

Treatment strategy for treating atrial-esophageal fistula: esophageal stenting or surgical repair?

A case report and literature review

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Abstract

Introduction: Atrial-esophageal fistula (AEF) is a rare severe disease, which may be associated with radiofrequency catheter ablation (RFCA) of atrial fibrillation (AF) or intraoperative radiofrequency ablation of atrial fibrillation (IRAAF).

Clinical Findings: We reported a case of a 67-year-old man with AEF following RFCA of AF, who treated with esophageal stenting and surgical repair.

Outcomes: He was attacked by out-of-control sepsis and infectious shock after surgery and died.

Literature review: We analyzed 57 relevant articles about AEF from 2003 to 2015 by searching PubMed database. According to literatures, the most common symptoms were fever, rigor, sepsis, and neurologic symptoms. Chest computer tomography (CT) and contrast enhanced CT may be the reliable noninvasive diagnosis methods because of high sensitive for AEF.

Conclusion: Make a definition diagnosis in time with early primary surgical repair may save their lives. Conservative treatment or esophageal stenting alone may not be a better choice for AEF patients.

Abbreviations: AEF = atrial-esophageal fistula, AF = atrial fibrillation, CECT = contrast-enhanced computer tomography, CT = computer tomography, ECG = electrocardiogram, EGD = esophagogastroduodenoscopy, ET = exploratory thoracotomy, GI = gastrointestinal, hsCRP = high sensitivity C reactive protein, IRAAF = intraoperative radiofrequency ablation of atrial fibrillation, LA = left atrium, MRI = magnetic resonance imaging, NT-proBNP = N-terminal pro brain natriuretic peptide, RFCA = radiofrequency catheter ablation, WBC = white blood count.

Keywords: atrial fibrillation, atrial-esophageal fistula, catheter ablation, esophageal stenting, surgical repair

1. Introduction

Atrial-esophageal fistula (AEF) is a rare severe disease, which may be associated with radiofrequency catheter ablation (RFCA) of atrial fibrillation (AF) or intraoperative radiofrequency ablation of atrial fibrillation (IRAAF). We presented a case of a 67-year-old man with AEF following RFCA treated with esophageal

stenting and surgical repair, and then reviewed literatures about AEF to explore the appropriate treatment options.

2. Case presentation

A 67-year-old man, who had coronary heart disease, AF, and heart failure, was admitted because of dyspnea and rapid AF. The symptoms improved after treating by diuretics, milrinone, and nitrate. Then RFCA of AF was performed to perform wide area circumferential ablations to achieve pulmonary vein isolation using a 3.5 mm tip electrode on a D-curved thermocool catheter through Agilis sheath. The lesions were applied for 30 to 60 s with 30 to 30 W. The patient discharged from the hospital 3 days after the procedure.

But 18 days after the procedure, the patient was admitted to local hospital with complaint of chest discomfort and fever for 1 day. His manifestation was confusion, gazing upwards, mild limb seizures, and vomiting. His temperature was 41°C, white blood count (WBC) $5.2 \times 10^9/L$, and high sensitivity C reactive protein (hsCRP) 18.56 mg/L. Electrocardiogram (ECG) showed supraventricular tachycardia.

Then he presented to emergency department of our hospital after hypothermia therapy. At admission, physical examination revealed that his temperature was 37.9°C, blood pressure was 99/73 mm Hg, and pulse rate was 78 beats/min. ECG showed AF. The initial laboratory studies showed N-terminal pro brain natriuretic peptide (NT-proBNP) 1550.00 pg/mL, alanine aminotransferase 21 U/L, cardiac troponin I 3.95 μg/L, WBC $10.58 \times 10^9/L$, neutrophil 97.2%, hsCRP 26.0 mg/L, and fecal occult blood 3+. Cerebral and pulmonary computer tomography

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BZ, HZ, and Y-HD planned the article and contributed to data collection, discussing content, and writing and reviewing the article. X-JC, L-YQ, and JP participated in its design, study supervision, and helping to writing the article.

Informed patient consent was obtained for publication of this case report.

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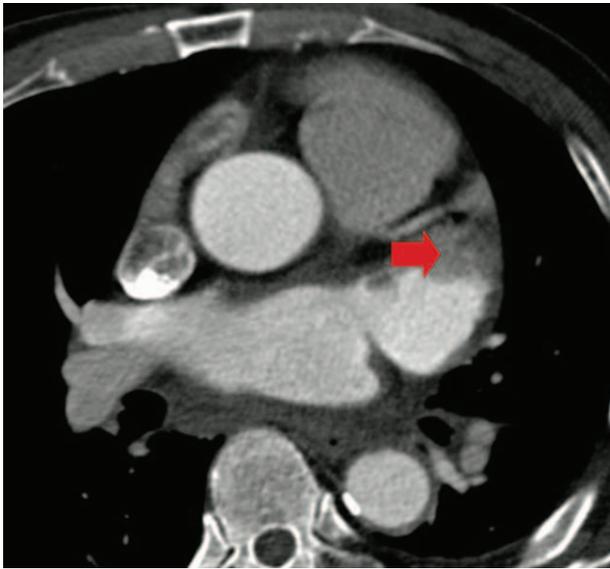


Figure 1. Contrast-enhanced computed tomography showed suspicious thrombus in left atrial appendage (indicated by red arrow).

