

↓ ATRIO-OESOPHAGEAL FISTULA COMPLICATING CRYOBALLOON WHICH PRESENTED WITH RECURRENT FEVER AND STROKE

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ABSTRACT

In this article, we report a case of atrio-oesophageal fistula (AEF) which occurred after a second-generation cryoballoon intervention was utilised for refractory atrial fibrillation. On the 4th post-procedural week, the patient was admitted to a hospital owing to recurrent febrile episodes, stroke and suspicion of bacteraemia. Sepsis occurred secondary to bacteraemia. Cerebral air emboli and gas bubbles in the left ventricle and mediastinum were shown by radiological images. Emergency surgical

treatment procedures were performed. Although the surgical procedure eliminated the air embolisation and drained the chest, the patient's clinical condition continued to worsen, presenting uncontrollable hemodynamic status and refractory septic shock, resulting death 24 h after the surgery.

Keywords: Atrio-oesophageal fistula, stroke, sepsis, fever, catheter ablation, atrial fibrillation. *Nobel Med* 2018; 14(3): 45-50

KRİYOBALON ABLASYON SONRASI TEKRARLAYAN ATEŞ VE İNME İLE PREZENTE OLMUŞ ATRİYOÖZOFAGEYAL FİSTÜL OLGUSU

ÖZET

Bu olguda, refrakter atrial fibrilasyon ile seyreden hastaya yapılan girişimsel 2. kuşak kriyobalon ablasyon sonrası gelişen atriyoözofageyal fistül vakası bildirilmektedir. İşlemden 4 hafta sonra tekrarlayan ateş ve bakteremi ile hastanemize başvuran hastada kısa sürede nörolojik semptomlar gelişmiştir. Hastanın takibinde bakteriyemi

zemininde sepsis oluştu. Serebral hava embolisi, sol ventrikülde ve mediastinumda gaz kabarcıkları radyolojik görüntüleme aracılığıyla gösterildi. Hasta acil olarak ameliyata alındı. Hava embolizasyonuna neden olan fistülün onarılmasına ve mediastinumun drene edilmesine rağmen hastanın klinik durumu kötüleşmeye devam etti. Kontrol edilemeyen hemodinamik durum ve refrakter septik şokun devam etmesi nedeniyle hasta cerrahi girişimden 24 saat sonra kaybedildi.

Anahtar kelimeler: Atriyoözofageyal fistül, inme, sepsis, ateş, katater ablasyon, atriyal fibrilasyon. **Nobel Med 2018; 14(3): 45-50**

INTRODUCTION

Atrio-oesophageal fistula (AEF) is a rare, but potentially fatal, complication associated with catheter ablation for atrial fibrillation (AF). Following cardiac tamponade, it is the second most frequent fatal complication with a reported incidence of 1 in 500–1000 cases and a mortality of 71%–83%.^{1,2} The causes of death include cerebral air embolism, massive gastrointestinal bleeding and septic shock. There is no consensus regarding the best treatment. The mortality rates of this complication are 96%, 100% and 33% with medical management alone, stent placement and surgical intervention, respectively.³ All evolving techniques may lead to unexpected complications.

Cryoballoon is the latest FDA-approved percutaneous ablation catheter which does not use radiofrequency (RF) energy. However, the long 7-year worldwide usage of the first-generation cryoballoon confirmed the possibility of AEF occurrence, with the first reported case published in 2012.⁴ More recently, a second-generation cryoballoon was developed with improved ability to create continuous lesions and more uniform cooling.⁵

This article describes a case of AEF which occurred 4 weeks after percutaneous circumferential pulmonary vein ablation using a cryoballoon catheter. Our case initially presented with recurrent fever and neurological symptoms.

CASE

A 59-year-old woman with drug-refractory paroxysmal AF underwent second-generation cryoablation in August 2016 at another hospital.

The ablation procedure was performed in all 4 pulmonary veins (PV) using a 28 mm Arctic Front R balloon-catheter, approved by the Food and Drug Administration (FDA) for the treatment of paroxysmal AF. Eight ablation applications were performed: two on each left-sided PV and two on each right-sided PV, with a lowest temperature of -67°C in the left inferior PV (LIPV).

