

Detection of an Atrioesophageal Fistula on a Transthoracic Echocardiogram



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INTRODUCTION

Atrial fibrillation (AF) is a dysrhythmia associated with a threefold increased risk for heart failure and a fivefold increased risk for stroke.^{1,2} It is typically associated with a left atrial (LA) arrhythmogenic substrate or trigger. Therefore, catheter ablation procedures, mainly the radiofrequency type, are widely used in its treatment.³ However, despite the efficacy of radiofrequency ablation, it has been associated with several complications, such as cardiac tamponade, embolic events, pulmonary vein stenosis, and extracardiac injuries.^{4,5} We hereby present a case of atrioesophageal fistula (AEF) complicating an AF ablation procedure that was first suspected on transthoracic echocardiography.

CASE PRESENTATION

A 65-year-old Caucasian man known to have hypertension and persistent AF for almost a decade presented to our institution for management of his arrhythmia. Because his AF was refractory to antiarrhythmic drug therapy, he successfully underwent a catheter-based radiofrequency pulmonary vein isolation procedure. At clinic follow-up 2 weeks following discharge, the patient reported an irritative cough, which was attributed to a respiratory origin. Three weeks later, he presented to the emergency department with a severe headache, a persistent cough, and fever. On physical examination, he was awake with unremarkable vital signs except for fever. His cough was incessant and was noted to exacerbate whenever he tried to speak. Blood workup showed significant leukocytosis (21,000 cells/mL). Brain computed tomography revealed a small area of subarachnoid hemorrhage, which was attributed by the neurology team to his excessive coughing on a background of anticoagulant therapy. The patient was admitted to the intensive care unit, warfarin was withheld, and he was started on broad-spectrum antibiotics. As part of workup for reported shortness of breath along with a possible cardiac etiology for his presentation, transthoracic echocardiography was ordered. During this study, bubbles intermittently appeared mainly in the left ventricle, with a less impressive number in the left atrium; the bubbles were particularly noted whenever the patient coughed (*Figure 1, Videos 1 and 2*). This finding, which is suggestive of the presence of

air within the heart, raised the possibility of a communication with either the esophagus or the bronchial tree. Before the next step to pinpoint the diagnosis was contemplated, the patient's clinical situation started to quickly deteriorate, with a remarkable decline in his consciousness level, presumably secondary to cerebral air emboli. This necessitated mechanical ventilation, in which the endotracheal tube was purposefully placed in the right main bronchus to avoid further damage in case a fistula between the left atrium and the airways existed. With the patient's recent LA radiofrequency ablation in mind, and because of his rapid clinical deterioration, a decision was made to proceed with upper endoscopy, a potentially diagnostic and therapeutic option. With insufflation turned off to prevent potential deleterious air entry into the heart, endoscopy was performed showing a large clot attached to the anterior aspect of the distal esophageal wall, a finding which was highly suggestive of an AEF. A covered stent was immediately placed over the fistula's entry point. Despite all these efforts, the patient's condition worsened further, until he eventually sustained a cardiac arrest, which could be the result of a coronary artery air embolization. Resuscitation attempts were unfortunately futile and he was pronounced dead. Autopsy later clearly delineated a fistula measuring 3 mm in diameter connecting the esophagus and the left atrium (*Figure 2*).

DISCUSSION

AEF is one of the most serious complications of LA ablation. First reported in the surgical literature in 2001, it has been widely recognized ever since.^{5,6} Although its true incidence may be underreported, it is still very low, ranging from 0.015% to 1.5%.⁵⁻¹¹ Most cases of AEF occur in men, with a range of 2 to 60 days between the procedure and presentation. AEF has a very high mortality rate ranging from 40% to 80%, and survivors are likely to have permanent neurologic deficits.^{4,6-8}

Despite efforts to determine the mechanism of development of this complication, its pathogenesis is still not completely understood. There are some known contributory anatomic factors, particularly the close proximity of the esophagus to the LA posterior wall.^{7,8,11,12} Interestingly, it has been proposed that the absence of a fat layer between the esophagus and the left atrium may be helpful in identifying patients who are at higher risk for developing AEF.^{7,11} Because esophageal thermal lesions are frequently delivered during AF ablation, the risk for developing an AEF is increased by the duration and magnitude of local heating.⁷

