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Case Report

Esophagopulmonary fistula causing pulmonary arterial pseudoaneurysms and massive hemoptysis ☆,☆☆

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ABSTRACT

An esophagopulmonary fistula (EPF) may occur in patients with esophageal carcinoma and result in pulmonary abscess formation. Lung abscesses may in turn cause pulmonary artery (PA) pseudoaneurysms and life-threatening hemoptysis. We report a 59-year-old man with past medical history of metastatic distal esophageal adenocarcinoma who presented with fever, cough, and massive hemoptysis. Imaging evaluation demonstrated an EPF, associated lung abscess, and PA pseudoaneurysms. The presented case illustrates that embolization of PA pseudoaneurysms to prevent bleeding, and endoscopic esophageal covered stent graft placement to divert esophageal contents from the abscess, may facilitate a favorable outcome.

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Introduction

Pulmonary artery (PA) pseudoaneurysms are at high risk of rupture and may cause massive hemoptysis [1]. Peripheral PA pseudoaneurysms occur most commonly after catheter-induced trauma, especially with Swan-Ganz catheters [2], or in patients with tuberculosis (Rasmussen's aneurysms) [3]. Lung abscesses, septic emboli, bronchiectasis, and lung neoplasms may also lead to rupture of PA branches [4]. An esophagopulmonary fistula (EPF) may complicate esophageal carcinoma and result in pulmonary abscess formation [5]. In this report, we describe PA pseudoaneurysms caused by a malignant EPF, leading to life-threatening hemoptysis. Bleeding resolved after embolization of PA pseudoaneurysms. In addition, placement of an esophageal stent graft to divert esophageal contents from the fistula, and percutaneous drainage of the lung abscess, contributed to the patient's recovery.

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Fig. 1 – A CT angiogram of the chest demonstrates a 12 x 11 x 9 cm air-fluid cavity within the right lower lobe with mild peripheral enhancement. Two pseudoaneurysms were visualized within the cavity (long arrows A, B) measuring 0.8 x 1.0 x 1.0 cm and 1.5 x 1.8 x 2.4 cm. Layering dependent contrast is visualized within the cavity (short arrow, A) from a prior esophagram, suggesting communication between the abscess and the gastrointestinal tract.

Fig. 2 – (A) A digital subtraction angiogram of the right pulmonary artery demonstrates a large bilobed pseudoaneurysm arising from the posterior segmental branch of the right pulmonary artery (long arrow) and a smaller pseudoaneurysm arising from the lateral segmental branch of the right pulmonary artery (short arrow). (B) The posterior and lateral segmental pulmonary arteries were then coiled to angiographic stasis.