(CT) did not reveal any abnormal findings. Contrast-enhanced CT (CECT) showed suspicious thrombus in left atrial appendage (Fig. 1).

Patient was asked to fast with gastrointestinal (GI) decompression, and used antibiotic, imipenem/cilastatin and linezolid, to control infections. He regained consciousness from the next day after admitting our institution, but he still had repeated epileptic seizures. Cerebral magnetic resonance imaging (MRI) was performed and showed lacuna infarctions (Fig. 2). Emergency esophagogastroduodenoscopy (EGD) showed a small and deep ulcer at 30 cm from incisors. The esophagus was stented with self-expanding, polyester-covered, length 100 mm, width 18 mm (Nanjing Micro-Tech, Nanjing, China) (Fig. 3). In the following days, WBC counts and hsCRP level decreased, epileptic seizures went away. But the symptom of acid reflux occurred from the 5th day after esophageal stenting when he ate fluid food. Temperature and WBC counts increased again from the 7th day. Cardiac CECT showed esophageal-mediastinum fistula with encapsulated effusion and pneumatosis behind left atrium (LA),

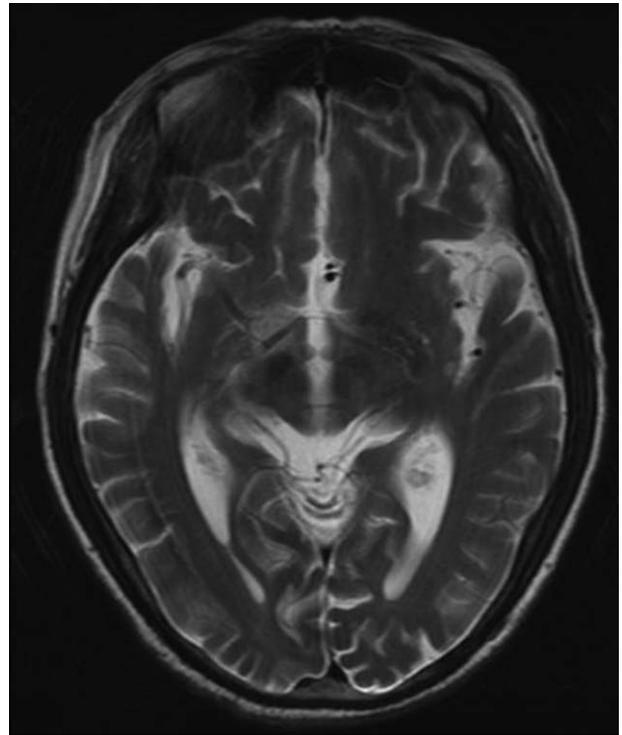


Figure 2. Cerebral magnetic resonance imaging showed lacuna infarctions.

and post wall of LA depression. Mediastinal abscess and AEF were considered and surgical repair was planned.

Right thoracotomy incision through the right 5th intercostal space was made under general anesthesia. During the surgery, we noticed the membranaceous and funicular adhesions between the right lung, diaphragm and mediastinum, white lamellar scars under azygos vein beside left atrial. After opening mediastinal pleura, tight adhesion between esophagus and pericardium was found and separated carefully, then a 6 mm diameter fistula in the anterior wall of esophagus was discovered (Fig. 4). Tight adhesion between LA and pericardium, combined with bleeding at the posterior wall of LA, was found during exploratory pericardiotomy. Rid-muscle flap pedicled with vessels was made to repair LA posterior wall and fistula between esophagus and LA

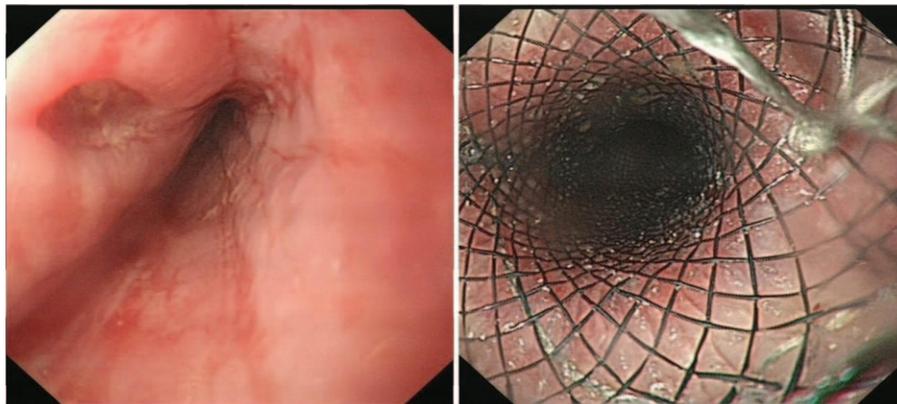


Figure 3. Emergency esophagogastroduodenoscopy showed a small and deep ulcer at 30 cm from incisors (left). The esophageal stent had been implanted (right).