Clinical presentation is variable, but the most reported signs and symptoms include fever, neurologic deficits, hematemesis, chest pain, and odynophagia.⁶⁻⁸ Although fever is the most common symptom, it is nonspecific because it can be the result of tissue inflammation related to the procedure. Neurologic manifestations range from mental status alteration to stroke or seizure, and these tend to occur 2 to 3 weeks after the procedure.⁶ Although brain computed

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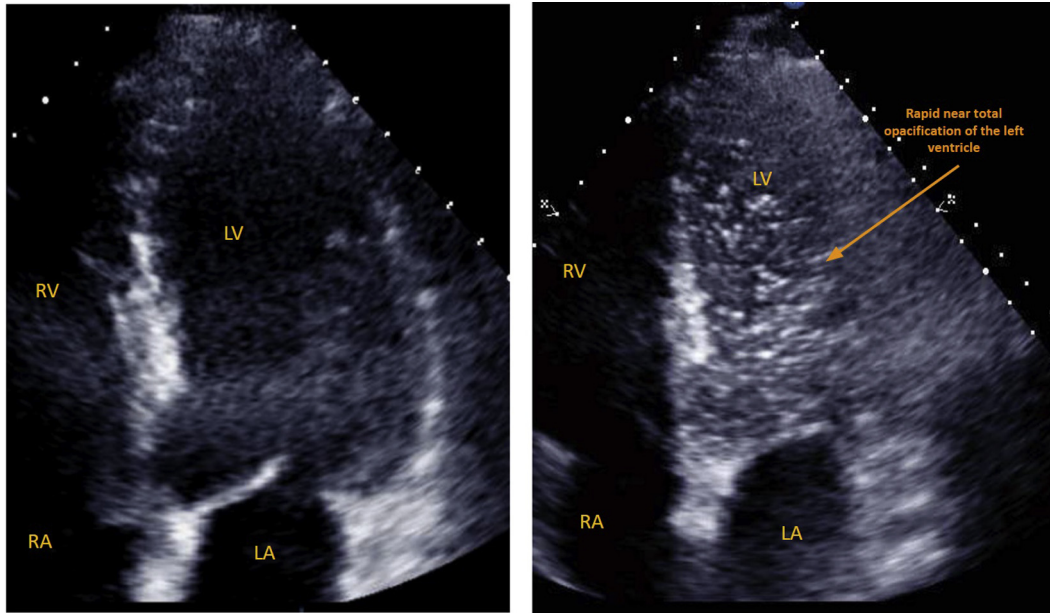


Figure 1 Transthoracic echocardiography revealed a significant number of bubbles, which were spontaneously opacifying most of the left ventricle (LV) whenever the patient coughed, seemingly filling the ventricle in a basal-to-apical fashion. LA, Left atrium; RA, right atrium; RV, right ventricle.

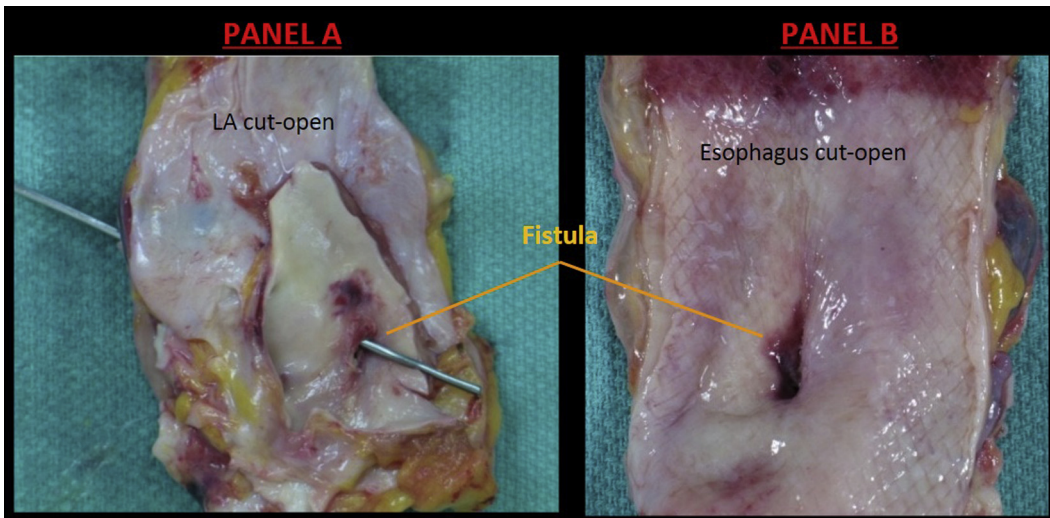


Figure 2 Postmortem autopsy study clearly defining the fistula with its atrioesophageal path. **(A)** Left atrium (LA) cut open with an inserted metallic rod clearly pinpointing the path of the fistula toward the esophagus (not seen) on the back. **(B)** Esophagus cut open, showing the entry point of the fistula. Also note the imprints on the esophageal mucosa from the previously present stent.

