Sistemska embolija s hrano zaradi atrio-ezofagealne fistule po alkoholni ablaciji Marshalove vene: prikaz primera

Systemic food embolism in a case of atrio-esophageal fistula after alcohol ablation of the Marshall vein: A case report

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Izvleček

Namen: *Ablacijski poseg je pogosta metoda, ki se uporablja v zdravljenju simptomatskih pacientov s persistentno atrijsko fibrilacijo (AEF). Po tovrstnih posegih so opisani možni različni zapleti, najhujši med njimi je pojav atrio-ezofagealne fistule. Čeprav pride do AEF redko, je smrtnost tega zapleta visoka. Prepoznava AEF je težavna, zlasti med urgentno obravnavo zaradi nenadnega poslabšanja kliničnega stanja, saj je klinična slika AEF nespecifična in vključuje predvsem nenadno nastalo nevrološko simptomatiko zaradi zračne embolije ali embolije s hrano.*

Predstavitev primera: *Pri 46-letni ženski z anamnezo deset let trajajoče AEF je bila opravljena etanolna ablacija Marshalove vene in elektrokonverzija. Približno mesec*

Abstract

Purpose: *Ablation procedures are common and standard therapeutic interventions for symptomatic patients with persistent atrial fibrillation (AF). Various complications have been observed after the procedure, with one of the most fatal being atrio-esophageal fistula (AEF). Although rare, AEF has very high morality. Diagnosis of AEF is challenging, especially in emergency settings, because of its unspecific clinical signs, such as atypical neurological signs after food or air embolism.*

Case Presentation: *A 46-year-old woman with a 10-year history of AF underwent ethanol infusion of the Marshall vein. Almost a month after the procedure, the patient collapsed and was brought to the emergency department with signs of right*

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dni po posegu je bila pacientka najdena nezavestna, in to z znaki desnostranske hemipareze. CT angiografija je pokazala okluzijo leve srednje cerebralne arterije brez jasno demarkiranih ishemičnih sprememb možganovine. Med trombektomijo je pričela pacientka nenadno bruhati kri, po opravljeni urgentni enzofagogastroskopiji je bila takoj prepeljana v operacijsko dvorano zaradi suma na rupturo požiralnika. Umrla je na operativni mizi. Obdukcija je potrdila diagnozo AEF z obsežno gastrointestinalno krvavitvijo in posledicami embolije s hrano. Predstavitev primera smo pripravili z namenom, da izboljšamo prepoznavanje klinične slike AEF po posegu etanolne ablacije za zdravljenje AF. Svetujemo aktivno spremljanje pacientov po tovrstnih posegih in pogostejše kontrolne preglede.

side hemiparesis. Occlusion of the left middle cerebral artery was identified with no ischemic change in brain parenchyma. During thrombectomy the patient had a sudden episode of hematemesis. After esophagogastroscopy, she was immediately transferred to the operating room for emergency surgery, but died on the operating table. Autopsy confirmed a clinical suspicion of AEF with massive gastrointestinal bleeding and systemic food embolism. We present this case to improve clinical recognition of the signs of AEF after alcohol ablation for AF. We also recommend active surveillance for patients who undergo similar procedures, including regular check-ups.

INTRODUCTION

Death due to massive food embolism is extremely rare in any setting, but has been described in cases with chronic esophageal ulcer [1], perforation of a peptic ulcer in hiatal hernia [2], and after a blunt force trauma to the chest and abdomen [3]. One of the mechanisms responsible for food embolism is the presence of atrio-esophageal fistula (AEF). This is a rare complication of ablation procedures used for the treatment of atrial fibrillation, with an estimated incidence of 0.05%–0.2% [4] after percutaneous radiofrequency ablation. There are no data available on the estimated incidence of AEF in patients who undergo alternative ablation techniques, such as ablation of the Marshall vein. AEF has an estimated mortality of 40%–80%, which is usually a result of either brain ischemia or sepsis [4,5]. High mortality is also a consequence of late recognition of clinical signs and symptoms, which delays treatment [4].

We present a case of a patient with persistent atrial fibrillation (AF), who developed an AEF fistula after ethanol cardiac ablation of the Marshall vein. We present this case to remind clinicians and patients about an atypical and less known clinical presentation of this condition, which often leads to late diagnosis and poor survival. We emphasize that AEF can be a complication after the use of alternative ablation techniques, and not just radiofrequency ablation, which has been previously reported.

