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Editorial board member of *World Journal of Gastroenterology*, Gülsüm Özlem Elpek, MD, Professor, Department of Pathology, School of Medicine, Akdeniz University, Antalya 07070, Turkey

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Tuberculous esophagomediastinal fistula with concomitant mediastinal bronchial artery aneurysm-acute upper gastrointestinal bleeding: A case report

Sultan R Alharbi

ORCID number: Sultan R Alharbi (0000-0001-7369-9869).

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Sultan R Alharbi, Interventional Radiology Unit, King Saud University Medical City, Collage of Medicine, King Saud University, Riyadh 11472, Saudi Arabia

Corresponding author: Sultan R Alharbi, MD, Assistant Professor, Doctor, Interventional Radiology Unit, King Saud University Medical City, Collage of Medicine, King Saud University, PO Box 7805, King Abdullah Street, Riyadh 11451, Saudi Arabia.
drsultan000@gmail.com

Telephone: +966-11-4761112

Abstract

BACKGROUND

Esophagomediastinal fistula is a very rare complication of tuberculosis in otherwise healthy adults, and mediastinal bronchial artery aneurysm is even rarer. In this case report, we describe a rare case of tuberculosis complication that presented with acute upper gastrointestinal (GI) bleeding. It also highlights the benefits of chest computed tomography (CT) as an excellent adjunct diagnostic tool to endoscopy and bronchoscopy and the role of trans-arterial embolization as a minimal invasive therapy alternative to surgery.

CASE SUMMARY

A 19-year-old medically free male patient presented with acute multiple episodes of hematemesis for 1 d. Upper GI endoscopy, bronchoscopy, and chest CT with IV contrast confirmed esophagomediastinal fistula with mediastinal bronchial artery aneurysm. After resuscitating patient with IV fluid and blood product transfusion, trans catheter embolization was performed for mediastinal bronchial artery aneurysm.

CONCLUSION

We successfully treated a patient with acute upper GI bleeding due to tuberculous esophagomediastinal fistula and mediastinal bronchial artery aneurysm using transcatheter coil embolization.

Key words: Mediastinal bronchial artery aneurysm; Esophagomediastinal fistula; Upper gastrointestinal bleeding; Mediastinal tuberculosis; Case report

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Core tip: A 19-year-old medically free male patient presented with acute upper gastrointestinal bleeding. He underwent endoscopy, bronchoscopy, and chest computed tomography with IV contrast. Diagnosis of pulmonary and mediastinal tuberculosis with esophagomediastinal fistula and mediastinal bronchial artery aneurysms was made. Patient was successfully treated with mediastinal bronchial artery aneurysm coil embolization and antitubercular medications.

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INTRODUCTION

Mediastinal tuberculosis (TB) lymphadenitis is a rare clinical manifestation of TB in adults, and it usually occurs secondary to pulmonary TB. Extrapulmonary lymph node TB was reported to be 17.6% of all TB cases^[1]. Esophagomediastinal fistula formation secondary to mediastinal TB lymphadenitis is an unusual complication that develops as a consequence of erosion of adjacent organs^[2]. Mediastinal bronchial artery aneurysms are extremely rare and can be idiopathic or secondary. Secondary aneurysms are usually related to an underlying inflammatory process like bronchiectasis, chronic bronchitis, or systemic vascular disease^[3,4]. Bronchial artery aneurysm with an esophageal fistula is an extremely rare and potentially fatal diagnosis, and there are few case reports in literature^[5].

CASE PRESENTATION

A 19-year-old medically free patient presented to our emergency department with 1-d history of 5 episodes of hematemesis with moderate amount associated with epigastric pain. His medical history is significant for chronic cough with occasional hemoptysis and weight loss for 5 mo. His vital signs are stable. Patient was pale, but there was no jaundice or cyanosis. Chest and abdominal examinations are unremarkable. Laboratory investigations show normal results except low hemoglobin level of 8 g/dL. Two units of packed red blood cells (PRBCs) were transfused. Upper gastrointestinal (GI) endoscopy shows an opening in the mid-esophagus without clot, bleeding, or mucosal lesions, otherwise a normal esophagus, stomach, and duodenum (**Figure 1**). Tracheoesophageal fistula was suspected, and no definite source of bleeding was identified.