tomography was not performed to confirm this theory, our patient's rapid in-hospital mental status decline was presumed to be secondary to cerebral air emboli. As in our case, leukocytosis is commonly reported and is likely secondary to bacteremia.^{6,9,10} Although cough is not a common clinical presentation, it was present in this case right after the ablation and evolved to become incessant and intractable, ultimately contributing to the subarachnoid hemorrhage.

Because AEF is a rare complication with nonspecific symptoms, it is often challenging to make the diagnosis. A high index of suspicion is required in patients with history of a recent ablation who present with fever, neurologic deficits, cough, or sepsis. As previously

mentioned, a blood test can reveal leukocytosis, which is common in these patients. The literature supports computed tomographic scanning of the chest with intravenous contrast as the most helpful test to make the diagnosis.⁶⁻¹³ It is considered diagnostic if intravenous contrast enters the esophagus or mediastinum from the left atrium.⁹ Thoracic computed tomography can also reveal pneumomediastinum, pneumopericardium, or the presence of air in the left atrium.^{6,9} Although transesophageal echocardiography had the potential ability to identify the source of microbubbles in our case, it was not considered because any type of esophageal instrumentation, including upper endoscopy, is generally precluded for fear of increasing the

fistula size and worsening the air embolization from insufflation and increased esophageal pressure.^{6,7,9,10}

In general, transthoracic echocardiography is not regarded as a helpful modality for the diagnosis of AEF.^{6,9,10,13} However, in this particular case, it was the main tool that paved the way toward the diagnosis. Echocardiography was ordered on the day of the patient's admission to the hospital, mainly because he was short of breath and had a subdural hematoma. The findings were unremarkable until the patient started coughing intensely. As can be imagined, it is challenging to achieve adequate acoustic windows when a patient is constantly moving because of persistent coughing. However, by firmly maintaining the probe on his chest it was observed that bubbles were suddenly appearing in the left ventricle and quickly disappearing. This was a critical observation, as no bubbles were seen on the right side (Video 2), which made it less likely that the connected intravenous line was the source. Furthermore, it was important to make sure that this was not a case of transpulmonary shunting through an arteriovenous fistula. Therefore, to determine how the bubbles entered the left ventricle, the strategy was to hold the probe on the patient's chest, set up the machine to register retrospectively, increase the number of acquired loops, and wait until the patient had another coughing episode. Immediately after the patient coughed, a significant number of bubbles suddenly appeared in the left ventricle, with a few bubbles also seen in the left atrium and no bubbles in the right chambers. The fact that the left atrium was not as strongly opacified as the left ventricle in the apical four-chamber view may have contributed to a delay in the diagnosis, as one would expect similar opacification of both chambers. A possible explanation would be the fact that the bubbles were originating posteriorly, outside the echocardiographic plane, with the added effect of the pulmonary venous flow rapidly pushing them into the left ventricle.

CONCLUSION

Catheter ablation has become the main treatment for patients with AF who are symptomatic and refractory to pharmacologic treatment. The number of ablation procedures has increased over the years, along with some of its most dreadful complications, such as AEF. It is crucial to be aware of this complication and to recognize its presenting signs and symptoms, which include fever, leukocytosis, neurologic deficits, cough, or hematemesis. Diagnostic tools could include comprehensive blood workup, chest computed tomography to look for pneumomediastinum or to directly visualize the fistula, or, as in our case, transthoracic echocardiography, which can show air bubbles localized to the left heart chambers. It is important to educate patients about this complication and its symptoms. Additionally, close follow-up should be undertaken for the first 2 months following ablation, particularly

if it is believed that a large number of ablation lesions were delivered to the posterior LA wall.

SUPPLEMENTARY DATA

Supplementary data related to this article can be found at <http://dx.doi.org/10.1016/j.case.2017.06.006>.

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