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CASE REPORT
Cardiovascular imaging

A case report of rapid diagnosis of atrial–oesophageal fistula by transthoracic echocardiogram with agitated saline injection

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Background Catheter ablation is a common treatment for atrial fibrillation (AF). Atrial–oesophageal fistula (AOF) is a rare yet fatal complication of catheter ablation. Chest computed tomography (CT) is the diagnostic modality of choice but may be undiagnostic in up to 24% of cases.

Case summary We present the case of a 61-year-old male who presented with pleuritic chest pain, hypotension, fever, and coffee-ground emesis 20 days after cryoablation for AF. His chest CT was undiagnostic. Atrial–oesophageal fistula was diagnosed by injecting agitated saline into the nasogastric tube during a transthoracic echocardiogram (TTE) that showed bubbles in the left atrium and ventricle.

Discussion In the case presented, as often happens, the diagnosis of AOF was delayed for several days, during which the patient presented with septic shock and concomitant multiorgan failure. The high mortality rate associated with AOF is partially attributable to delayed diagnosis. As prompt surgical intervention offers the best chance of survival, a high level of suspicion is of the utmost importance. We suggest contrast-enhanced TTE as a potential diagnostic tool when a rapid and definitive diagnosis is crucial and CT is inconclusive. Since this procedure is not without risk, proper risk consideration and management are necessary.

Keywords Agitated saline • Atrial fibrillation • Atrial–oesophageal fistula • Case report • Catheter ablation • Contrast-enhanced echocardiogram

ESC Curriculum 2.1 Imaging modalities • 2.2 Echocardiography • 6.4 Acute heart failure • 5.3 Atrial fibrillation • 2.4 Cardiac computed tomography

Learning points

- Atrial–oesophageal fistula (AOF) is a rare though fatal complication of atrial fibrillation ablation. Patients may present with neurological, gastrointestinal, and cardiac symptoms. Prompt diagnosis is crucial. Surgical repair is the treatment of choice.
- Contrast chest computed tomography (CT) is the gold standard for diagnosis, though up to 13% of initial CT scans are undiagnostic.
- Transthoracic echocardiogram with agitated saline injected threw the nasogastric tube into the oesophagus may serve as a diagnosis modality of AOF in patients with undiagnostic CT.

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Introduction

Atrial fibrillation (AF) is the most commonly sustained arrhythmia in clinical practice, and its global prevalence and incidence are rising.[1]

The utilization of catheter ablation as a treatment for AF is increasing significantly.[2] The periprocedural complication rate of AF ablation is estimated at 5%, and death occurs in 0.1% of patients.[2] Among the leading causes of death are cardiac tamponade, stroke, and atrial–oesophageal fistula (AOF).[2]

Although rare, AOF is often fatal. The gold standard for diagnosis is chest computed tomography (CT), though first CT may not be diagnostic in up to 24% of cases.[3]

We present a case of AOF in a haemodynamically unstable and rapidly deteriorating patient with an undiagnostic CT scan. The diagnosis was made at the bedside using transthoracic echocardiography (TTE) and an injection of agitated saline through a nasogastric tube.

Case presentation

A 61-year-old male presented to the emergency department (ED) with pleuritic chest pain. Ten days earlier, he underwent pulmonary vein isolation (PVI) by cryothermal ablation for treatment of paroxysmal AF at another hospital.

His medical history included diabetes mellitus, hypertension, and AF, for which he was treated with empagliflozin, dabigatran, and bisoprolol. His physical examination was unremarkable except for a pericardial friction rub. His electrocardiogram (ECG) showed diffuse ST-elevation with a concave pattern and PR depression. Laboratory results, including troponin levels, were within normal limits. Transthoracic echocardiogram demonstrated preserved ventricular function and a small to medium circumferential pericardial effusion (*Figure 1*). He was hospitalized with a presumptive diagnosis of pericarditis and promptly treated with 0.5 mg/kg/day of prednisone and 0.5 mg colchicine twice daily. Clinical improvement followed, and after 4 days, he was discharged with a recommendation to complete the course of the treatment and follow-up in the community.

