

A Unique Case of Esophageal Bleeding: Arterioenteric Fistula Secondary to Intercostal-Bronchial Trunk Pseudoaneurysm Rupture

Tej Mehta, BS^{1,2}, Oluwagbenga Serrano, MD², Cristina Jensen, MD², and Douglas Yim, MD^{1,2}

¹Sanford School of Medicine, University of South Dakota, Sioux Falls, SD

²Avera McKennan Hospital and University Health Center, Sioux Falls, SD

ABSTRACT

Esophageal bleeding has a broad differential. One rare cause of esophageal bleeding is an intercostal artery pseudoaneurysm, which usually presents as hemothorax secondary to trauma or an iatrogenic cause; we identified only 9 reported cases in the English literature. Rarer still is pseudoaneurysm of the intercostal-bronchial trunk, which has not been reported in the literature. We report a 61-year-old man with an intercostal-bronchial trunk pseudoaneurysm who presented with hematemesis and signs of upper gastrointestinal bleeding without previous history of trauma. This case report serves to broaden the differential for esophageal bleeding.

INTRODUCTION

Intercostal artery pseudoaneurysm is a rare but potentially life-threatening cause of gastrointestinal bleeding. Pseudoaneurysms form secondary to disruption of the outer layers of arterial walls, resulting in a balloon-like, blood-filled dilatation of the muscularis propria and adventitia.¹ They may form arterioenteric fistulae, which can result in gastrointestinal bleeding of varying severity. Nine cases of intercostal artery pseudoaneurysm have previously been reported, and none of these reported cases involved intercostal-bronchial trunk (ICBT) pseudoaneurysm.¹⁻⁹

CASE REPORT

A 61-year-old man presented to our emergency department with hematemesis and dysphagia. His past medical history included non-small cell lung cancer (treated with radiotherapy 15 years prior), radiation fibrosis of the posterior mediastinum, hypertension, and gastroesophageal reflux disease. His vital signs included pulse 123 bpm, respiratory rate 26 breaths/min, blood pressure 93/63 mm Hg. Workup revealed hemoglobin 8.7 g/dL and hematocrit 27.8%.

The patient was stabilized, and computed tomography (CT) angiography of the chest, abdomen, and pelvis revealed a focal hyperdensity in the esophageal mucosa, which was originally interpreted by diagnostic radiology as a pill lodged in the esophagus (Figure 1). This hyperdensity was later determined by the interventional radiologist to be a pseudoaneurysm of the right ICBT. Other pertinent findings on CT were calcified right-paratracheal and middle-mediastinal abnormalities with scarring and fibrosis consistent with post-radiation changes, which were the presumed etiology of the pseudoaneurysm. Gastroenterology was consulted.

After CT angiography, endoscopy revealed bright red blood in the first and second portions of the duodenum, the stomach, and the middle third of the esophagus. Large blood clots were found in the mid-esophagus and removed. However, upon removal of these clots, brisk bleeding was observed in the area (Figure 2). This area was injected with epinephrine, yet bleeding persisted. Argon plasma coagulation also failed to stop the bleeding. Finally, 3 hemoclips were deployed, after which no active bleeding was noted. A large, fresh blood clot was seen in the area, and an

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Correspondence: Tej Mehta, University of South Dakota, Sanford School of Medicine, 1400 W 22nd St, Sioux Falls, SD 57105 (tej.mehta@coyotes.usd.edu).



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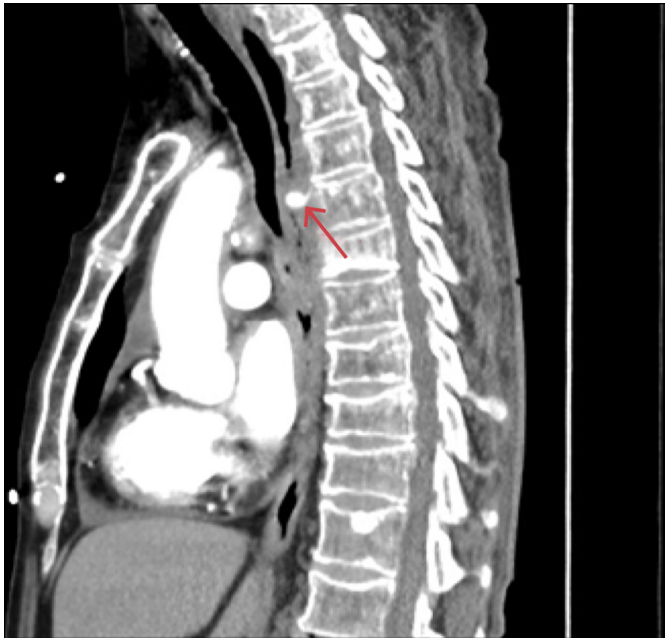


Figure 1. Contrast-enhanced computed tomography before surgery showing an area of hyperdensity projecting into the esophagus consistent with an intercostal-bronchial trunk pseudoaneurysm (arrow).

ulcer approximately 7 mm in length covering more than half of the luminal circumference was visualized. The patient was admitted to the ICU and Interventional Radiology was consulted.

After reviewing the endoscopic findings and the CT angiography with Gastroenterology, it was determined that the suspected lodged pill was actually a pseudoaneurysm, and the patient was taken urgently for fluoroscopic angiography and embolization. Angiogram of the right ICBT showed the bleeding pseudoaneurysm with an arterioenteric fistula to the esophagus (Figure 3). The proximal length of the artery that was the source of the active bleeding was coil-embolized back to the takeoff from the aorta. A post-embolization angiogram showed excellent hemostasis. The patient tolerated the procedure and was transferred to the recovery area.

