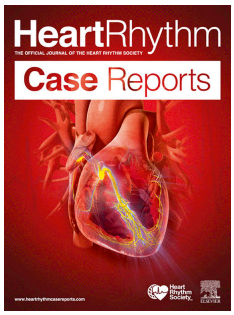
Accepted Manuscript



Massive air embolism caused by an atrio-esophageal fistula following isolation of the pulmonary veins for atrial fibrillation

Vassil Traykov, MD, Galina Kirova, MD, PhD, Petko Karagyozov, MD, Daniel Marchov, MD, Genadi Kaninski, MD, Valeri Gelev, MD

PII: S2214-0271(18)30277-X

DOI: https://doi.org/10.1016/j.hrcr.2018.11.003

Reference: HRCR 624

To appear in: *HeartRhythm Case Reports*

Received Date: 19 September 2018

Revised Date: 20 October 2018

Accepted Date: 7 November 2018

Please cite this article as: Traykov V, Kirova G, Karagyozov P, Marchov D, Kaninski G, Gelev V, Massive air embolism caused by an atrio-esophageal fistula following isolation of the pulmonary veins for atrial fibrillation, *HeartRhythm Case Reports* (2018), doi: https://doi.org/10.1016/j.hrcr.2018.11.003.

Title: Massive air embolism caused by an atrio-esophageal fistula following isolation of the pulmonary veins for atrial fibrillation.

Short title: Atrioesophageal fistula with air embolism after PVI with RF energy

Vassil Traykov, MD[a]

Galina Kirova, MD, PhD[b]

Petko Karagyozov, MD[c]

Daniel Marchov, MD[a]

Genadi Kaninski, MD[a]

Valeri Gelev, MD[a]

[a] Clinic of Cardiology, Acibadem City Clinic Tokuda Hospital, Sofia, Bulgaria

[b] Imaging department, Acibadem City Clinic Tokuda Hospital, Sofia, Bulgaria

[c] Department of Interventional Gastroenterology, Acibadem City Clinic Tokuda Hospital, Sofia, Bulgaria

Conflict of interests: none

Address for correspondence:

1407 Sofia, N. Vaptzarov blvd. 51B, Acibadem City Clinic Tokuda Hospital, Clinic of Cardiology

Dr. Vassil Traykov, Tel: +35924034606, Mobile: +359879430365, Email: vtraykov@yahoo.com

Word count: 1672

Keywords:

Atrial fibrillation, catheter ablation, cerebral air embolism, atrio-esophageal fistula, upper endoscopy.

Introduction

Catheter ablation with radiofrequency or cryoenergy is a well-established therapeutic method for symptomatic atrial fibrillation (AF) especially in its paroxysmal form (1, 2). The number of ablations of AF worldwide is growing rapidly with more than 50000 ablations performed in the US on an yearly basis (3). The procedure is associated with a number of potential complications occurring in 4.5 % of cases (4). Atrio-esophageal fistula (AEF) is a very rare but devastating complication of catheter ablation of atrial fibrillation (AF). The reported incidence of this complication reaches up to 0.11% among patients ablated for AF (5). Due to the very low total number of cases exact preventive measures have not been systematically studied. Among the most important preventive measures applied in routine practice are esophageal temperature monitoring, limiting the power of delivered energy at the posterior left atrium (LA) and treatment with proton pump inhibitors following catheter ablation (2).

We describe a case of a patient developing an AEF resulting in massive multifocal air embolism following a routine pulmonary vein isolation (PVI) for paroxysmal AF.

