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Case Report

Spontaneous esophageal rupture resulting in formation of multiple pulmonary artery pseudoaneurysms: A case report☆,☆☆

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ABSTRACT

Esophageal rupture and pulmonary artery pseudoaneurysm are 2 rare but life-threatening conditions that are challenging to detect but require prompt intervention once diagnosed. The present case describes a patient with spontaneous esophageal rupture resulting in necrotizing pneumonia and multiple PAP that were ultimately managed via endovascular embolization combined with surgical resection. Although atypical, it emphasizes the need for a high degree of clinical suspicion when either of these diagnoses are on the differential and additionally illustrates another scenario in which interventional radiology and surgery can collaborate to optimize patient care.

Introduction

Pulmonary artery pseudoaneurysms (PAP) are rare but portend a poor prognosis as a result of both the risk of rupture, which carries a mortality rate as high as 50%, and the severity of the underlying conditions that lead to their development [1–3]. They are the cause of up to 11% of cases of massive hemoptysis, although they are increasingly incidentally discovered in asymptomatic patients [1,4,5]. Thus, early detection is essential to avoid adverse outcomes but necessitates a high degree of suspicion.

The most common causative etiology is infection, although a variety of both iatrogenic and noniatrogenic causes have been reported. Diagnosis is made by computed tomography angiography (CTA), which shows focal dilatation of a pulmonary artery with adjacent consolidation; catheter angiography is useful when the diagnosis is in question. Treatment has traditionally consisted of surgical resection of the affected lung but endovascular methods have increasingly been shown

Abbreviations: COPD, chronic obstructive pulmonary disease; CTA, computed tomography angiography; ED, emergency department; GERD, gastroesophageal reflux disease; PAP, pulmonary pseudoaneurysm; pRBC, packed red blood cells.

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to be effective, particularly in patients who are poor surgical candidates [1,6].

This report describes a case of spontaneous esophageal rupture resulting in necrotizing pneumonia and multiple PAP managed via endovascular embolization followed by surgical resection.

Case presentation

Institutional review board exemption was granted for this case report. A 67-year-old woman with a history of chronic obstructive pulmonary disease, hiatal hernia with severe gastroesophageal reflux disease on proton pump inhibitor therapy, chronic malnutrition and electrolyte abnormalities requiring frequent infusions, chronic renal insufficiency, and monoclonal gammopathy of undetermined significance presented to an outside hospital with small-volume hemoptysis and was found to be febrile with leukocytosis. A noncontrast CT showed right lower lobe consolidation (Fig. 1), so she was diagnosed with pneumonia and discharged on a 5-day antibiotic course.

Four days later, she returned to the ED with massive hemoptysis resulting in blood-loss anemia for which she was transfused and transferred to the authors' institution. On arrival, vital signs were significant for tachycardia to 100-120 beats per minute, blood pressure 90s/50s mmHg, and oxygen saturation 96% on room air. Labs were notable for a decrease in white blood cell count from 14.4 $\times$ $10^3/\mu L$ to 10.8 $\times$ $10^3/\mu L$ and a hemoglobin of 7.9 g/L after transfusion of 2 units packed red blood cells (pRBCs). CTA of the chest demonstrated extensive right lower lobe consolidation and necrosis containing 2 adjacent PAP; additionally discovered was rupture of the distal esophagus into the right chest (Fig. 2). After multidisciplinary discussion, it was decided to first embolize the PAPs and then surgically resect both the involved lobe and perforated esophagus.

Under moderate sedation, the right pulmonary artery was selected with a 6 French pigtail catheter via right common femoral vein access. A 9 French 75 cm curved Check-Flo introducer sheath (Cook Medical, Bloomington, IN) was then introduced and the feeding branch selected with a 5 French Bernstein catheter (Merit Medical Systems, South Jordan, UT) with 2.4 French Progreat microcatheter (Terumo Corporation, Shibuya, Tokyo, Japan). Angiography confirmed the presence of 2 adjacent PAPs supplied by the superior segmental artery of the right lower lobe (Fig. 3A). Embolization was then performed by injecting liquid embolic (Obsidio, Boston Scientific, Marlborough, MA) to occlude the outflow and then using fibered coils to occlude the inflow (Fig. 3B). Postembolization angiography showed complete occlusion of the pseudoaneurysms with preserved flow to the remainder of the lung (Fig. 3C).