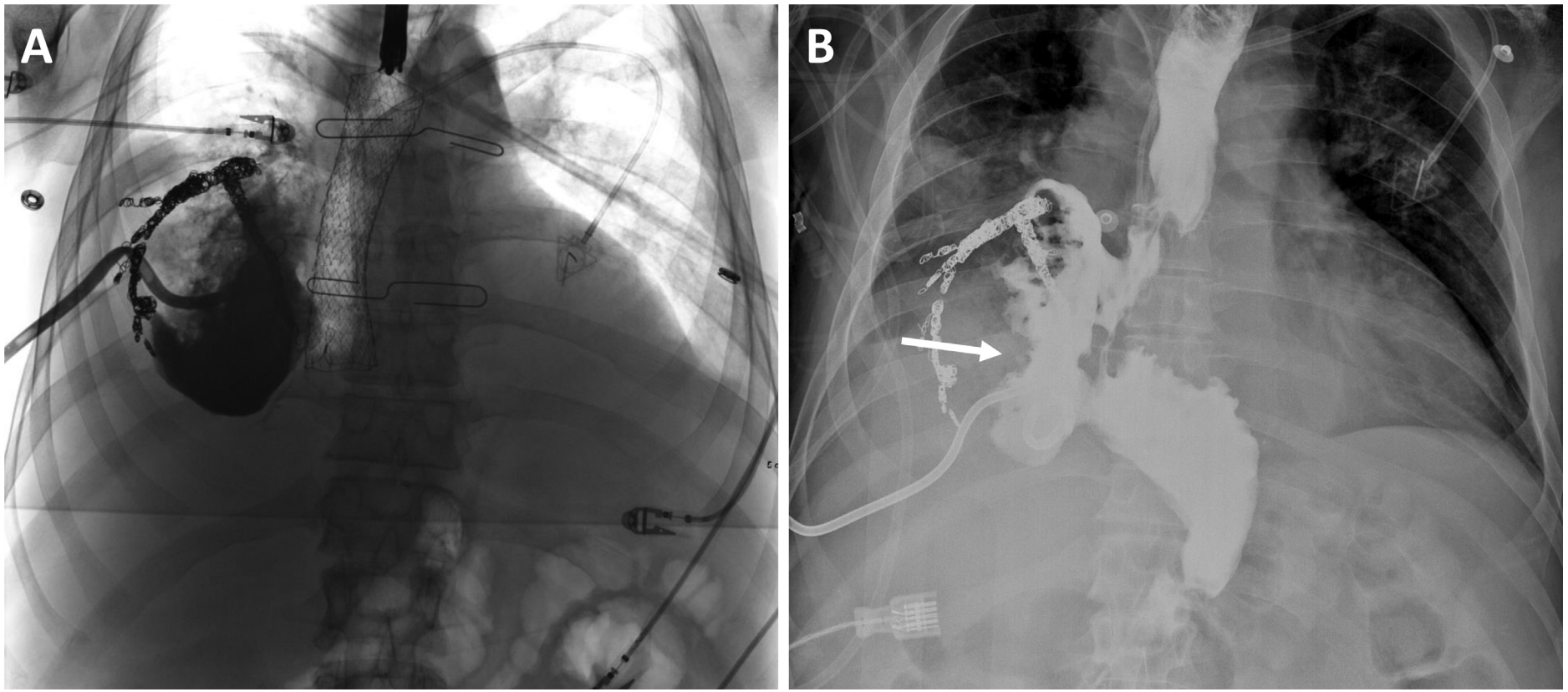

Fig. 3 – (A) A single 23 mm x 120 mm stent graft was deployed across the fistula tract under fluoroscopic and endoscopic guidance. (B) The esophagram confirmed a fistula between the distal esophagus and the right lower lobe abscess cavity (arrow).

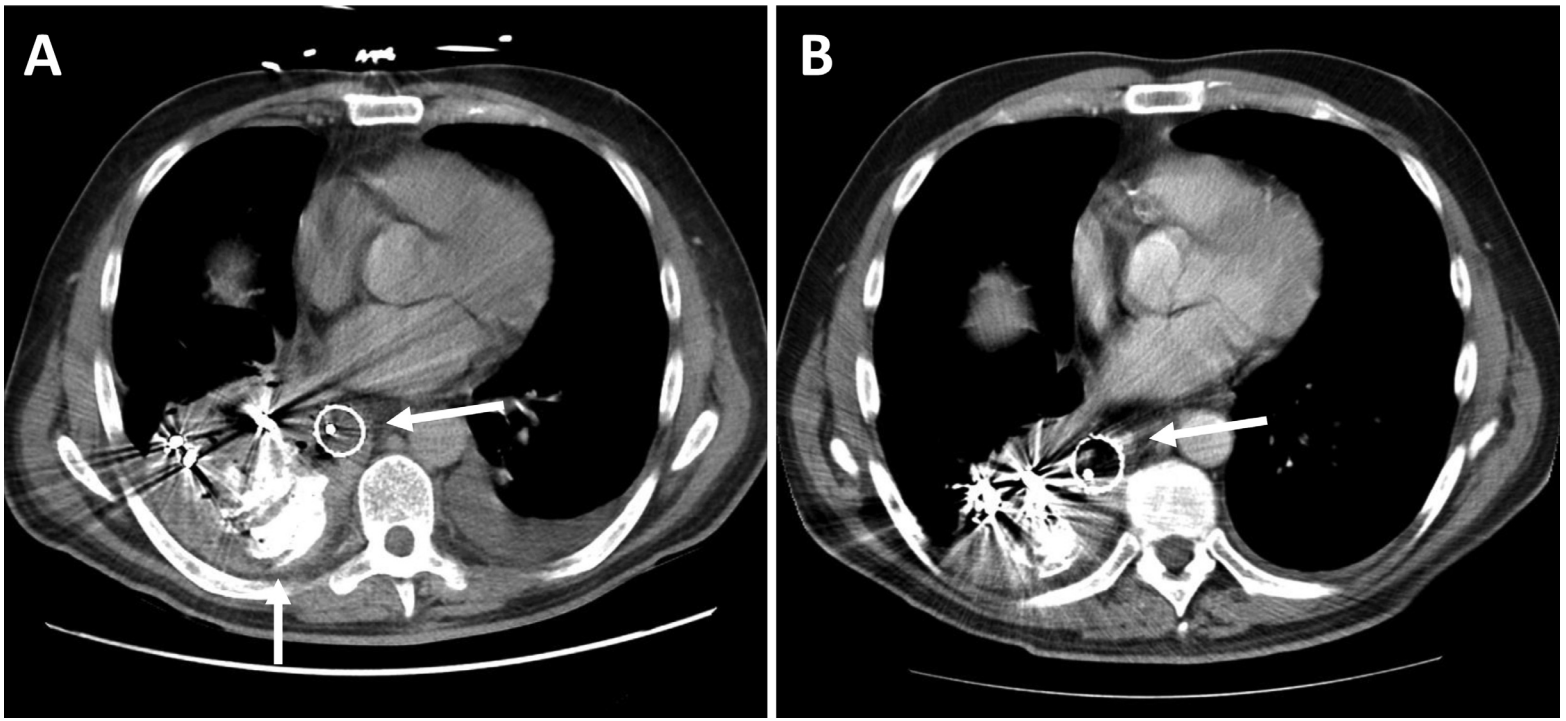

Fig. 4 – Follow up contrast-enhanced chest CT at 2 (A) and 4 (B) weeks after embolization showed a progressive decrease in the size of right lower lobe abscess cavity. The esophageal stent (long arrows) is partially imaged.