Case report

A 62 year-old male was referred to our center for catheter ablation because of very frequent, symptomatic episodes of atrial fibrillation. He had previously undergone cavotricuspid isthmus ablation for typical atrial flutter. Following chest computed tomography for evaluation of LA anatomy the patient underwent radiofrequency (RF) pulmonary vein isolation. The procedure was carried out under general anaesthesia. Following double transseptal puncture a circular mapping catheter (Reflexion Spiral, Abbott, Minneapolis, MN, USA) and a four-millimeter, irrigated tip catheter (Flexability, Abbot, Minneapolis, MN, USA) were introduced into the LA. Three-dimensional

anatomical map of the LA was created with the EnSite Velocity system (Abbott, Minneapolis, MN, USA). Power delivered to the left atrium was 20 Watts on the posterior wall and 30 Watts at all other locations. Flow rate was set to 17 ml/min. Lesion duration was not limited and catheter dragging was applied constantly during RF power delivery not allowing it to remain at one spot for more than 20 seconds. Esophageal temperature monitoring was not used. Wide area circumferential ablation of left and right pulmonary venous antra was carried out resulting in entrance and exit block to all four pulmonary veins. The procedure was carried out uneventfully. Total RF ablation time was 1962 sec. Procedure time was 162 minutes. The patient was discharged on the following day and was prescribed propafenone, proton-pump inhibitor and NOAC as per protocol at our center.

In the evening hours of day 21 following the procedure the patient complained of sudden onset of malaise, dyspnea and fever up to 38.9°C (102.02 °F). He also reported vomiting, severe chest pain of sudden onset and left arm numbness. Thirty minutes following initial symptom onset the patient lost consciousness. An emergency unit was called in and the patient was taken to the hospital. At the emergency room he presented in deep coma (Glasgow coma scale 3 points). The patient was intubated and laryngoscopy during intubation revealed severe edema of oropharynx. Electrocardiogram at presentation demonstrated ST segment elevation in leads I and aVL. Troponin was found to be elevated and STEMI was diagnosed. No primary PCI strategy was undertaken due to the very poor condition of the patient. Due to fever and neurological signs a concomitant neuroinfection was suspected but was excluded following the results of lumbar puncture. As a part of the regular work-up of comatose conditions a noncontrast head computed tomography (CT) scan was performed shortly after presentation to the emergency room. It showed massive air embolism to the brain with clearly visible gas collections in the cerebral arteries (Figure1). The patient was later admitted to the intensive care unit with an unclear diagnosis because the team on call was not aware of the potential complications of AF ablation. The following morning the patient had complete resolution of the ECG findings and deep coma still persisted. Because of cerebral air embolism and STEMI most likely due to coronary air embolism, AEF following catheter ablation of AF was strongly

suspected and a cardiac CT scan was performed. It revealed an air collection in the left ventricular apex (Figure 2A). A leak of contrast to the periesophageal tissues was also clearly visible (Figure 2B). There was also a massive thrombosis of the LA attached to the posterior wall close to the antrum of the left inferior pulmonary vein antrum (Figure 2C). The patient was referred to a gastroenterologist who performed esophagoscopy using carbon dioxide for insufflation. It revealed a deep lesion at the anterior wall of the esophagus forming a fistula to the adjacent LA with active bleeding into the esophagus (Figure 3, Supplementary Video 1). Following all these findings AEF resulting from previous catheter ablation of AF was diagnosed. The patient was also referred to a thoracic surgeon for further management, but an operation was deemed high risk because of the critical condition of the patient. Following diagnosis, the patient was treated conservatively. During the following 24 hours deep coma persisted, the patient developed hemodynamic instability and succumbed to severe mediastinitis.

Discussion

Currently there is no data to support benefit of catheter ablation of AF in terms of hard cardiovascular endpoints. Therefore, the procedure is directed mainly towards improvement of quality of life. Any complication in this setting has a major negative impact. AEF is associated with a high mortality, even after early surgical treatment making it the most feared complication of catheter ablation of AF (6).

Clinical manifestation of AEF includes variety of symptoms most common of which are fever and neurological deficit (7). All symptoms develop some time after the procedure in the range 2-60 days according to the results of one published series of patients with AEF. Late occurrence of symptoms might lead to late diagnosis and delayed treatment. In our case symptoms suggestive of cerebral and possible coronary air embolism developed on day 21 following the ablation which is in the typical time frame for AEF occurrence following catheter ablation of AF. Patient was diagnosed 10-12 hours after initial presentation which probably reflects low awareness of this condition by the

emergency and intensive care unit teams. Neurological impairment upon presentation is a very strong predictor of mortality according to recently published series (7). In our case the patient had severe neurological symptoms upon presentation that determined poor outcome from the very beginning and the 10-12 hours delay in the diagnosis would have been unlikely to change the therapeutic strategy. The treatment associated with the highest (although moderate in absolute numbers) survival rate in these patients is in the group of surgically treated patients (7, 8). In our case surgery was deemed very high risk and was not carried out because of the very poor condition of the patient.