Six days after discharge, he presented to the ED again with severe weakness and fatigue, a single syncopal episode, blood-tinted vomitus, and pleuritic chest pain. His blood pressure was 80/50 mmHg, his temperature 38.5 °C, and his oxygen saturation 96% in room air. Laboratory examination revealed haemoglobin of 11.2 gr/dL (normal range 12–16 gr/dL), mild neutrophilia of 9.23 10^3/uL (normal range 1.9–8 10^3/uL), and a creatinine level of 1.72 mg/dL (normal range 0.5–0.95 mg/dL) with a high urea concentration of 114.1 mg/dL (normal range 17–43 mg/dL). Troponin levels were normal. A nasogastric tube was inserted and drained a large amount of coffee-ground matter with no evidence of active bleeding. Bedside TTE showed no differences from the previous study. The presumptive diagnosis was mediastinitis due to AOF following PVI cryoablation. The patient received 2000 cc of intravenous fluids, 0.5 mcg/kg/min of norepinephrine, and broad-spectrum antibiotics: amoxicillin clavulanate (1 g tid) and fluconazole 400 mg once daily. Non-gated CT angiography demonstrated multiple mediastinal lymph nodes and a small amount of fluid in the mediastinum and pericardium without extravasation of contrast medium from the left atrium into the oesophagus and with no pneumomediastinum or pneumopericardium (*Figure 2*). Brain CT showed no focal lesion.

During the next several hours, the patient developed multiorgan failure, his haemoglobin level declined to 9.7 mg/dL, and his creatinine level rose to 1.83 mg/dL. He was intubated and mechanically ventilated, yet there was still no definitive proof of AOF. At this time (the day after his second admission), a third TTE showed the same amount of pericardial effusion, preserved biventricular function, and no air in the cardiac cavities (Clip 1). The study was completed by injection of 10 mL of agitated saline through the nasogastric tube, which was retracted to the mid-oesophagus in order for the tip to be at the presumptive level of the atrium. Immediately after the injection, bubbles appeared in the left atrium and left ventricle, confirming the diagnosis of AOF (*Figure 3*/Clip2).

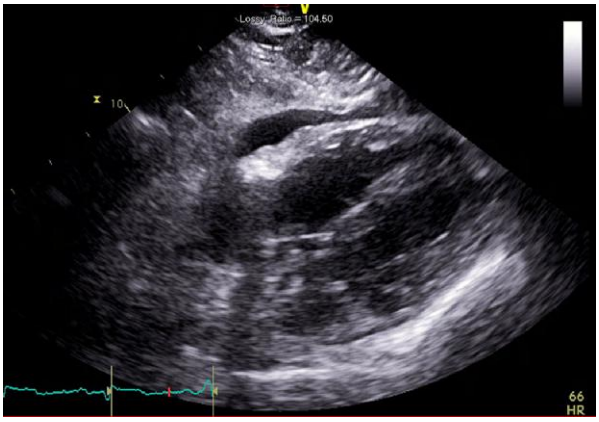

Figure 1 Subcostal view demonstrating a small to moderate amount of pericardial fluid located mainly in the anterior area of the pericardium.

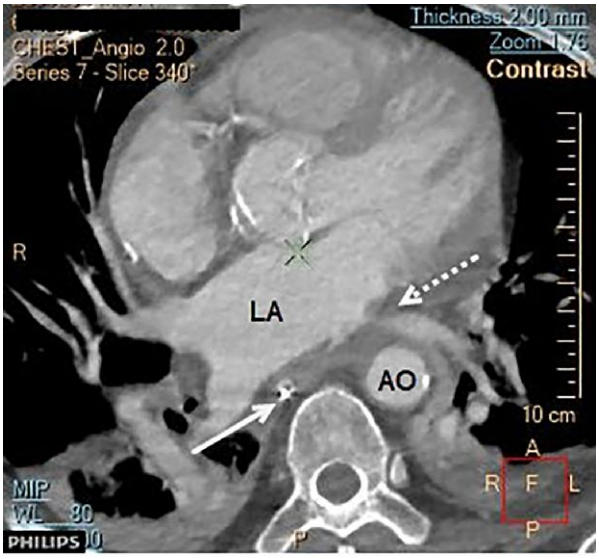

Figure 2 Axial imaging of a non-gated chest computed tomography angiogram. The computed tomography shows left inferior pulmonary vein stenosis (interrupted arrow), probably as consequence of the recent cryoablation procedure, and a small amount of pericardial fluid. The continuous arrow indicates the location of the nasogastric tube inserted into the oesophagus. AO, descending aorta; LA, left atrium.

