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Esophagopericardial Fistula Complicated by Tension Pyopericardium Managed with a Three-incision Esophagectomy

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Central Message:

We report a case of esophagopericardial fistula after dilation for the recalcitrant peptic stricture complicated by tension pyopericardium and successfully managed with urgent esophagectomy.

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Introduction

Esophagopericardial fistulization (EPF) is an extremely rare complication after esophageal stricture dilation.\textsuperscript{1,2} Much rarer is fistulization into the pericardium causing cardiac tamponade, a life-threatening event.\textsuperscript{3,4} A single case of tension pericarditis after esophageal dilation has been reported in the literature in a patient who expired, and the diagnosis was confirmed on postmortem examination.\textsuperscript{1} We report the first case of EPF after stricture dilation resulting in tension pyopericardium that was successfully managed with an urgent, three-incision esophagectomy.

Clinical Summary

Our patient is a 38-year-old female with history of peptic esophageal stricture who had undergone nine prior esophageal dilations and an intralesional injection of triamcinolone. The patient provided written consent for publication; IRB approval was not required. The patient has history of diabetes mellitus, a body mass index of 39, and cannabis use. During her last endoscopy 12 days prior to presentation, a tight stricture 33 cm from the incisors could not be traversed with a 9.2-mm endoscope and was dilated to 17 mm using a Savary bougie over a guide wire (Figure 1). One day prior to admission the patient developed vomiting and chest pain. The patient was not in acute distress, temperature 37.5 °C, heart rate 119/min, blood pressure 125/90 mm Hg, respiratory rate 20/minute, saturation 93% on room air, and had tenderness in the subxiphoid area. Chest radiogram was unrevealing. Chest computed tomography (CT) with oral contrast was consistent with EPF (Figure 2).

Given the refractory esophageal stricture, now complicated by EPF, the patient was offered an esophagectomy. A preoperative echocardiogram was not performed given normal
blood pressure and plan for urgent repair facilitated by a cardiothoracic anesthesiologist. Patient was positioned in the left lateral decubitus position with right groin prepped into the operative field should femoral cannulation be needed for initiation of cardiopulmonary bypass. During a right posterior lateral thoracotomy in the 5th intercostal space and initiation of a single lung ventilation, the patient developed significant hypotension requiring fluid resuscitation and high doses of vasopressors. The pericardium was tense and erythematous, and pericardiotomy yielded a large amount of pus under pressure (Supplemental Figure 1A, 1B). The patient’s hemodynamics promptly improved.

The esophagus was circumferentially dissected and retracted proximally and distally to the area of inflammation to expose the location of the fistula (Supplemental Figure 2A). Once the esophagus was freed from the pericardial adhesions, the EPF became apparent (Supplemental Figure 2B). Dense intrapericardial adhesions in the area of the fistula were present and were left undisturbed. Given no findings of malignancy in prior endoscopic biopsies, lymphadenectomy was not pursued.

The patient was repositioned supine, and the rest of the operation was performed according to the standard technique of a three-incision esophagectomy. Through a midline laparotomy the distal esophagus was divided, 5-cm-wide gastric conduit was prepared and advanced through the posterior mediastinum into the left neck incision for the cervical gastroesophageal anastomosis. Given normal hemodynamics, euhemeria, lack of significant acidosis, non-inflamed tissues of the stomach and neck, primary anastomosis rather than temporary esophagostomy was completed and a feeding tube jejunostomy was placed. Pathology of the esophagus confirmed perforation in the area of the esophageal ulcer with no evidence of malignancy.
The patient was extubated on postoperative day (POD) 1. Her hospital course was uncomplicated although discharge from the hospital was delayed due to social issues. Per our protocol we delay oral intake until POD 21 after esophagectomy. The patient was not compliant with nil per os status, and on POD 17 an esophagram demonstrated no anastomotic leak or aspiration. The patient was discharged on POD 22 and was doing well at her follow-up clinic visit 3 months later. The patient continued to report no dysphagia 19 months later at the time of admission for the management of diabetic ketoacidosis and pneumonia, complicated by embolic cerebrovascular accident. Chest CT demonstrated normal size of her gastric conduit and echocardiogram demonstrated normal cardiac function without pericardial effusion. Patient died at home 2 months after admission for pneumonia.

**Discussion**

Most reports in the literature regarding esophagopericardial fistulization are within the setting of cardiac ablation procedures or esophageal carcinomas, with management including primary repair or, in severe cases, esophagectomy. Esophagopericardial fistula secondary to endoscopic interventions are extremely rare.