CASE PRESENTATION

Clinical history and presentation

A 46-year-old woman with a 10-year history of persistent AF underwent ethanol infusion of the Marshall vein and electro conversion. Because the procedure did not result in a stable sinus rhythm, she was scheduled for a repeat electro conversion. Three days before the planned repeated electro conversion, she presented with aphasia and right side hemiparesis. First, computed tomography (CT) of the head and neck, thorax, and abdomen was performed (Figure 1), which showed spleen and bilateral kidney infarctions This was followed by CT angiography of the brain, which identified an occlusion of the left middle cerebral artery with no ischemic change in brain parenchyma. The patient's personal history was also significant, involving

vomiting after meals, beginning the day after the initial procedure, and episodes of dull pain between the shoulder blades. However, she did not seek medical attention for these symptoms. Apart from her regular medical therapy, including bisoprolol, amiodarone, pantoprazole, and ibuprofen (if necessary), she was taking 20 mg of rivaroxaban daily since the ablation procedure.

Figure 1. *An abdominal computed tomography scan shows bilateral kidney infarctions (arrows).*

Therapeutic interventions

Based on CT angiography results, she was transferred to our institution for a planned thrombectomy. Thrombectomy resolved the occlusion of the left middle cerebral and right renal arteries. During the procedure, she experienced a sudden episode of abundant hematemesis, requiring sedation, intubation, and inotropic support with noradrenaline. She was transferred to the ICU for urgent endoscopy. Esophagogastroscopy showed a rupture of the esophageal wall with a formed thrombus in the middle third of the esophagus, without active bleeding, so she was immediately transferred to the operating room. Soon after the induction of anesthesia, she became hypotonic, and the ECG showed pulseless electrical activity. During resuscitation she was actively bleeding from the mouth. Sternotomy was performed in an attempt to save the patient, but she died on the operating table. All diagnostic and therapeutic interventions are summarized in Figure 2.

Autopsy findings

Postmortem examination confirmed a clinical suspicion of atrio-esophageal fistula (Figure 3) with massive gastrointestinal bleeding, which was determined to be the cause of death. The autopsy revealed hypoxic changes of the brain with no clearly demarcated infarction. Gastric content with identifiable plant cells and skeletal muscle was found in the lumen of the left ventricle of the heart. Food emboli (Figure 4) were found in the vessels of the epicardium and smaller vessels of the myocardium, lungs, and in renal arteries. There were hypoxic changes of cardiomyocytes, indicating an early stage of myocardial infarction. Splenic and kidney infarctions were also present.

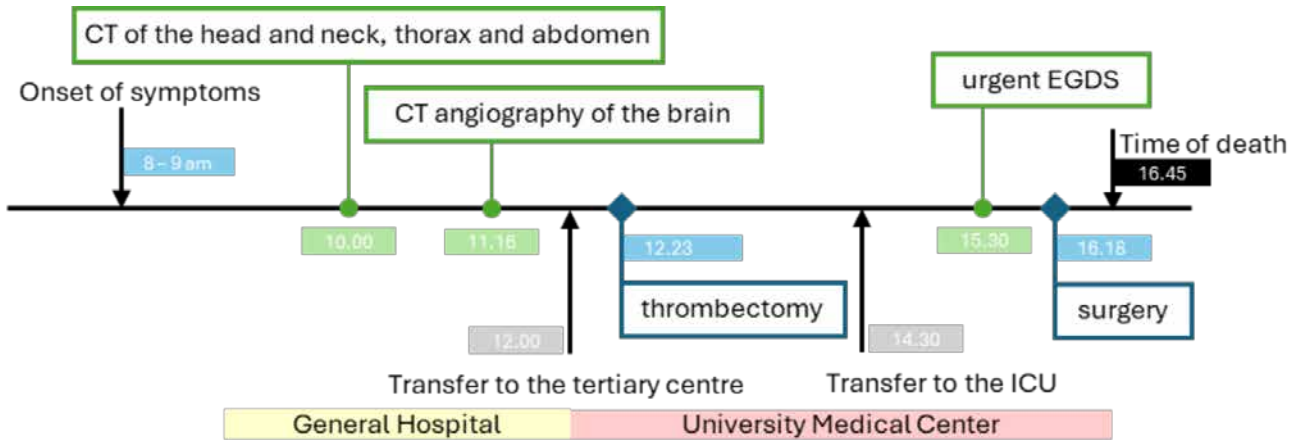

Figure 2. *Timeline of diagnostic and therapeutic interventions after the onset of neurological symptoms.*