The procedure was uneventful and well-tolerated, and the patient was discharged from the hospital next day. Medications on discharge comprised dabigatran, atenolol and ramipril. Five days after the procedure, the patient developed post-prandial chest pain and swallowing difficulty. Transthoracic echocardiography (TTE) and cardiac biomarkers were unremarkable at that time; additionally, there was no pericardial effusion or evidence of vegetation. Upper gastrointestinal (GIS) endoscopic examination revealed signs of esophagitis and gastric ulcer. She was treated with antacids and proton-pump inhibitors, which improved the symptoms to some extent.

On the 3rd post-procedural week, the patient presented to her local hospital with fever, chills, shivering, sweating, nausea, vomiting, chest pain, elevated white cell count and inflammatory markers. Based on the clinical, laboratory and radiographic examinations, lower respiratory tract infection was diagnosed and oral quinolone and nonsteroidal anti-inflammatory medications prescribed. Meanwhile, the patient travelled to the city where our institution is located and presented to the Yeditepe University Hospital with fever, chills, shivering, elevated inflammatory markers and low platelet counts. The patient was admitted to the hospital owing to recurrent febrile episodes and suspicion of bacteraemia.

Blood samples were taken for cultures and procalcitonin test, and we initiated broad-spectrum intravenous antibiotic (moxifloxacin) therapy. The next day, she developed new-onset weakness in her right extremities, which became acutely aphasic with a right hemiplegia. She soon required intubation and mechanical ventilation owing to rapid and progressive loss of consciousness. Her Glasgow Coma Scale score was 6, blood pressure was 90/50 mmHg and pulse rate was 110 beats per minute. She was immediately transferred to the intensive care unit (ICU).

Early head computed tomography (CT) revealed a mild hypodensity at the posterior side of left centrum semiovale and the subcortical area of the precentral gyrus (Figure 1a). A TTE did not show any evidence of an atrial clot.

Blood cultures were positive for *Streptococcus mitis*, *Streptococcus oralis* and *Capnocytophaga* spp and procalcitonin were 80 ng/ml. She was treated with parenteral carbapenem and linezolid.

Ten hours later, a follow-up CT (Figure 1b) revealed an extensive right cerebral hypodensity and cortical swelling consistent with right parietal infarction, extensive cerebral air emboli and right parenchymal hematoma (arrow). Gas bubbles in the left ventricle (arrows) were shown by repeated transthoracic echocardiography (Figure 2). Chest CT (Figure 3) demonstrated pneumomediastinum (arrows) and air images in the left ventricle (arrowhead).

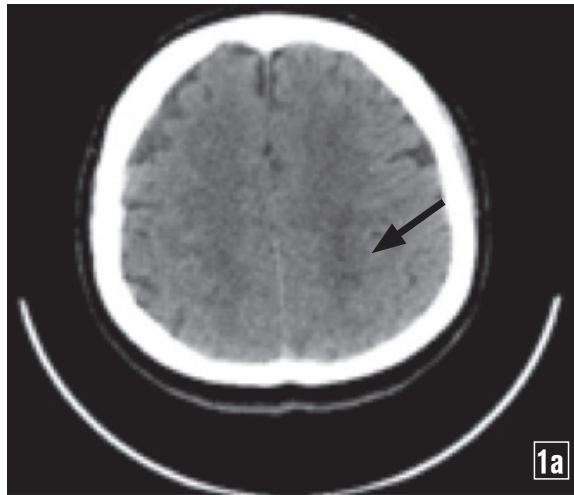


Figure 1a. Brain computed tomography (CT) image. Mild hypodense areas.

Upper GIS endoscopy demonstrated fragile, exudative, ulcerative fistula in the 30th cm of the oesophagus and air bubbles passing into the left atrium and building in the cranial ventricles, worsening her general condition.