Obtaining a personal history of prior foregut symptoms and conditions, past surgeries, and esophageal endoscopic procedures are essential for timely diagnosis and optimal treatment of EPF. Depending on the time of presentation, symptoms and physical signs may vary in severity and include chest pain, dysphagia, shortness of breath, fever, and tachycardia. Tachycardia and hypotension may be signs of sepsis, hypovolemia, or pericardial tamponade. Physical examination is generally nonspecific and in an advanced stage may include classic signs of pericardial effusion and tamponade.
Chest X-ray findings are nonspecific but may reveal presence of pneumomediastinum or pleural effusions. Optimal imagining modalities require oral, water-soluble contrast to confirm the presence of a fistula and its location; the main modalities used are fluoroscopic esophagogram and/or CT esophagram. The benefits of the CT are its ability to establish the diagnosis of EPF, demonstrate the level of the fistula, indicate the presence of esophageal dilation and inflammation, and confirm the existence of a mediastinal abscess and intrapericardial air or effusion. Pericardial air and oral contrast efflux from the esophagus into the pericardium are diagnostic. An echocardiogram can demonstrate pericardial effusion and reveal evidence of tamponade effect. Endoscopy should be avoided unless performed in the operating room, as insufflation of air may precipitate pericardial tamponade. In cases of atrioesophageal fistula (AEF), air insufflation may result in air embolism if the patient has a patent foramen ovale.

Esophageal perforation with pericardial fistulization requires urgent intervention. Typical management strategies of esophageal perforation and fistula include primary repair, esophagectomy, and esophageal diversion or esophageal stenting in high-risk patients.\textsuperscript{7-10}

Surgical management of necessitates evacuation of purulent debris with drainage of the pericardium and pleural space, repair of perforation, or esophagectomy. In most cases this can be achieved via right thoracotomy, although a median sternotomy approach has been described.\textsuperscript{6} In cases of primary repair, the best practice is to interpose vascularized tissue between the esophagus and the pericardium. Flap options include an intercostal muscle flap, pericardial fat pad, pleural flap, and less commonly diaphragm or omental flaps.

Esophageal stenting, especially in a contaminated field, has a high failure rate and rather is used as a bridge for operative intervention.\textsuperscript{6,10} In our opinion stenting should not be used as
definitive management for cases involving esophageal fistulae and should only be reserved for patients unfit for immediate surgery or for palliative purposes.

EPF should be differentiated preoperatively from even more lethal AEF, which is caused by cardiac ablations. In the latter cases, patients typically present with fever, neurologic changes, and chest pain, among other symptoms approximately 3 weeks after ablation. Coagulation necrosis fuses all layers of the cardiac wall to the pericardium and esophagus with possible subsequent fistulization. For best outcomes AEF repair requires cardiopulmonary bypass.\textsuperscript{11} Surgical approaches utilized included median sternotomy, right thoracotomy, or a combination of both.

Complications of radiofrequency ablations resulting in pericardioesophageal fistula without communication to the atrium have been reported.\textsuperscript{6} Endoesophageal procedures, like dilations, on the other hand, typically result only in EPF and do not affect the heart itself given different mechanisms and direction of traumatic force. In cases where EPF and AEF cannot be differentiated preoperatively, surgical planning should include possibility of a cardiopulmonary bypass.

Pericardial tamponade that is clinically evident on presentation should be managed emergently with a percutaneous drain to stabilize the patient, akin to tension pyopericardium from other etiologies.\textsuperscript{12}

Our case highlights the importance of early recognition and prompt management of EPF, as the tamponade effect of pyopericardium may remain compensated for some time but promptly progress and lead to shock. Our patient, who had normal blood pressure preoperatively and sustained hemodynamic decompensation at induction of general anesthesia that resolved with pericardiotomy, is an attestation of a tamponade effect of tension pyopericardium.
Esophagectomy is an accepted management option for cases of an end-stage achalasia and refractory benign strictures that failed non-operative management, as is the case for this patient. In young, select patients with peptic esophageal strictures refractory to numerous dilations, earlier elective esophagectomy should be considered to avoid complications.

There are very few reported cases of esophageal fistulization into pericardium from various etiologies. Based on a thorough literature search of the English language, this is the first report of a patient who developed tension pyopericardium after dilation of a benign esophageal stricture and was managed with urgent esophagectomy. In patients with refractory benign esophageal strictures presenting with esophagopericardial fistula, we recommend prompt surgical management with esophagectomy.
References


Supplemental References

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Figure Legends

Figure 1
A—Endoscopic view of a tight fibrotic esophageal stricture at 33 cm from incisors.
B—Stricture after dilation to 17 mm.

Figure 2
CT esophagram demonstrates contrast-enhanced small pericardial effusion with intrapericardial air and esophageal fistula leading into the pericardium. Note the absence of mediastinal or pleural fluid collections.

Supplemental Figure 1
A—Incision of tense, inflamed pericardium anterior to the phrenic nerve with immediate drainage of pus under pressure.
B—Wide pericardiotomy was created with an energy device and the fibrinopurulent debris surrounding the heart was evacuated.

Supplemental Figure 2
A—Esophagus dissected circumferentially and encircled distally and proximally to the area of dense esophagopericardial adhesions.
B—Esophagopericardial fistula divided, exposing a small pericardial opening.