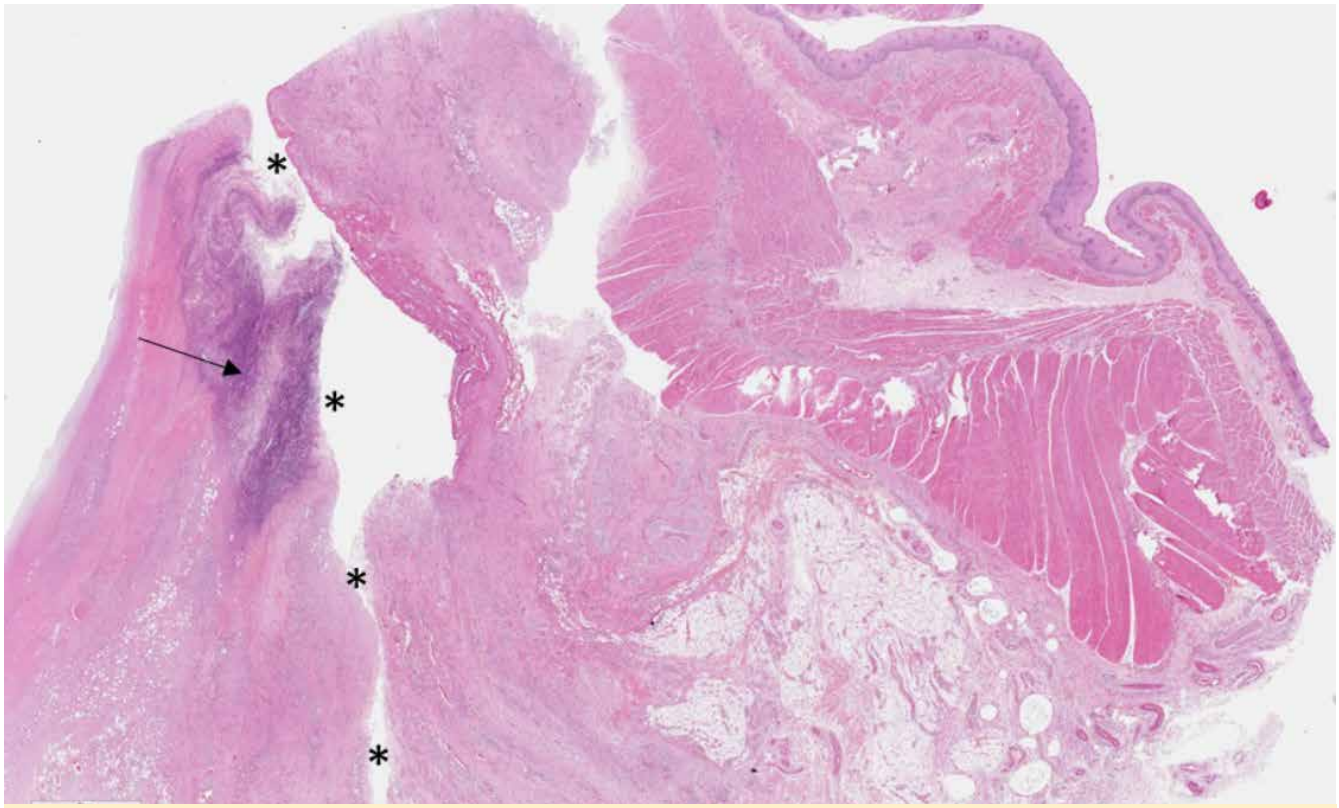

Figure 3. *Rupture in the esophageal wall (marked with ****). There were areas of necrotic tissue (arrow) surrounding the defect in the esophageal wall (hemoxylin & eosin staining). The image was captured with an Aperio ImageScope, with the scale in the lower left corner of the image.*