Due to its low incidence management strategies and preventive measures to avoid AEF occurrence have not been extensively studied. Prevention of AEF during the procedure includes mainly limiting power to the posterior wall of the left atrium, implementing esophageal temperature monitoring, esophageal cooling or mechanical displacement of the oesophagus (9-11). The value of all these preventive measures has not been studied in large trials. Current practice guidelines recommend limiting the power on the posterior wall of the LA while applying RF energy. This was also done in our case and power was limited to 20W on the LA posterior wall. Duration of RF energy delivery was not limited but left to operator discretion. However, with constant catheter dragging during RF delivery the catheter was not allowed to remain at one spot for more than 20 seconds. It is logical to assume that shorter lesions allowing more time between them for tissue cooling in the LA region overlying the esophagus would potentially diminish the risk for esophageal injury. However, this would come in return of decreased lesion efficacy and durability. Given the lack of firm evidence the benefits of such an approach in terms of safety remain unproven. Several studies have shown that esophageal temperature monitoring might potentially reduce the incidence of esophageal injury following ablation (9) and it has therefore been implemented as a routine practice in many centres worldwide (2). However, taking all these measures is not able to completely eliminate the risk occurrence of AEF and there are reports of high incidence of esophageal injury despite the use of esophageal temperature monitoring (5).

Another widely used preventive measure to reduce the incidence of esophageal injury following catheter ablation is postprocedural administration of proton pump inhibitors. Despite its wide adoption its value has never been shown in trials but is currently recommended and used by many (2). Our postablation treatment protocol also includes routine use of proton-pump inhibitors.

Performing the ablation under general anaesthesia is reported to be among the factors associated with esophageal injury. A small randomized study demonstrated a higher rate of oesophageal tissue damage assessed by capsule endoscopy following the ablation (12). Whether this is translated in an increased rate of AEF formation in the patients ablated under general anaesthesia remains unclear because no cases of AEF were reported in the studied population. Possible mechanisms associated with increased incidence of esophageal injury in anesthetised patients are absence of pain perception as a potential sign to limit or stop energy delivery, reduced esophageal motility and the lack of swallowing. While there are no firm data showing higher incidence of AEF in these conditions general anaesthesia continues to be widely used in routine practice (2).

In our case we observed AEF formation resulting in multifocal air embolism (cerebral and coronary) despite the preventive measures undertaken in accordance with the recommendations. The procedure was short lasting and RF time was also relatively short but sufficient to achieve pulmonary veins isolation. General anaesthesia is a potential, although unproven risk factor that could have aided the process. Diagnosis was made late after initial presentation and surgery was not undertaken because of the poor condition of the patient. However, earlier recognition would have been unlikely to result in a surgical intervention because the patient was already in deep coma upon presentation.

References:

1. Kirchhof P, Benussi S, Kotecha D, et al. 2016 ESC Guidelines for the management of atrial fibrillation developed in collaboration with EACTS. Europace. 2016;18:1609-78.

2. Calkins H, Hindricks G, Cappato R, et al. 2017 HRS/EHRA/ECAS/APHRS/SOLAECE expert consensus statement on catheter and surgical ablation of atrial fibrillation. Heart Rhythm 2017; 14:e257-e444.

3. Freeman JV, Wang Y, Akar J, Desai N, Krumholz H. National Trends in Atrial Fibrillation Hospitalization, Readmission, and Mortality for Medicare Beneficiaries, 1999-2013. Circulation. 2017;135:1227-39.

4. Cappato R, Calkins H, Chen SA, et al. Updated worldwide survey on the methods, efficacy, and safety of catheter ablation for human atrial fibrillation. Circ Arrhythm Electrophysiol. 2010;3:32-8.

