

CASE REPORT

Successful management of left-atrial oesophageal fistula following radiofrequency ablation

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Abstract

We present the case of a 67-year-old patient who presented to the ICU with fever and neurological deficits, mimicking a cerebrovascular accident, due to a left atrial oesophageal fistula (LAOF) after surgical radiofrequency ablation for atrial fibrillation. Due to the relative rarity of the complication and the diverse clinical presentation of this condition, both the diagnosis and management of LAOF is challenging.

Introduction

Atrial fibrillation (AF) is the most common form of tachyarrhythmia. If pharmacotherapy is unsuccessful, radiofrequency ablation (RFA) is the next treatment of choice.^[1] A rare but lethal complication is left atrial oesophageal fistula (LAOF). Recognition by physicians is paramount, because early diagnosis and management can reduce morbidity and mortality.^[2]

Case description

A 67-year-old man was admitted with unexplained collapse and AF. His history included therapy-resistant AF, catheter-guided RFA five months earlier and surgical RFA five weeks prior to admission. The postoperative period was complicated by symptomatic pericardial effusion (10-15 mm), which was drained (400 ml). Blood cultures were negative.

Physical examination revealed an irregular pulse of 180 beats/min and fever of up to 39°C. Laboratory findings only showed an elevated C-reactive protein (CRP) (83 mg/l) without leucocytosis. Transthoracic echocardiogram showed normal ventricular function, bi-atrial dilation, no valve anomalies and no vegetation. Several hours later the patient developed symptoms of stroke with left-sided hemiparesis, hemianopsia, hyperreflexia and bilateral plantar reflex according to Babinski. A computed tomography (CT) scan of the brain showed no signs of haemorrhage, infarction or vascular occlusion. Subsequently the patient developed focal epilepsy, periods of apnoea and hypotension, for which he was transferred to the intensive care unit (ICU).

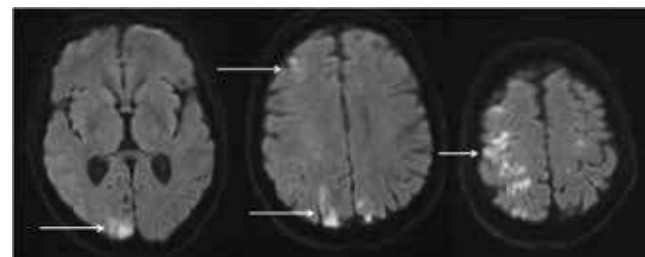


Figure 1. Diffusion-weighted images of MRI scan of the brain revealing recent ischaemia of the right and left hemispheres depicted by arrows (white, hyperintense lesions)

Due to positive blood cultures with *Streptococcus* species, a cardiac embolism source was suspected. Brain magnetic resonance imaging (MRI) revealed multiple areas of diffusion restriction confirming recent ischaemia in both hemispheres, suggestive of a cardiac embolic source (*figure 1*). Transoesophageal echocardiogram (TEE) showed no signs of endocarditis or endocardial thrombus formation. Chest CT scan showed signs of oesophageal perforation and mediastinitis, as well as an air bubble in the apex of the left ventricle (*figure 2*). The combination of the patient's history and clinical signs of a cardiac embolism source suggested an LAOF. He was transferred to the primary cardiac surgery centre which had performed the RFA. Initially a thoroscopic reconstruction of the left atrial roof was performed, as well as endoscopic placement of two clips on the oesophageal lesion during oesophagoscopy. Postoperatively, the patient was admitted to the ICU for supportive care, antibiotic treatment and a nil by mouth diet for the following two weeks, after which an oesophageal stent was inserted during oesophagoscopy. The postoperative course was prolonged due to severe mediastinitis, recurring cerebral embolism, slow neurological recovery and stenting procedures. Two months after the initial oesophageal stent placement, another stent was placed over the initial stent due to stent displacement. Finally, reconstruction of the oesophagus with the intercostal muscle flap was successfully performed six months after the initial diagnosis. At discharge the

patient had somewhat recovered. He had succeeded in gaining weight. AF was no longer present, but he is now suffering from focal epilepsy. The strength in his right arm and leg had recovered completely, strength in the left arm is now diminished. He is now able to walk by himself without supportive aids.



Figure 2. Chest CT scan depicting an air bubble in the left ventricular apex, depicted within the circle

Epidemiology

Ablation therapy for AF can be performed as a catheter-guided procedure or surgically. As we are seeing a rise in the number of procedures, an increase in the number of atrial-oesophageal fistulas is also observed. The incidence of reports of LAOF following surgical AF ablation is low. From 1990 to 2005 the incidence of AF rose from 270,000 to 2.2 million. During this period there was a 15% increase in RFA procedures, resulting in an increase in reported LAOF from 0.06% to 0.79%.^[2]

Pathophysiology

During RFA, dysfunctional conductive tissue surrounding the four pulmonary veins is ablated to isolate AF triggers using heat generated from medium frequency alternating electric current. The proposed pathophysiological mechanism of LAOF is through RFA inducing high temperatures in the oesophageal wall (>39°C).^[3] Patient-related factors include small body size, resulting in a thinner posterior wall of the left atrium,^[4] atrial enlargement,^[5] or the absence of a sufficient fat layer between the left atrium and the oesophagus.^[6] Procedural factors include the use of a TEE probe during the procedure. The probe may displace the ventral oesophageal wall towards the left atrium which increases heat transfer to the tissue.^[7] High temperatures in the oesophageal wall cause ulceration extending into the muscular and vascular tissue, compromising blood flow to the affected area and impairing wound regeneration.^[3,8-10] Due to the minimal distance between the posterior left atrial wall and the oesophageal lumen (5 mm), there is a high risk of perforating

the oesophageal wall, pericardium and left atrial wall, creating a fistula. The intraluminal oesophageal pressure is higher than the intra-atrial pressure during swallowing, retching and coughing,^[11] forcing oesophageal contents into the left atrium, thus entering the arterial circulation.^[2,12]