The next day, the patient underwent right lower lobectomy and esophagectomy with latissimus dorsi muscle flap placement. One unit of packed red blood cells were transfused intraoperatively. She was extubated 2 days later and ultimately discharged 1 month after surgery. Pathology confirmed esophageal perforation in the setting of patchy esophageal ulceration; no malignancy was identified.

Discussion

Esophageal rupture and PAP each represent rare but critical emergencies that require prompt diagnosis and intervention. Esophageal rupture is accompanied by a mortality rate of 13%–20% that doubles when treatment is delayed by more than 24 hours [7,8]. The most common cause is iatrogenic, which comprises approximately half of cases; spontaneous perforation underlies 15%–30% [7]. Presentation is nonspecific and dependent in part upon whether the perforation occurs at the cervical, thoracic, or abdominal level. The thoracic esophagus is the most affected, and patients present with chest pain, dyspnea, and subcutaneous emphysema [9]. Diagnosis is best

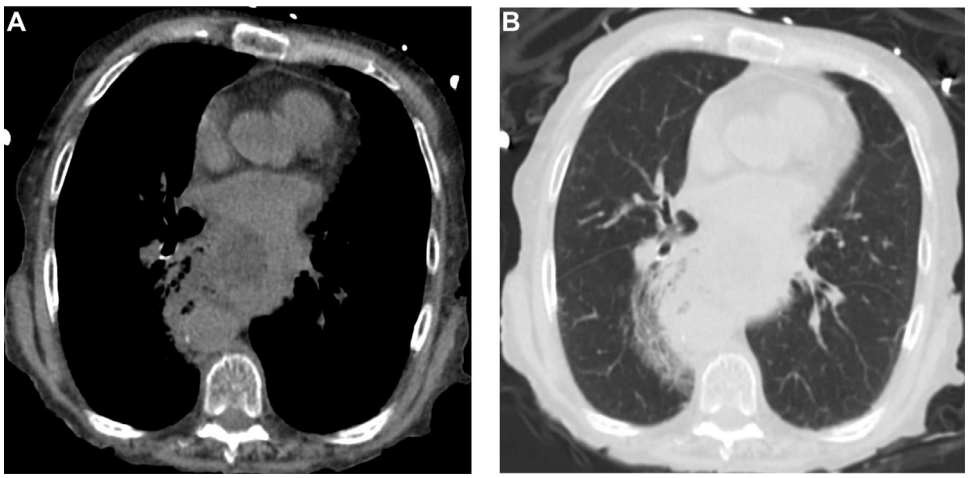

Fig. 1 – Axial noncontrast CT images in mediastinal (A) and lung (B) windows showing early esophageal rupture (arrows) with adjacent right lower lobe consolidation (arrowheads).

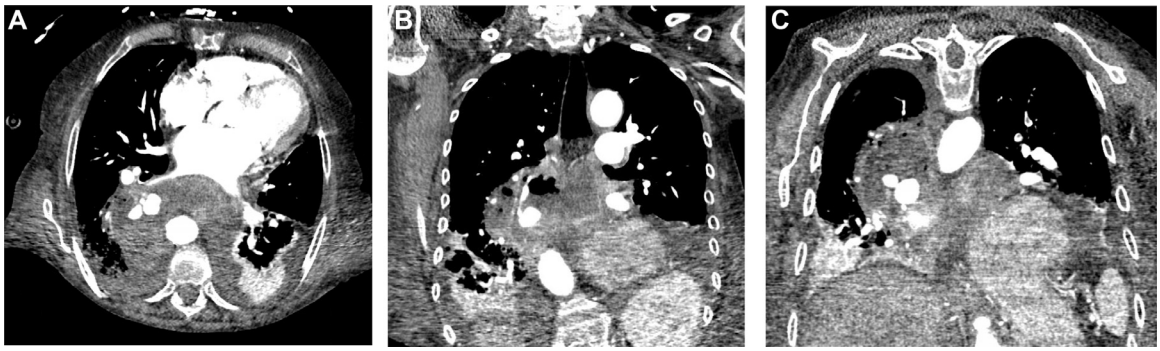

Fig. 2 – Axial (A) and coronal (B, C) contrast-enhanced CT images demonstrating the site of esophageal rupture (arrows) and adjacent PAPs (arrowheads).