5. Medeiros De Vasconcelos JT, Filho S, Atie J, Maciel W, De Souza OF, Saad EB, Kalil CA, De Castro Mendonca R, Araujo N, Pisani CF, Scanavacca MI. Atrial-oesophageal fistula following percutaneous radiofrequency catheter ablation of atrial fibrillation: the risk still persists. Europace. 2017;19:250-8.

6. Singh SM, d'Avila A, Singh SK, Stelzer P, Saad EB, Skanes A, Aryana A, Chinitz JS, Kulina R, Miller MA, Reddy VY. Clinical outcomes after repair of left atrial esophageal fistulas occurring after atrial fibrillation ablation procedures. Heart rhythm : the official journal of the Heart Rhythm Society. 2013;10:1591-7.

7. Han HC, Ha FJ, Sanders P, Spencer R, Teh AW, O'Donnell D, Farouque O, Lim HS. Atrioesophageal Fistula: Clinical Presentation, Procedural Characteristics, Diagnostic Investigations, and Treatment Outcomes. Circ Arrhythm Electrophysiol. 2017;10:e005579.

8. Chavez P, Messerli FH, Casso Dominguez A, Aziz EF, Sichrovsky T, Garcia D, Barrett CD, Danik S. Atrioesophageal fistula following ablation procedures for atrial fibrillation: systematic review of case reports. Open Heart. 2015;2:e000257.

9. Leite LR, Santos SN, Maia H, et al. Luminal esophageal temperature monitoring with a deflectable esophageal temperature probe and intracardiac echocardiography may reduce esophageal injury during atrial fibrillation ablation procedures: results of a pilot study. Circ Arrhythm Electrophysiol. 2011;4:149-56.

10. Chugh A, Rubenstein J, Good E, Ebinger M, Jongnarangsin K, Fortino J, Bogun F, Pelosi F, Oral H, Nostrant T, Morady F. Mechanical displacement of the esophagus in patients undergoing left atrial ablation of atrial fibrillation. Heart Rhythm. 2009;6:319-22.

11. Mateos JC, Mateos EI, Pena TG, Lobo TJ, Vargas RN, Pachon CT, Acosta JC. Simplified method for esophagus protection during radiofrequency catheter ablation of atrial fibrillation--prospective study of 704 cases. Rev Bras Cir Cardiovasc. 2015;30:139-47.

12. Di Biase L, Saenz LC, Burkhardt DJ, et al. Esophageal capsule endoscopy after radiofrequency catheter ablation for atrial fibrillation: documented higher risk of luminal esophageal damage with general anesthesia as compared with conscious sedation. Circ Arrhythm Electrophysiol. 2009;2:108-12.

Figure legends

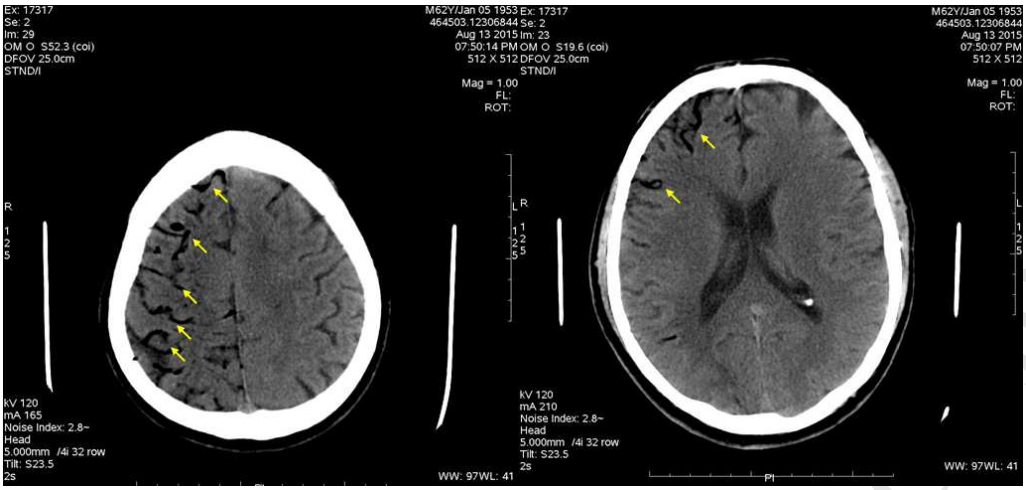


Figure 1. Native head CT scan demonstrating massive air collections in the cerebral arteries (arrows).