Follow-up esophagogastroduodenoscopy (EGD) one month later revealed 5 cratered, clean-based ulcers with no

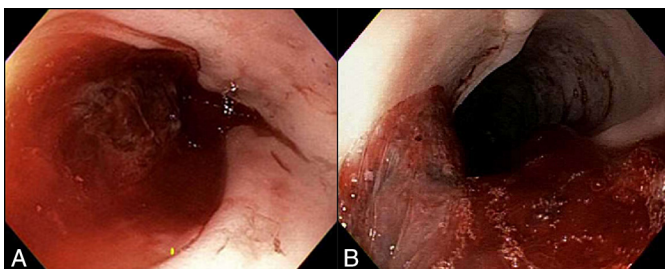


Figure 2. Active, bleeding ulcer of the mid-esophagus.



Figure 3. Angiogram of the right intercostal-bronchial trunk (black arrow) demonstrating extravasation of contrast into the esophagus (red arrow). The blue arrow indicates the catheter in the thoracic aorta with the tip engaged in the intercostal-bronchial trunk.

bleeding, the largest of which was 11 mm in its largest dimension. The previously placed coils were visualized (Figure 4). Biopsies of the area revealed benign squamous mucosa with areas of fibrinopurulent debris. Follow-up CT of the chest revealed a stable coil pack between the esophagus and the aorta without abnormal contrast enhancement (Figure 5). As of the time of this writing, the coils have not caused any apparent adverse effects.

DISCUSSION

Although rare, recognition of intercostal artery pseudoaneurysms is important due to the potential for rapid hypovolemic shock and decompensation. Among the 9 cases of intercostal artery pseudoaneurysm identified in the English literature, 6 cases presented with hemothorax, one presented with recurrent flank pain, one presented as a pulsatile mass, and one

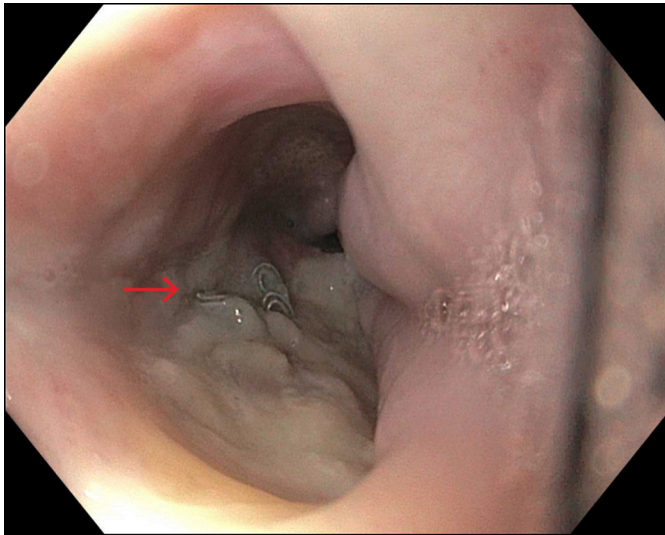


Figure 4. Esophagogastroduodenoscopy after 1 month revealing previously placed coils visible below the mucosa near the area of prior hemorrhage.

was an incidental finding.¹⁻⁹ Of these identified cases, embolization was the first-line treatment and was successful 85% of the time.

The case we have presented is rare in terms of the location of the pseudoaneurysm and the potential underlying cause. No other cases of ICBT pseudoaneurysm were identified in the English literature. This poses the question of whether ICBT pseudoaneurysm is a rare condition or underreported. Moreover, it is possible that ICBT pseudoaneurysms are generally asymptomatic, but when they become symptomatic (as in this case) they quickly become implacable. The presenting symptom of hematemesis in this patient is



Figure 5. Contrast-enhanced computed tomography follow-up demonstrates coil pack (arrow) and no active extravasation of contrast into the esophagus.

unusual and implies the formation of a sudden arterioenteric fistula.

Hematemesis suggests bleeding proximal to the ligament of Treitz, and it may have a “coffee grounds” appearance (suggesting limited bleeding) or be frankly bloody (suggesting moderate to severe bleeding, as in this case). The most common causes of hematemesis include peptic ulcer disease, gastroesophageal varices, and erosive esophagitis.¹⁰⁻¹² In this case, the past medical history of radiotherapy to the area around the ICBT, the presence of post-radiation fibrosis, and the lack of known trauma to the area are suggestive of pseudoaneurysm formation secondary to radiotherapy. Three bronchial and 2 intercostal arterioesophageal fistulae have been reported in the setting of radiotherapy, one of which presented with hematemesis.¹³ Therefore, for our case it is likely that a combination of post-radiotherapy dystrophic changes led to the formation of an ICBT pseudoaneurysm which, coupled with chronic gastroesophageal reflux/erosive esophagitis, caused an arterioesophageal fistula presenting with hematemesis.

Gastroenterology and interventional radiology often play a combined role in the management of gastrointestinal bleeding. In this case of massive hematemesis, the rapid identification and treatment of the bleeding source through CT angiography, EGD, and endovascular embolization highlights the multispecialty approach. The unique anatomy surrounding the esophagus demonstrates the importance of understanding the diversity of pathophysiologies behind esophageal bleeding and highlights the symptoms of arterioesophageal fistulae that should prompt early and aggressive management.

DISCLOSURES

Author contributions: T. Mehta wrote and edited the manuscript. O. Serrano and C. Jensen provided the images. D. Yim edited the manuscript. T. Mehta is the article guarantor.

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Informed patient consent was obtained for this case report.

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