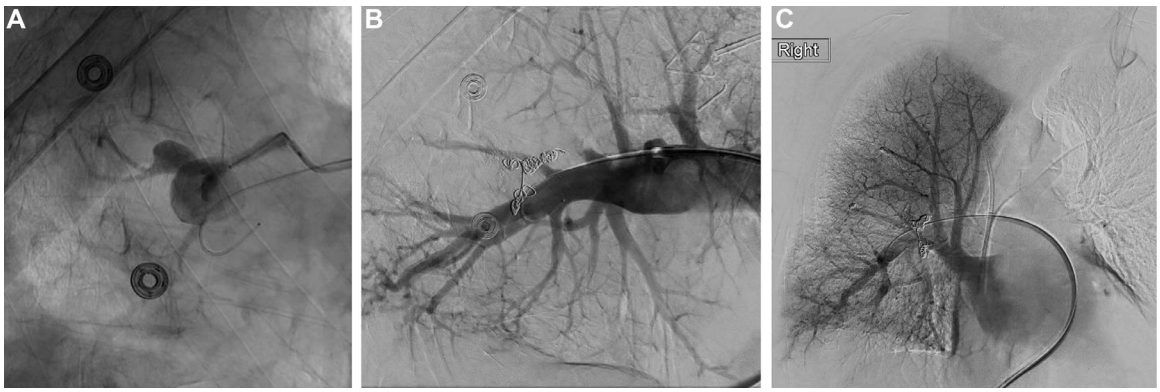

Fig. 3 – (A) Oblique frontal angiogram demonstrating the PAPs. Multiple outflow vessels are opacified. (B) Postembolization frontal digital subtraction angiogram confirming stasis within the embolized arterial branch. (C) Final digital subtraction angiogram demonstrates largely preserved parenchymal opacification.

achieved via contrast-enhanced CT, which has a sensitivity of 92%–100%; swallow studies and endoscopy should be used in equivocal cases [10]. CT findings include esophageal discontinuity, which may be difficult to visualize, pneumomediastinum, pleural effusion, and mediastinal fluid [11,12]. In the present case, esophageal discontinuity and mediastinal fluid were both present on the initial noncontrast CT but difficult to detect; by the time of the contrast-enhanced scan 6 days later, the condition had progressed such that all 4 findings were present, facilitating diagnosis.

PAP can similarly result from a variety of causes, the most common of which are infectious. Commonly cited pathogenic organisms include pyogenic bacteria (Staphylococcus aureus, Streptococcus, Klebsiella, Actinomyces), Mycobacterium tuberculosis (Rasmussen aneurysm), and fungi (Mucormycosis, Aspergillus, Candida albicans), which cause destruction either through hematogenous dissemination or direct spread from adjacent lung [13]. In the present case, the most likely etiology was perforation of a chronically inflamed esophagus, which allowed leakage of pathogenic organisms and digestive enzymes into the right hemithorax. Together these eroded both lung and blood vessels, ultimately resulting in pulmonary cavitation and PAP formation. To the authors' knowledge, this sequence of causative events has not previously been recognized.

This case additionally illustrates the difficulty in making either of these diagnoses. The nonspecific nature of the presenting complaint combined with the patient's underlying renal dysfunction prompted initial evaluation with a noncontrast CT. This allowed for ready diagnosis of the pneumonic process, which adequately explained the initial symptoms and thus obviated the need for further investigation. However, the absence of contrast precluded detecting either of the underlying life-threatening pathologies; it wasn't until the hemoptysis had significantly progressed that the necessary study was performed.

Conclusions

In conclusion, this report describes a case of spontaneous esophageal rupture resulting in necrotizing pneumonia and PAP that were successfully managed via preoperative embolization and surgery. Although atypical, it emphasizes the need for a high degree of clinical suspicion when either of these diagnoses are on the differential and additionally illustrates another scenario in which interventional radiology and surgery can collaborate to optimize patient outcomes.

Consent for publication

Consent for publication was obtained for every individual person's data included in the study.

Data availabilty statement

Data supporting Figs. 1 and 2 are not publicly available in order to protect patient privacy. Anonymized data may be shared upon reasonable request.

Patient consent

We confirm that written, informed consent for the publication of their medical case details was obtained from the patient(s) prior to submission of this manuscript.

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