Case Report

A 59-year-old man with past medical history of metastatic distal esophageal adenocarcinoma (T3N0M1) presented with fever, cough, and massive hemoptysis for 2 days (approximately 250 mL over 24 hours). The patient was status post neoadjuvant chemoradiation, distal esophagectomy and partial gastrectomy with esophagogastric anastomosis 2 years before presentation. He had undergone multiple endoscopic dilatations of an anastomotic stricture.

An esophagram at an outside hospital revealed a fistula from the esophagus to the right lower lung zone. A chest CT demonstrated a right lower lobe pulmonary abscess. Upon administration of intravenous iodinated contrast, PA pseudoaneurysms were noted to arise from right lower lobe PA branches encased in the abscess (Fig. 1). He was then transferred to our hospital for further management.

Interventional radiology was consulted, and PA angiography was performed, confirming hypoperfusion of the right lower lung zone with 2 pseudoaneurysms arising from the posterior and lateral segmental pulmonary arteries (Fig. 2A). The feeding arteries were identified, selectively catheterized and embolized with Nester microcoils (Cook Medical Inc., Bloomington, IN, USA) (Fig. 2B). Subsequently, a 14 Fr chest tube (Flexima, Boston Scientific, Natick, MA, USA) was placed under ultrasound and fluoroscopic guidance within the lower lobe abscess cavity, yielding scant, thick purulent fluid. The day after embolization, a follow-up esophagram confirmed a fistula between the distal esophagus and the abscess cavity within the right lower lobe. The patient then underwent upper gastrointestinal endoscopy which demonstrated an erythematous esophagogastric anastomosis with an esophageal defect approximately 2 cm from the anastomosis. A single 23 mm x 120 mm stent graft (EndoMaxx; Merit Medical Endotek, South Jordan, Utah, USA) was placed over the defect under fluoroscopic and endoscopic guidance to divert flow from the fistula (Fig. 3).

A fluid sample from the chest tube grew *Candida albicans*. The patient was started on fluconazole flushes through the chest drain along with systemic antibiotics and antifungal medications. He remained afebrile and improved clinically, with resolution of hemoptysis, and was discharged 2 weeks after admission with oral fluconazole and antibiotics. The chest tube was removed prior to discharge. A follow-up contrast-enhanced chest CT 4 weeks after embolization showed decrease in the size of right lower lobe abscess cavity (Fig. 4).

Discussion

The reported patient presented with massive hemoptysis, defined as greater than 100-600 cc in volume over 24 hours [6]. Massive hemoptysis is supplied by the bronchial artery in approximately 90% of cases, often secondary to chronically infected or inflamed lung tissue in the setting of bronchiectasis or cystic fibrosis [7]. Bronchoscopy may be performed to localize and treat bleeding, though bronchial arteriography and embolization may be necessary if endobronchial therapy is infeasible [4]. Arterial phase contrast enhanced CT with multiplanar reconstructions may also identify bronchial arterial pathology and identify common anatomical variants [8]. In this patient CT angiography identified pseudoaneurysms of pulmonary arterial branches, obviating the need for bronchoscopy or bronchial artery embolization.

CT angiography may be superior to catheter angiography in detection of PA pseudoaneurysms, as thrombus, peripheral location, vascular tissue flaps acting as valves, and slow blood flow within the pseudoaneurysm may all impede visualization on digital subtraction angiography [9]. Maximal intensity projections and 3-dimensional reconstructions may be helpful for endovascular or surgical planning [9]. Treatment of PA pseudoaneurysms include coil embolization across the arterial defect, endovascular stent-graft repair, or surgical resection [10–13].

An EPF may occur in patients with esophageal, tracheal, or bronchogenic carcinoma and lead to pulmonary abscess [14,15]. Chemotherapy and radiation therapy may be associated with development of EPF in patients with esophageal cancer [15]. Treatment of EPF is influenced by anatomical and clinical factors, and may involve esophagopulmonary resection, endoscopic placement of self-expanding covered stents, and non-invasive supportive therapy [14]. Placement of a covered self-expanding metallic stent graft has been reported as a safe and effective palliative treatment for patients with EPFs, with clinical success rates greater than 80% [15,16]. Fistulas may recur in 30% of patients after stent placement, but repeat stent placement or balloon dilation may effectively seal reopened EPFs [17]. The mean survival rate in patients with malignant EPF has been reported as 13.4 weeks with a range of 1-56 weeks post stent placement [17].

In summary, we report a rare case of EPF-associated lung abscess causing PA pseudoaneurysms and life-threatening hemoptysis. CT angiography, and minimally invasive percutaneous and endoscopic therapies, may facilitate diagnosis and management with a favorable outcome.

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