tachycardia with no ST segment or T wave changes. Computed tomography (CT) scan of brain was negative for bleeding or any mass. A magnetic resonance imaging (MRI) of the brain revealed multiple infarcts which raised the possibility of a cardioembolic phenomenon (Figure 1).

Because the patient was on warfarin with an INR of 3.4, he was not a candidate for tissue plasminogen activator. The patient underwent a transesophageal echocardiography (TEE) that showed turbulent blood flow, raising the suspicion for vegetation at the right atrial and inferior vena caval junction.



Figure 2. First EGD showing completely normal esophagus with no visible ulcers or fistulous opening.



Figure 4. Image of the axial section of chest CT with oral contrast showing air near the posterior wall in the left atrium adjacent to esophagus (white arrow).



Figure 3. Image of sagittal section of chest CT with oral contrast showing air between left atrium and esophagus with oral contrast along posterior left atrial wall (black arrow).

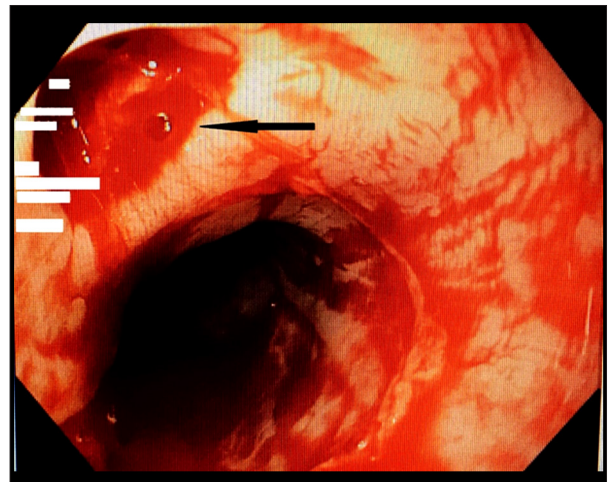


Figure 5. Repeat EGD showing the 1 cm fistulous opening in the anterior wall of lower third of esophagus showing blood gushing out (black arrow).

On the second day of hospitalization, he had an episode of melena. An esophagogastroduodenoscopy (EGD) did not reveal any lesions in the esophagus or stomach (Figure 2).

Warfarin was stopped and the patient received packed red blood cells as his hemoglobin was below 7 g/dL. He also became febrile with a temperature of 104°F (40°C). He was empirically started on vancomycin, cefepime and metronidazole. Blood cultures grew *Clostridium perfringens*. CT scan of the chest, abdomen and pelvis did not show any foci of infection but revealed a 3-mm pocket of air in the dependent portion of the left atrium, which was considered to be iatrogenic (Figures 3, 4).

A day later, he had another episode of massive hematemesis followed by cardiac arrest from which he was successfully

resuscitated. He was intubated during the cardiac arrest and was placed on mechanical ventilation. A CT brain scan showed interval progression of bilateral infarcts compared to previous brain imaging. The patient was transferred to another facility where repeat EGD again failed to reveal any active source of bleeding, but on retraction of the endoscope, a flap opened up in the lower third of esophagus and a 1 cm fistulous opening was seen with fresh blood gushing out of it (Figure 5).

The patient had another cardiac arrest during the endoscopy and he died despite all measures.

Discussion

Several possible factors could be responsible for the formation of LAEF after RFA, such as thermal injury to the esophagus,

injury to the end arterioles leading to ischemia, enzymatic injury, or an infection from a GI source of organisms [6,7]. Another potential risk factor for fistula formation is esophageal dysmotility after general anesthesia, rendering that area of esophagus vulnerable to persistent thermal injury during the ablation procedure. Also, due to general anesthesia, the physiologic cooling mechanism of the esophagus that results from swallowing is absent and this may increase the likelihood of esophageal injury [8]. However, even though esophageal injury is frequently reported, the incidence of LAEF is fortunately rare. Esophageal wall injury after RFA varies from erythema to ulcers, and is reported after endoscopy in nearly 47% of patients after pulmonary vein isolation (PVI) [8]. Although EGD is the gold standard for diagnosing upper GI bleed, it can be catastrophic if used in cases of suspected LAEF due to the risk of massive air embolization [9].