At this point, the patient's Glasgow Coma Scale score was 3 and she started to require incremental inotropic agents, but exhibited little hemodynamic improvement. After thorough discussion, among the medical staff and family members, the decision to pursue surgical intervention was made despite its high risk of morbidity and mortality. During the operation there was no spillage in the mediastinum or the pleural space due to dense adhesion between oesophagus and pericardium encircling the

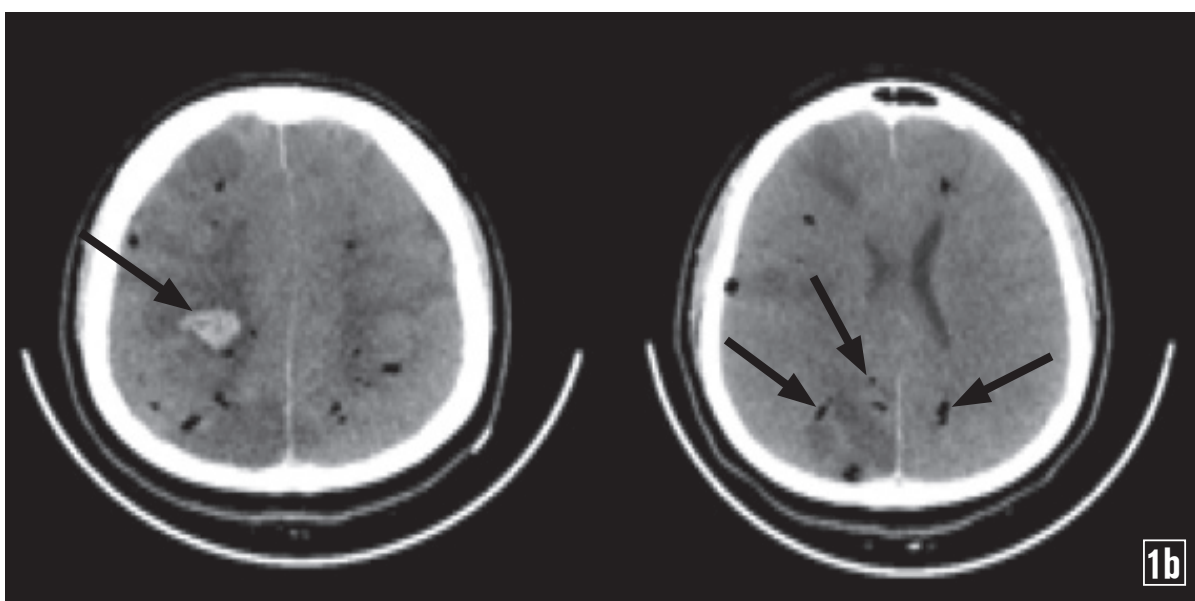


Figure 1b. Control brain CT image, 10 hours later. Right parenchymal hematoma (arrow), cerebral air emboli (arrowheads)



Figure 2. Transthoracic echocardiography. Gas bubbles in the left ventricle (arrows).

fistula. The pericardium was opened by the cardiac surgeons and the atrium isolated and repaired primarily. Then, a 2-cm fistula was exposed on the oesophagus, which was then resected and reconstructed by Ivor Lewis oesophagectomy. The chest was temporarily closed and the patient was laid supine. Her stomach was prepared for reconstruction and a jejunostomy tube was placed. An Ivor Lewis gastroesophageal anastomosis was successfully performed using a 25-mm circular stapler at the level above the azygos vein. The preoperatively-started inotropes were continued during the surgery, and the patient was postoperatively transferred to the ICU.

Although the air embolisation was eliminated and the chest was drained following the surgical intervention, the patient's clinical condition continued to worsen, ultimately resulting in an uncontrollable hemodynamic status and refractory septic shock leading to death 24 h after the surgery.

DISCUSSION

Although according to radiofrequency ablation data AEF complications are less common in cryoablation, there is an increase in AEF cases with the widespread application of cryoablation.