Bronchoscopy was performed and showed no tracheoesophageal fistula but edematous mucosa with nodular lesions in the left lower lobe bronchus. Biopsy was performed. Initial chest X-ray shows cavitory left lung lesion. Three sputum samples were sent for Ziehl-Neelsen (ZN) staining and were negative. Chest computed tomography (CT) with IV contrast was performed and showed evidence of esophagomediastinal fistula, multiple necrotic mediastinal lymph nodes, cavitory lung lesion, and two small mediastinal bronchial artery aneurysms indicating pulmonary and mediastinum TB (**Figure 2**). Patient had another episode of hematemesis with moderate amount. Upper GI endoscopy was repeated and unremarkable except a small opening in the mid-esophagus. Another two units of PRBC were transfused. Conventional angiography was performed and showed bronchial artery arising from the left subclavian artery with two small mediastinal bronchial artery aneurysms. Embolization was performed using coils (**Figure 3**). Histopathological results of bronchial biopsy showed necrotizing granulomatous inflammation suggesting TB, and two additional sputum samples were sent for ZN staining and were positive. Anti-TB medication was started. Patient did not have hematemesis or melena after embolization. Patient was discharged in good condition with anti-TB medications. At 3-mo of follow-up, patient was asymptomatic and had no episode of GI bleeding. Follow-up chest X-ray shows interval improvement (**Figure 4**).

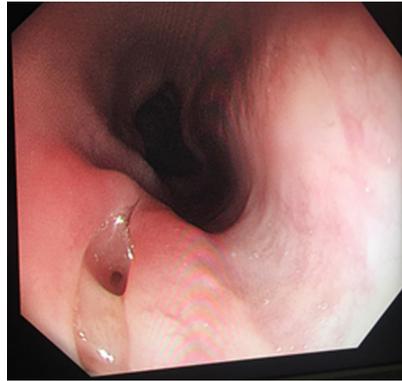


Figure 1 Endoscopy image: An opening is seen in the mid-esophagus without active bleeding or clots.

FINAL DIAGNOSIS

Tuberculous esophagomediastinal fistula with mediastinal bronchial artery aneurysm.

TREATMENT

Patient was treated by mediastinal bronchial artery aneurysm trans catheter coil embolization and antitubercular medications.

OUTCOME AND FOLLOW-UP

After 3 mo of mediastinal bronchial artery aneurysm embolization and initiation of antitubercular medication, patient become asymptomatic and no further episode of GI bleeding was encountered. Follow-up chest X-ray shows interval resolution of lung cavity.

DISCUSSION

Esophageal involvement in TB is extremely rare in otherwise healthy individuals. It mostly affects the mid-esophagus and is secondary to direct extension from the surrounding structures such as mediastinal lymph nodes, lungs, and vertebrae. The usual presenting symptoms are dysphagia or odynophagia^[2]. There are several reports on esophageal TB that appeared as submucosal lesions mimicking a mass, ulcer, diverticulum, or only sinus opening^[1].

Bronchial artery aneurysm is a rare condition with reported rate of < 1% in selective bronchial arteriogram^[6]. It can be classified according to location as mediastinal or intrapulmonary. Clinical presentation is variable depending on the size, location, concomitant disease and if it is ruptured or not. Although it usually detected incidentally on chest CT, the most frequent symptoms are hemoptysis, chest pain, and hemomediastinum^[7]. Chest CT is greatly helpful in detecting TB manifestation in the chest and mediastinal lymph nodes as well as associated fistula formation^[8]. CT angiography and conventional angiography are commonly used tools to diagnose mediastinal bronchial aneurysm^[3].

Mediastinal bronchial artery aneurysms are fatal and even asymptomatic, requiring treatment regardless of the diameter. Endovascular embolization is the first line of treatment, and surgery is reserved as the last option for some patients with contraindication to endovascular therapy^[3,5,7]. Anti-TB medication is the mainstay treatment of mediastinal TB. A large esophageal fistula secondary to TB could be treated by surgery or endoscopic clipping^[2,8].

In this case report, although TB was suspected from the beginning, the presence of acute upper GI bleeding made it challenging to determine its source and treat it. Endoscopy and bronchoscopy – the examinations of choice for upper GI bleeding and suspected tracheoesophageal fistula – failed to identify the source. Chest CT with IV contrast and conventional angiography followed by coil embolization were of great value for diagnosis and treatment of such a rare condition. We believe that TB is a