With a definitive diagnosis of AOF, the patient was transferred to the operating room and underwent surgical repair of a left atrium necrotic rupture in the posterior wall and adjacent to the left inferior pulmonary vein using a pericardial patch. Gastroscopy was performed within 24 h for definitive closure of the fistula from the oesophageal side. On examination, a 12 mm tear was found in the low oesophagus and closed with four endoclips. The postoperative period was characterized by

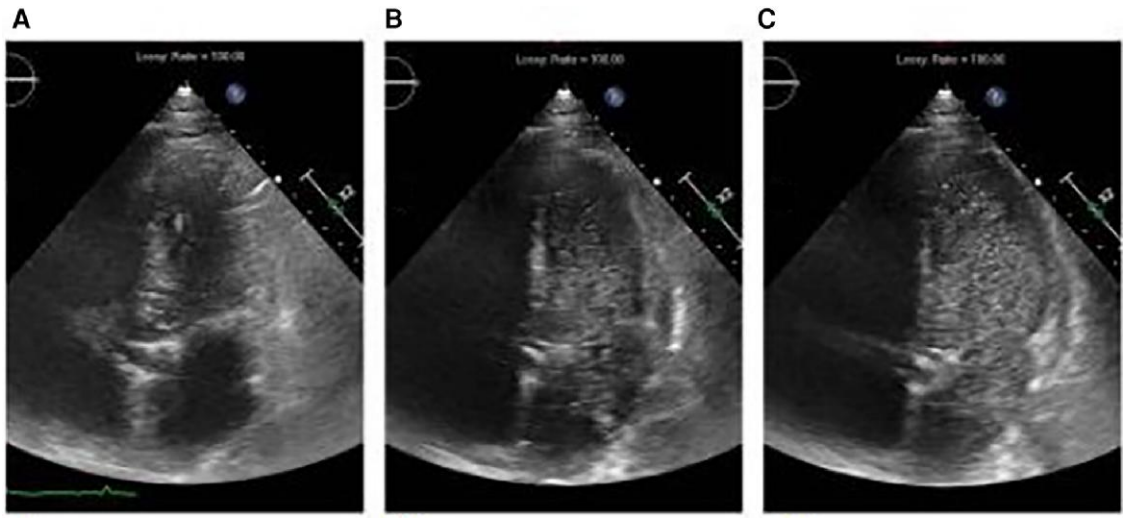

Figure 3 Three sequential images from the apical four-chamber views. *(A)* Before the injection of agitated saline through the nasogastric tube. *(B)* Immediately after the injection, bubbles appear in the left atrium and ventricle. It can be clearly seen that the bubbles are entering the left atrium from the posterior aspect of the roof. *(C)* The bubbles fill the left atrium and ventricle.

high-grade fever and haemodynamic instability. The patient was treated with meropenem (1 g tid) and 100 mg micafungin per day due to positive polymicrobial (*Streptococcus constellatus*, *Candida glabrata*, and *Candida albicans*) pleural fluid cultures.

After 30 days of hospitalization, the patient was discharged for rehabilitation.

Discussion

Although rare, AOF is a fatal complication of PVI.[4]

Time to clinical presentation may vary, ranging from 0 to 60 days after the procedure (median 21 days).[3] Usually, the patient seeks medical attention more than once before a definitive diagnosis is reached.[4]

Over 70% of patients develop fever.[5] Neurological (72%), gastrointestinal (GI) (41%), and cardiac (40%) symptoms are frequent as well.[5] Gastrointestinal symptoms include upper GI bleeding, dysphagia, nausea, and vomiting.[5] Cardiac symptoms include chest pain, dyspnoea, and palpitations.[5] Electrocardiogram abnormalities are found in 45% of patients.[5] Leukocytosis and elevated CRP and troponin are also common. Infection with oral cavity organisms may occur in up to 96% of cases.[6] The high mortality rate of AOF is at least partially attributable to late diagnosis.[7] Surgical repair is considered the standard of care, yet mortality rates remain high (33%).[8] A high level of suspicion is the key to early diagnosis, as prompt surgical intervention can be lifesaving.[5,7,9]