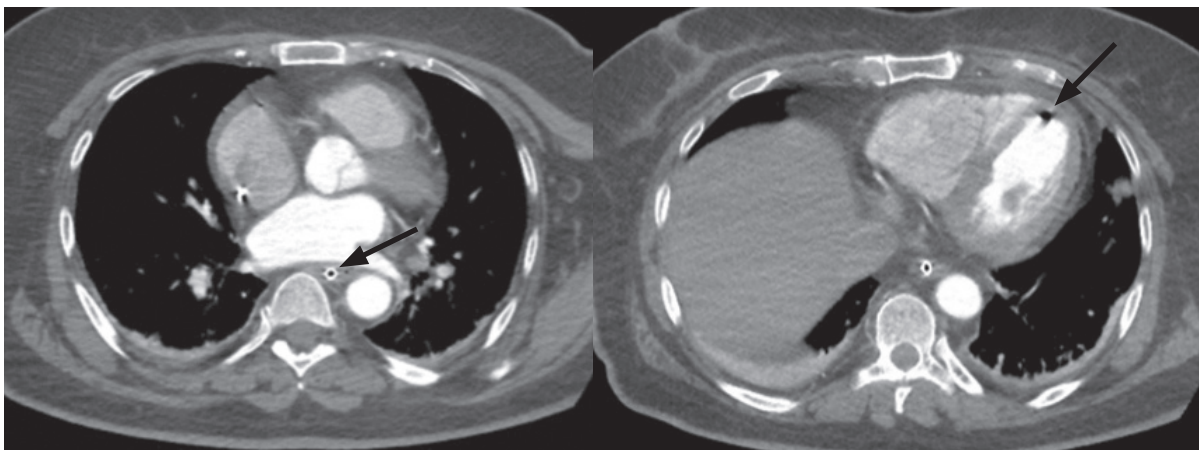


Figure 3. Chest computed tomography images. Pneumomediastinum (arrows) and air images in the left ventricle (arrowhead).

Patients who consent to catheter ablation must understand the risks associated with the procedure. Major complications include cardiac tamponade (1.31%), thromboembolic events (stroke 0.23%, transient ischaemic attacks 0.71%), pulmonary vein stenosis (0.29% requiring dilation), oesophageal injury (AEF 0.04%) and death (0.1 5%).⁶

The clinical presentation of AEF may include fever, chest pain, dysphagia, melena, hematemesis, sepsis and neurological symptoms in the spectrum of mild altered consciousness to coma and paralysis. Our case presented with fever, chest pain, sepsis and neurological manifestations including stroke 4 weeks after ablation.

Blood tests showed leukocytosis, elevation of serum C-reactive protein levels, thrombocytopenia and an elevated erythrocyte sedimentation rate. Blood cultures are frequently positive for bacteria. These laboratory findings were present in our patient during her presentation. A barium swallow study should be avoided with these patients because an intravascular leak of barium could have detrimental consequences. In our case, TEE was not accomplished, but upper GIS endoscopy was performed. The patient's neurological symptoms emerged before the endoscopic procedure. The air embolism to the heart, mediastinum and finally cerebrum was suspected and detected on endoscopy. Early recognition is important, as patients often develop endocarditis with septic emboli leading to neurological manifestations such as altered mental status, seizures and coma within hours of symptom onset.⁷

Endocarditis should be suspected in cases with signs of infection and positive blood cultures after invasive cardiac interventions. In case of radiofrequency ablation (RFA) history, AEF should be strongly considered. Embolic stroke due to recurrent atrial fibrillation may frequently be a differential diagnosis

because atrial fibrillation after RFA may recur and a fistula between the oesophagus and the left atrium may also induce atrial fibrillation.⁸ Patients with AEF frequently present with transient ischaemic attacks or stroke. An atrio-oesophageal fistula should be strongly considered if the complaints are associated with fever or occur after meals.

There are some measures which can minimise the risk of oesophageal injury. Gastroesophageal reflux may play a role in aggravating the initial oesophageal damage and hinder appropriate healing, thereby promoting the development of AEF; consequently, prophylactic proton-pump inhibitors (PPIs) have been recommended for patients undergoing AF ablation.⁹ In our case, there was no use of PPIs before and early after the procedure, but PPIs and antacids were given a week after the detection of esophagitis and peptic ulcer. PPIs are extremely effective at reducing gastric acidity, allowing for healing of oesophagitis and reducing the size of iatrogenic ulcers.

CONCLUSION

Currently there is no AF ablation method which is 100% free of complications or risk of AE fistula. Consequently, more open reporting is necessary when these rare, but potentially fatal, complications occur. The results of our case presentation confirm the high mortality rate associated with AEF, following left atrial ablation and the necessity for diagnosing atrio-oesophageal injury in a timely manner. Atrio-oesophageal injury and subsequent AEF is an infrequent but potentially fatal complication of atrial ablation. We recommend that cases should be followed closely for infections, gastrointestinal disturbance and neurological symptoms within 2 months after cryoablation procedure.

*The authors declare that there are no conflicts of interest.



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