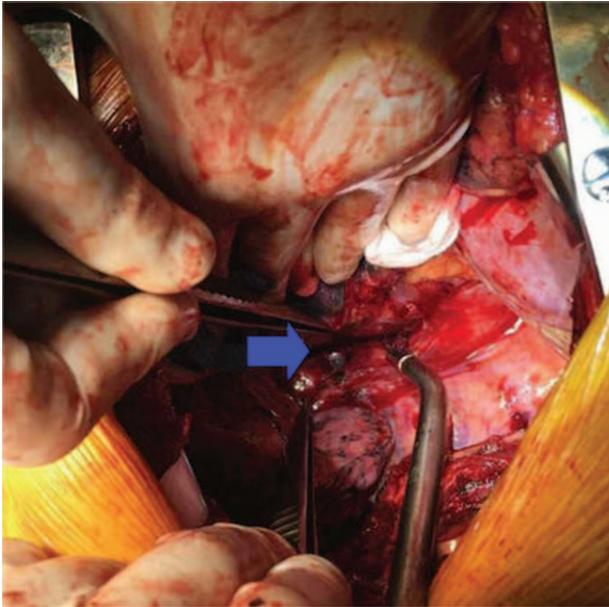


Figure 4. A 6 mm diameter fistula (indicated by blue arrow) in the anterior wall of esophagus.

(Fig. 5). Pleuroclysis using povidone iodine and nature saline was performed before sternal closure.

After surgery, the patient was transferred to intensive care unit, and then out-of-control sepsis and infectious shock, which showed repeated fever and chills, metabolic acidosis, hypotension, tachycardia and oliguria, attacked the patient, and caused his death.

3. Discussion

AEF, a rarely complication of RFCA of AF, was first reported in 2004.^[1] Some surveys revealed that the incidence was 0.03% to



Figure 5. Rib-muscle flap pedicled with vessels (indicated by blue arrow) was used to repair LA posterior wall and fistula between esophagus and LA. LA= left atrium.

Table 1	
Characteristics of patients.	
Characteristics	Value
No. of cases, n	86
Male gender, n	60
Age (mean ± SD), y	56.3 ± 12.2
Procedure	
Catheter ablation, n	78
Surgical, n	8
Mean timing of presentation (range), d	24.8 (2–60)
Outcome (survived), n/n	36/86

SD = standard deviation.

0.04% in RFCA of AF and about 1% in IRAAF, and its mortality ranging was from 71% to 83%.^[2–4] As the number of RFCA of AF cases grows, more AEF cases could be found in recent years. It was considered that the ablation of the posterior wall of LA by the high temperature might be an important risk of AEF.^[5] Elevated esophageal temperatures during LA ablation may occur in more than 50% of patients.^[5,6]

Esophageal stenting and surgical repair are the commonest treatment for AEF. To date, we still did not know which one is the best treatment for AEF. So we searched PubMed database about AEF from 2003 to 2015. Fifty-seven relevant articles, including 86 AEF patients (60 males), were analyzed.^[1,6–61] Seventy-eight patients were secondary to RFCA of AF, and 8 patients were secondary to IRAAF. The incubation period of AEF symptoms was from 2 to 60 (average: 24.8) days. The mortality of the patients was 58.1% (Table 1).

The most common symptoms were fever, rigor, and sepsis. The symptoms of neurological symptoms, including hemiplegia and epilepsy, consciousness disorder, GI bleeding, chest pain, were relatively common. The other uncommon symptoms included: nausea and vomiting, dysphagia, dyspnea, cough, arrhythmia, headache, blindness, syncope, abdominal pain, and petechiae (Fig. 6).