Clinical findings and radiology studies

LAOF can present between two days to six weeks after RFA. Presenting symptoms include fever (83%), neurological deficits (51%), haematemesis (36%) and an altered mental state (28%).^[2] Neurological manifestations of LAOF include stroke (58%), transient ischaemic attacks (TIAs) (11%), and seizures (17%).^[2,13] Chest CT scan showing pneumomediastinum or air bubbles in the left atrium is the most suggestive of LAOF. Brain imaging may show multifocal ischaemic lesions. Transthoracic echocardiogram or chest CT may show pericardial effusion, although this is not a common finding.^[2,14]

Prevention of LAOF

Proposed methods to prevent LAOF include modulation of power (Watt) and time of RFA, orientation of the ablation catheter, monitoring of the oesophageal temperature and three-dimensional electroanatomical mapping of the left atrium and oesophagus. In procedures using TEE it is important to retract the probe during ablation to prevent displacement of the oesophageal wall.^[2,9] TEE probes also may act as antennae drawing energy to the oesophageal wall. Additionally prolonged TEE usage increases the risk of pressure-related tissue necrosis.^[7]

Management of LAOF

Surgical management of LAOF has been reported to be most successful. It is generally advised to refrain from oesophagoscopy and stenting to minimise the risk of intra-atrial air emboli during insufflation.^[5,10,15]

Surgical management has generally been described by right thoracotomy or thoracoscopy and occasionally by median sternotomy. As the oesophagus and left atrium are most accessible from the right, posterolateral thoracotomy is the most versatile approach for both oesophageal and left atrial repair and extensive debridement. Cardiopulmonary bypass is necessary in procedures requiring atriotomy for left atrial repair. Surgical oesophagus repair is necessary to limit postoperative morbidity. Ongoing oesophageal leaking will cause and maintain mediastinitis, impairing recovery of the left atrial repair. Maintaining physical separation between the left atrium and oesophagus by means of an intercostal muscle flap or omental wrapping, which should be used as a living pedicle to promote blood flow to the area, appears to be beneficial for wound healing and outcome.^[15]

Discussion

This case report illustrates a rare, but severe complication of surgical AF ablation treatment. The complication can manifest

weeks after the procedure and rarely presents with primarily thoracic or cardiac symptoms. Widespread awareness among physicians has not yet been achieved, possibly causing delay in establishing the diagnosis. Due to the severity of this complication, there must be a clear indication for performing ablative therapy, and patients and physicians should not think too lightly of AF ablation. Given the rarity of LAOF after RFA, no uniform management strategy has been agreed upon to date. Mortality and morbidity rates remain high. Without immediate surgical management of LAOF, mortality rates have been reported to be as high as 83%, whereas adequate and immediate surgical management can reduce mortality to 34%. There is no difference in morbidity or mortality rate of LAOF following catheter-guided RFA compared with surgical RFA.^[2]

It is possible that the LAOF in our patient could have been prevented; it may have manifested due to a combination of factors, including a relatively low body mass at the time of the procedure, usage of too much power for a relatively long period during RFA in combination with leaving the oesophageal ultrasound probe in situ during the procedure displacing the ventral oesophageal wall towards the left atrium, drawing energy to the oesophageal wall and inducing pressure related oesophageal injuries.^[7] Unfortunately, due to limited clinical documentation on power and TEE usage it is not possible to confirm this hypothesis.

In retrospect, bacterial cultures of the pericardial effusion in our patient should have been acquired, because non-sterile bacterial effusion could have suggested LAOF at an earlier stage and have warranted further diagnostic procedures before discharge, although pericardial effusion is not a frequently reported finding in LAOF.^[2]

Although most contemporary literature suggests primary surgical management of oesophageal lesions is necessary to achieve the best outcome and least risk of iatrogenic air emboli,^[2,15] minimally invasive repair of the oesophagus through clipping and stenting during oesophagoscopy was successful in our patient.

No cases have been described in which non-surgical management of LAOF led to survival of the patient,^[2] although cases where there was only an oesophageal perforation or oesophago-pericardial fistula have been successfully managed by non-surgical management alone.^[15,16]

Despite the long and complicated hospital admission, neurological and functional recovery was significant in our patient and further recovery is to be expected in time with adequate rehabilitation. The rapid diagnosis and immediate non-surgical (antibiotics, nil by mouth and endoscopic oesophagus repair) and surgical atrial repair may have contributed to this outcome.

Conclusion

LAOF is a very rare, but lethal, complication of RFA if unrecognised. Since the incidence of RFA is increasing, LAOF incidence will increase accordingly. In patients presenting with fever, neurological deficits and with a recent history of RFA for AF, this diagnosis should be considered.

Early recognition and immediate surgical management of the atrium and oesophagus in combination with adequate antibiotic treatment and necessary supportive care improve the chances of survival compared with a non-surgical strategy.^[2] Although in this case surgical management of the atrial lesion combined with endoscopic closure of the oesophageal lesion led to a favourable outcome for our patient.

Disclosures

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