The modality of choice for diagnosing AOF is chest CT angiogram, which may show free air in the left atrium or the pericardium, or both (56%), AOF tract (12%), atrial wall abnormalities (7%), oesophageal perforation (5%), mediastinal inflammation (1%), or left atrium thrombus.[7] However, up to 24% of cases may show no abnormalities on CT, especially in the early phase of AOF.[5,7,10] In a systematic review by Han et al., the initial CT scan was undiagnostic in 7% of 120 confirmed cases of AOF.[5] Della Rocca et al. found that the initial chest CT had unremarkable findings in 24% (42 out of 192) of patients with AOF.[10] Repeated scans are often needed to establish the diagnosis. Magnetic resonance imaging may be more sensitive than CT but is not the modality of choice, especially in unstable patients.[7] Brain CT or MRI may be useful in high-suspicion cases. Multi-territory infarcts, pneumocephalus, and intravascular air are the most common findings.[7]

Other less commonly used diagnostic methods such as oesophagram and explorative thoracotomy are highly sensitive but not exempt from adverse events. Urgent endoscopy and transoesophageal echocardiogram (TOE) in these patients may result in clinical deterioration and should be avoided when AOF is suspected due to the potential of air embolization during oesophageal insufflation. Transoesophageal echocardiogram may also increase fistula size and haemorrhage.[4,5,7,9,11–14]

Transthoracic echocardiogram is feasible and may identify features that suggest AOF, such as echogenic material in or around the left atrium, pericardial effusion, and pneumopericardium. However, due to its high rates of false negatives, non–contrast-enhanced TTE should not be used as a first-line imaging tool for AOF diagnosis.[5,9,12] Angulo et al. described spontaneous bubbles, seen intermittently, in the left cardiac chambers during a TTE in an AOF patient who was coughing.[15] To prevent iatrogenic air embolism, Rong et al. used TTE to monitor carbon dioxide insufflation during an oesophageal endoscopy for AOF diagnosis. Bubbles visualized in the left atrium confirmed the diagnosis, and the study was terminated.[16]

In the case presented, as frequently occurs, the diagnosis of AOF was delayed for several days, during which the patient deteriorated into septic shock. Although mediastinitis due to AOF was the leading assumption, CT did not provide a definitive diagnosis, as often happens in these cases. Due to the patient's rapid deterioration and multiorgan failure, other diagnostic modalities, namely cardiac MRI and repeated CT, were impractical.

Considering AOF's extremely high mortality rate, prompt surgical intervention offers the best chance of survival. However, in this case, surgeons were reluctant to operate until a definitive diagnosis was made. Another TTE using an agitated saline injection in the oesophagus was attempted to make a definitive diagnosis, and the diagnosis was confirmed. This procedure is not without risk as it may cause air embolization or push contaminated material into the circulatory system, though the risk is probably low due to the small amount and low pressure of the fluid injected into the oesophagus. The injection through a peripheral vein of microbubbles generated by agitating 9 mL of saline

solution and 1 mL of air in a syringe has been proven safe in the diagnosis of intracardiac shunt.[16]

Contrast-enhanced TTE is a potential rapid diagnostic tool for AOF and may be used when CT is inconclusive and following proper risk consideration and management.

Lead author biography

Dr Ori Galante has graduated with honours from medical school at faculty of health in the Ben Gurion university. He is a specialist in internal medicine since 2011 and in intensive care medicine since 2014. He is the head of the extracorporeal membrane oxygenation (ECMO) service and the head of resuscitation committee in the Soroka Medical Center in Beer-Sheba Israel.

Consent: The patient reported in this case is deceased. Despite the authors' best efforts, they have been unable to contact the patient's next of kin to obtain consent for publication. Every effort has been made to anonymize the case. This situation has been discussed with the editors.

Conflict of interest: All authors declare they have no conflict of interest.

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Data availability

No new data was generated or analysed in support of this research.

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