Chest CT and CECT were the most common noninvasive diagnosis methods with high sensitive for AEF, which could show the air bubbles or vegetation at the site near the esophagus of LA. Sometimes mediastinum emphysema could be found. EGD was a

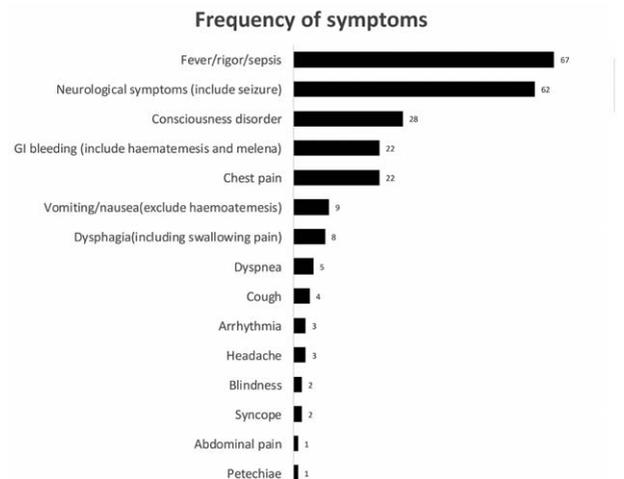


Figure 6. Frequency of symptoms at the time of hospitalization.

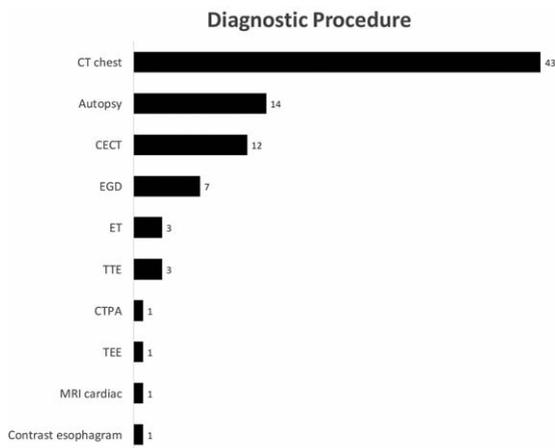


Figure 7. Diagnostic procedure at the time of hospitalization. CECT = contrast enhanced computed tomography, CTPA = computed tomography pulmonary angiogram, EGD = esophagogastroduodenoscopy, ET = exploratory thoracotomy, TEE = transesophageal echocardiography, TTE = transthoracic echocardiography.

common diagnostic way too. In some cases, AEF was only diagnosed during autopsy or exploratory thoracotomy. Transthoracic echocardiography, transesophageal echocardiography, cardiac MRI, and contrast esophagram could discover AEF (Fig. 7). Abnormal cerebral CT findings, such as cerebral air embolism and multifocal cerebral ischemia, revealed in most of patient on admission.

Hemorrhage from AEF, sepsis, air embolism, and cerebrovascular accidents are the major reasons for a fatal outcome.^[2,6,9,12,23,57] Esophageal stenting and surgical repair were the major therapies of AEF. Different therapeutic strategy might influence outcome of patients. Twenty-eight patients without esophageal stenting or surgical repair had higher mortality, only 1 patient who performed endoscopic ligation survived. Ten patients who treated with esophageal stenting all died. But relatively, surgical repair decreased the mortality of the patients significantly, only 12 patients died in 43 patients who only performed surgical. Three patients implanted esophageal stenting after performing emergency atrial wall repair with thoracotomy, 2 patients survived. Two patients, who received esophageal stenting and subsequently performed surgical, were all survived (Table 2).

It is known to all that AEF is a life-threatening complication of ablation of AF. An appropriate treatment is the key to reduce mortality of patients with AEF. According to the literature review of AEF, we found that conservative treatment or esophageal stenting alone seemed to be a poor clinical outcomes, and surgical repair combined with or without esophageal stenting may be the preferred option for the patients. Early surgical treatment have lower mortality than delayed strategy.^[8,19,23,26,37] Though there were many disputes about the opportunity to put esophageal stent, stenting alone was not recommended for treating AEF.^[26,37] The clinical outcomes of combination therapy of esophageal stenting and surgical repair were not clear because of few cases.

In our case, the patient underwent early esophageal stenting with temporary improvement of the symptoms in the first few days. But the conditions of the patient were worse, and eventually caused death despite surgical repair was performed. Early

Table 2

Treatments and outcomes.

Intervention for AEF		Death	Survived	No sequels
Stent (+)	Surgical (-)	10	0	
Stent (-)	Surgical (+)	12	30	1
Stent (-)	Surgical (-)	27	1*	
Stent (+)	Surgical (+) [†]	1	2	
Stent (+)	Surgical (+) [‡]	0	2	

AEF = atrial-esophageal fistula.
 * Underwent endoscopic ligation.
 † Primary surgical repair.
 ‡ Primary esophageal stenting.

resuming oral intake and delayed surgical intervention may be the important factors of relapse.

4. Conclusion

Chest CT and CECT are high sensitive noninvasive diagnostic methods for AEF. Make a definition diagnosis in time with early primary surgical repair may save their lives. Conservative treatment or esophageal stenting alone may not be a better choice for AEF patients.

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