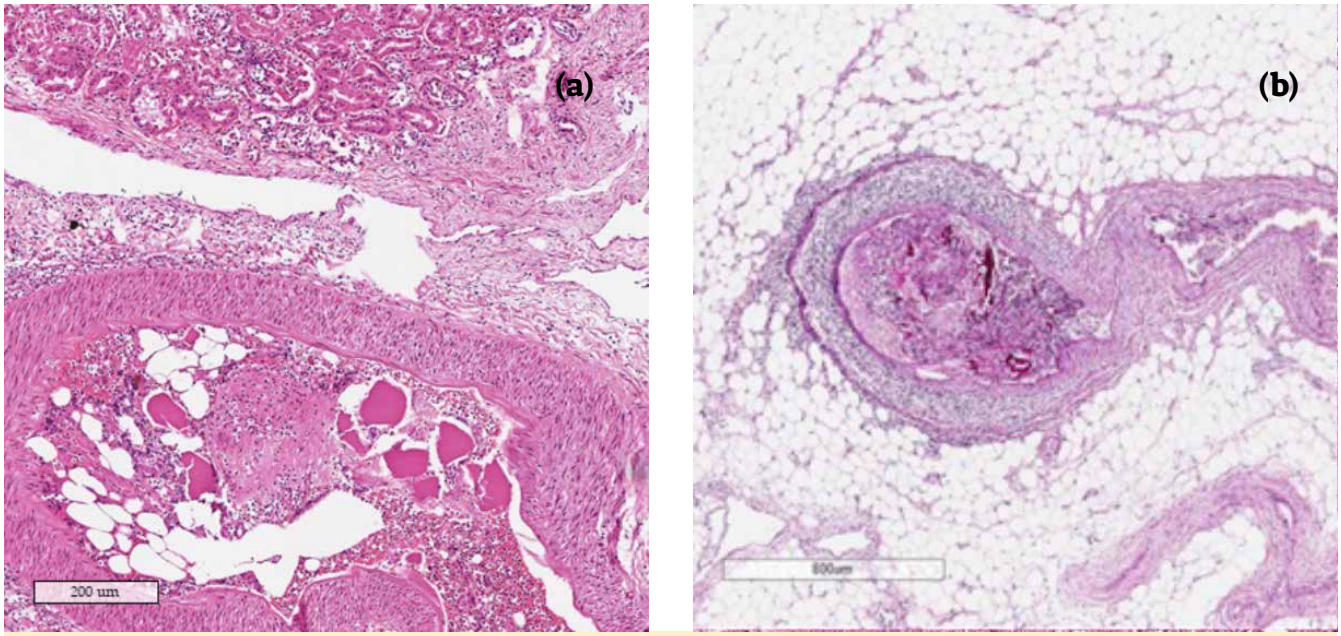

Figure 4. *Examples of food emboli in the (a) renal (hemoxylin & eosin staining) and (b) coronary (Periodic Acid-Schiff staining) artery. The image was captured with an Aperio ImageScope, with the scale in the lower left corner of the images.*

DISCUSSION AND CONCLUSIONS

Ethanol infusion ablation of the Marshall vein is an alternative ablation technique, used in the treatment of persistent cardiac arrhythmias, which do not respond to antiarrhythmic drugs. The Marshall vein is an embryological remnant in the left superior vena cava, and is a common source of AF, because of sympathetic and parasympathetic innervations that affect the electrophysiology of heart tissues [6,7]. High concentrations of ethanol are applied to cardiac tissue to solubilize cell membranes, which ultimately causes cell death. Dead cells are replaced by fibrous tissues, which isolate electrical signals from arrhythmogenic foci [8]. Various complications are associated with EIA, including pulmonary vein thrombosis, pericardial effusion, pericardial tamponade, and left atrial wall hematoma [8]. The occurrence of AEF has thus far not been described after EIA. Although rare, even after radiofrequency ablation, AEF is a serious and often under-recognized complication with a mortality of 40%–80% [9]. Delayed diagnosis of AEF can be attributed to unspecified clinical signs, which occur from a few days, up to more than a month after the procedure. Signs and symptoms include general weakness and lethargy, but also include chest pain, fever, dysphagia, melena, hematemesis, and even sepsis. When AEF results in a food or air embolism, as well as subsequent infection, neurological signs usually prevail [4,10].

Systemic food or air embolism develops due to a reverse pressure gradient, which occurs during food swallowing and emesis. When there is no active passage of food through the esophagus, pressure in the esophageal lumina is lower than pressure in the atrium, causing persistent bleeding in the esophagus. When food passes through the esophagus, either by swallowing or by vomiting, pressure during a rise in intraluminal pressure enables food particles as well as air to enter the blood stream. This causes systemic embolism, leading to infarction in target organs and septicemia. The causes of death after AEF can involve septic shock, significant brain ischemia, or as in our case, hypovolemic shock [4,10].

The diagnosis of AEF is challenging. If AEF is suspected, contrast enhanced thoracic CT scans should be performed. Endoscopy is generally contraindicated due to the risk of systemic air embolization and stroke from air insufflation during the procedure [11]. In our case, the diagnosis of AEF was not suspected, and the patient was only treated for neurological complications, because the full patient history was later obtained only during treatment. If the patient had earlier reported any of the symptoms, and if the information about the cardiac ablation procedure was known beforehand, diagnostic and treatment procedures could have been planned differently.

In conclusion, we presented this case to improve the clinical recognition of AEF symptoms after alcohol ablation for AF. We recommend active surveillance for patients who undergo similar procedures, including regular check-ups, at least using phone calls, to confirm or deny any gastrointestinal problems. We especially emphasize the relevance of educating the patient and his/her family about the importance of immediately reporting any symptoms. Finally, we want to raise awareness among general practitioners and emergency physicians, to recognize such symptoms and plan treatment interventions accordingly.

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