Several studies have been done to detect early esophageal changes after ablation using different modalities, such as endoscopic ultrasound (EUS) and capsule endoscopy [8,10]. Zellerhoff et al. used EGD and EUS as a diagnostic modality in the followup after PVI for atrial fibrillation ablation in patients at high risk for LAEF, and showed EUS was highly sensitive in not only diagnosing the esophageal injury but also diagnosing the injury of adjacent mediastinal structures [8]. They noted that 27% of patients demonstrated structural mediastinal changes as detected by EUS [8]. EUS combined with immediate operative repair can evade the potential sequelae of LAEF. If diagnosed early, this fatal complication can be managed and result in lifesaving [7]. Di Biase et al. utilized capsule endoscopy for detecting esophageal changes after RFA. Their study revealed that capsule endoscopy was better tolerated and provided satisfactory images for detecting esophageal injury without the potential risk of insufflation which occurs with regular EGD. However, further studies are required to determine whether capsule endoscopy can be a useful clinical tool [10]. CT angiography of the chest is the diagnostic modality of choice as it can show the connection between esophagus and left atrium with extravasation of contrast from the left atrium into esophagus via a fistulous tract. A review of the literature suggested changes, such as air in the left atrium, esophageal injury, or diverticulum on the posterior wall of the left atrium, can also be seen. MRI or CT of the brain usually reveals either septic, air emboli, or pneumocephalus. In one case report, intra-cerebral abscesses were reported [2].

Per our literature review, one of the major reasons for the delay in diagnosis of LAEF was its similar clinical presentation to that of infective endocarditis, which can also present as multifocal strokes secondary to septic emboli, high grade fever, and positive blood cultures. These patients are usually first evaluated in a stroke unit or they end up on the infectious disease service ward. Early recognition of LAEF is vital



Figure 6. Chest CTA with contrast sagittal view, showing air in the mediastinum between esophagus and left atrium (white arrow).

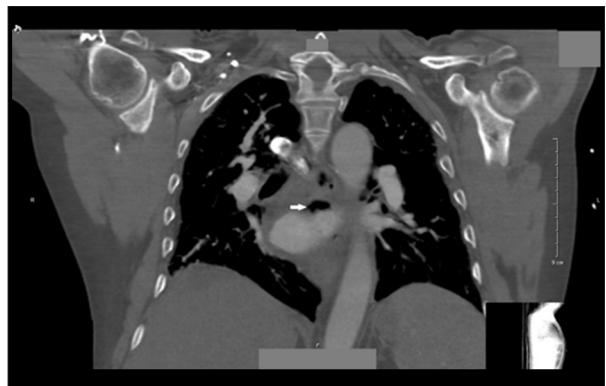


Figure 7. Chest CTA with contrast coronal view showing air in the mediastinum around the left atrium and pulmonary veins (white arrow).

in patients who recently underwent RFA for atrial fibrillation. Due to the lack of awareness and the rarity of this fatal condition, the diagnosis is often delayed. Literature suggests several precautionary measures that might help prevent esophageal injury during atrial fibrillation ablation, such as the use of prophylactic proton pump inhibitors, esophageal temperature monitoring with esophageal visualization, and protective devices. Once the diagnosis of LAEF is made, early surgical repair is essential, as the mortality rate is 100% without treatment [5]. There are various surgical approaches available for the repair of LAEF, which includes primary esophageal repair and placing a biologic barrier between the esophageal and atrial repair. This approach helps prevent refistulization and is associated with lower morbidity and mortality [11].

Although surgery is the gold standard treatment for LAEF, there was one reported case in which the patient was conservatively managed with repeated endoscopic snaring of the esophageal mucosa, accompanied by antibiotic therapies which eventually lead to improvement in the patient's condition without any evidence of symptoms recurrence [12]. Our case highlights the need for a high degree of clinical suspicion when investigating patients with sepsis and neurological signs following atrial fibrillation ablation. The timing of the events, along with a careful history taking and radiographic findings, can guide the clinician towards the precise diagnosis and early surgical intervention that can be lifesaving. The CT angiography can show subtle findings, such as in our case where the covering mucosal flap prevented visualization of the fistula and showed a 3 mm air pocket in the left atrium (Figures 6, 7). Interestingly, this mucosal flap may have

also prevented the patient from having life threatening upper GI exsanguination.

Conclusions

LAEF is a rare but fatal complication post RFA. A high degree of clinical suspicion is key to early diagnosis as prompt surgical intervention can be lifesaving. Advanced modalities like EUS in cases with equivocal conventional imaging can be of great help in diagnosis. This approach can help prevent delay in diagnosis and preclude catastrophic events.

Conflict of interest

None.

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