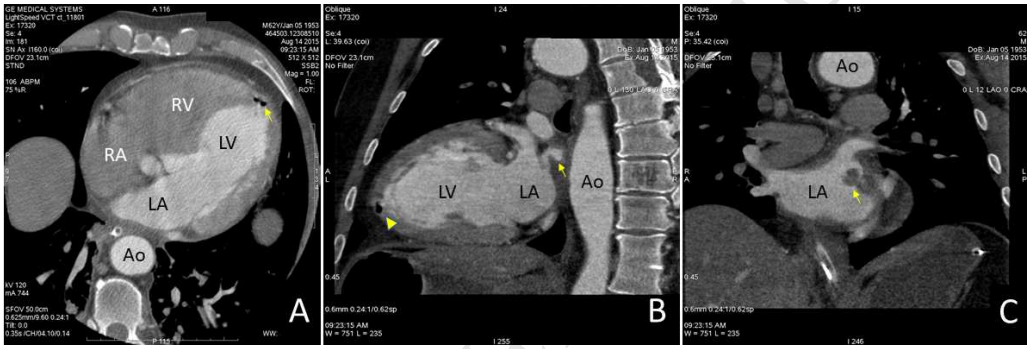


Figure 2. Cardiac CT scan. Panel A: Horizontal plane section demonstrating air collection at the left ventricular apex (arrow). Panel B: Sagittal plane section demonstrating contrast leakage from the left atrium to the periesophageal tissues (arrow). The air collection at the left ventricular apex is visible in this section as well (arrowhead). Panel C: Frontal plane section at the level of left atrium demonstrating intracavitary thrombosis attached to the posterior wall close to the antrum of the left inferior pulmonary vein. Left atrium (LA), left ventricle (LV), right atrium (RA) and right ventricle (RV).

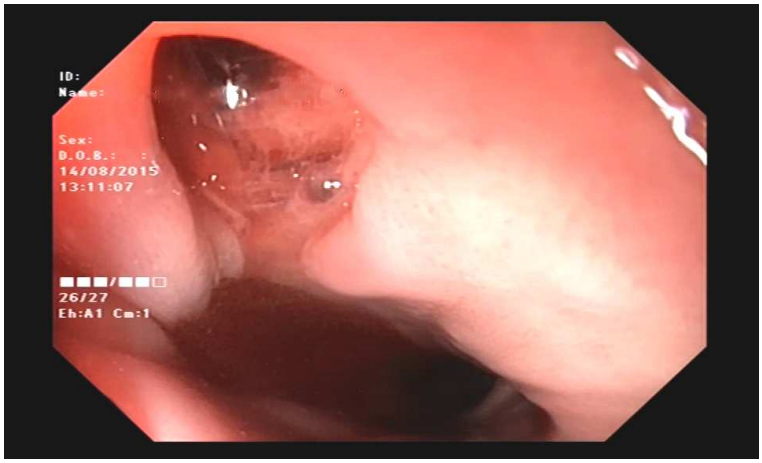


Figure 3. Upper endoscopy demonstrating a deep ulcerative lesion at the anterior esophageal wall consistent with atrioesophageal fistula.

Key teaching points

- Massive air embolism especially when multifocal should raise suspicion for AEF at the first medical contact. History of recent ablation for atrial fibrillation should be actively sought.
- Atrio-esophageal fistula might occur even after procedures in which power applied to the posterior left atrial wall is limited in accordance with current recommendations.
- Left atrial thrombosis at or close to the presumed location of the atrioesophageal fistula could also occur.