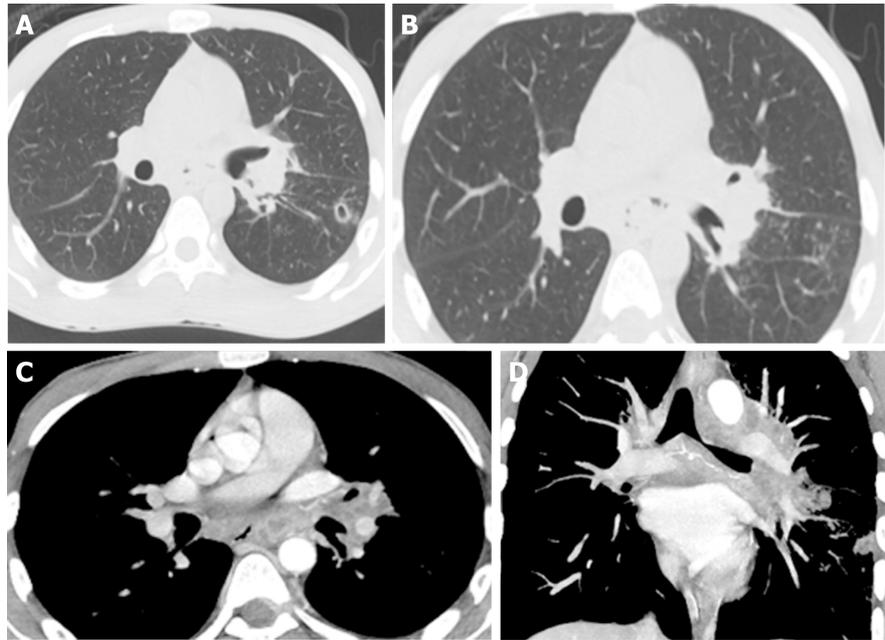


Figure 2 Computed tomography chest. A: Computed tomography lung window axial image shows left lower lobe lung cavitary lesion with multiple nodules in tree-in-bud configuration, a classical sign of lung tuberculosis; B: Chest computed tomography lung window axial image shows multiple mediastinal air pockets anterior to the esophagus, indicating an esophageal fistula; C: Computed tomography chest with IV contrast mediastinal window axial image shows multiple mediastinal necrotic lymph nodes and mediastinal air pocket anterior to esophagus indicating mediastinal tuberculosis and esophageal fistula; D: Computed tomography chest with IV contrast maximum intensity projection coronal reformatted image shows mediastinal bronchial artery aneurysm.

primary disease that involves the lung and then extends to the mediastinum and leads to esophageal fistula and mediastinal bronchial artery aneurysm formation. This rare presentation of acute upper GI bleeding is most likely due to mediastinal bronchial artery aneurysm that caused bleeding and drained into the esophagus by the esophagomediastinal fistula.

CONCLUSION

We successfully treated a patient who presented with acute upper GI bleeding due to tuberculous esophagomediastinal fistula and mediastinal bronchial artery aneurysm using transcatheter coil embolization.

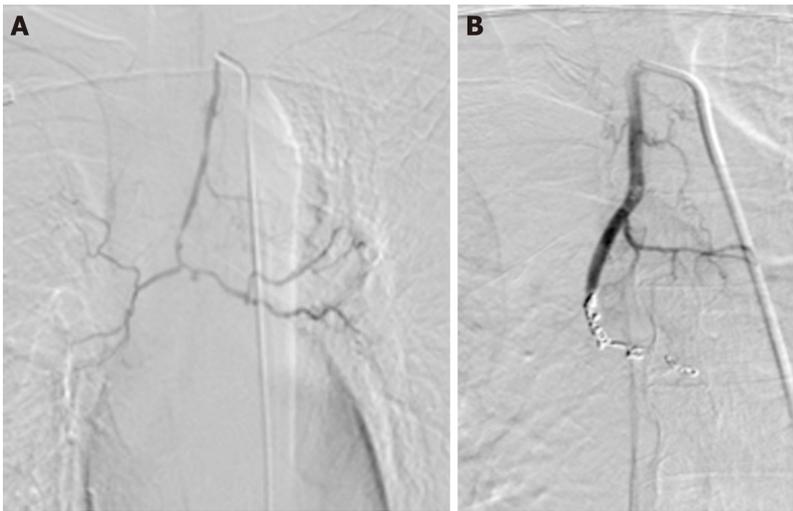


Figure 3 Conventional angiography. A: Selective angiogram of the bronchial artery showing 2 small mediastinal aneurysms; B: Post-coil-embolization angiogram shows complete occlusion of the artery and aneurysms.



Figure 4 Follow-up chest X-ray shows resolution of cavitory lesion and no consolidation. Coils are seen in the mediastinum.

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