

Case Report

Esophageal Cancer Complicated by a Distal Acquired Esophagopulmonary Fistula

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Abstract

Description

Esophageal respiratory fistulas, commonly found as a tracheoesophageal fistula (TEF), are abnormal connections between the esophagus and trachea. These can be congenital (infants) or acquired (malignancy). A more rare form of an esophageal respiratory fistula is an abnormal connection between the esophagus and the lung parenchyma—also known as an esophagopulmonary fistula. In our case, we present a middle-aged male with a history of esophageal cancer undergoing chemotherapy and radiation presenting into the intensive care unit for increasing shortness of breath and vomiting after eating found to have a rare form of a TEF causing his symptoms.

Keywords

gastroenterology; critical care; esophageal cancer; malignancy complications; fistula; esophageal neoplasms; esophageal neoplasms/complications; esophageal fistula

Introduction

Esophageal cancer is most commonly squamous cell carcinoma, comprising approximately 9 out of every 10 cases worldwide.¹ Tobacco and alcohol use are the two most common risk factors for esophageal squamous cell carcinoma within the United States. Gastroesophageal reflux disease (GERD) and *Helicobacter pylori* are the two most common risk factors for the development of esophageal adenocarcinoma.² The overall peak age of onset is between 50 to 75 years old. Males typically are twice as likely to develop esophageal cancer regardless of the histologic subtype.³ Esophageal cancer should be suspected in individuals presenting with known risk factors—along with persistent dyspepsia despite medically optimized treatment—progressive dysphagia—usually solids first then liquids—and significant weight loss. An esophagogastroduodenoscopy (EGD) is the diagnostic test of choice with endoscopic biopsies making the diagnosis. The most common complication of standard therapy for esophageal cancer (preoperative chemo radiation, surgery or perioperative chemotherapy depend-

ing on the disease stage) is dysphagia and an esophageal obstruction.³ Another complication of therapy is the development of an abnormal connection between the esophagus and pulmonary system. An acquired tracheoesophageal fistula is the most familiar subtype. However, a more rare form is an esophagopulmonary fistula—an abnormal connection between the esophagus and the lung parenchyma. In our case, a middle-aged male, with a history of esophageal cancer undergoing chemotherapy and radiation, presented into the intensive care unit (ICU) for increasing shortness of breath and vomiting after eating and drinking.

Case Presentation

We present a case of a 60-year-old male with a past medical history of esophageal squamous cell carcinoma status post-chemotherapy and radiation. The patient used tobacco and alcohol and was medically noncompliant. The patient presented with a 3 to 4 day history of shortness of breath with productive cough as well as nausea and vomiting after eating or drinking. The symptoms worsened when lying flat. He

Table 1. Emergency Department Laboratory Values.

Test	Patient's Value	Reference Range
Hemoglobin	11.8 g/dl	12.0–18.0 g/dl
Serum leukocyte count	17.4 x 10 ⁹ /L - 88.5% Neutrophils	4.8–10.8 x 10 ⁹ /L
Platelets	363 x 10 ⁹ /L	125–400 x 10 ³ /mcL
Aspartate transaminase (AST)	96 U/L	15–37 U/L
Alanine transami-nase (ALT)	95 U/L	12–78 U/L

stated that when he consumed either food or drink he began to cough persistently, which led to emesis. These symptoms prompted the patient to seek help as he was unable to swallow with worsening shortness of breath.

Pertinent history for his esophageal squamous cell carcinoma shows he underwent a combination of chemotherapy (carboplatin-paclitaxel) for 6 weeks and had 28 radiation visits from the time of his diagnosis in July 2018 to when he presented to the ICU. A recent positron emission tomography (PET) scan revealed localized disease in the esophagus with a decrease in size of the malignant tumor compared to the prior radiograph. He had been referred to multiple thoracic surgeons and was offered the option of surgical resection. Due to insurance issues, this resection was unfortunately delayed, and the patient's condition worsened, leading to the current presentation.

The patient presented to our ICU around 2 months after his last visit with a thoracic surgeon. The patient's initial vital signs included a temperature of 38.6°F, heart rate of 122 beats per minute, blood pressure of 162/84 mmHg

and 92% oxygen saturation on 2 liters nasal cannula. Initial labs revealed a neutrophilic leukocytosis, mild normocytic anemia and liver transaminase elevation. (**Table 1**)

A chest x-ray revealed bilateral pulmonary opacities, suspicion for pneumonia and the possibility of COVID-19. These results led to appropriate testing and isolation precautions. He was started on empiric antibiotics with vancomycin, metronidazole and cefepime. Oncology, gastroenterology and pulmonology consultations were ordered by the primary care physician with admission to the medical floor.

COVID-19 PCR testing was negative. A computed tomography (CT) angiogram was ordered and is shown in **Figure 1**. The findings included a large right upper lobe airspace opacity, consolidation with a large area of necrosis, tissue fragmentation and a thick-wall cavitary lesion with significant pleural thickening medially with fistula formation between the esophagus and lung parenchyma. A barium esophagram confirmed an esophagopulmonary fistula. (**Figure 2**)



Figure 1. A CT Angiogram shows a large right upper lobe airspace opacity, consolidation with a large area of necrosis, tissue fragmentation and a thick-wall cavitary lesion with significant pleural thickening medially with fistula formation between the esophagus and lung parenchyma.

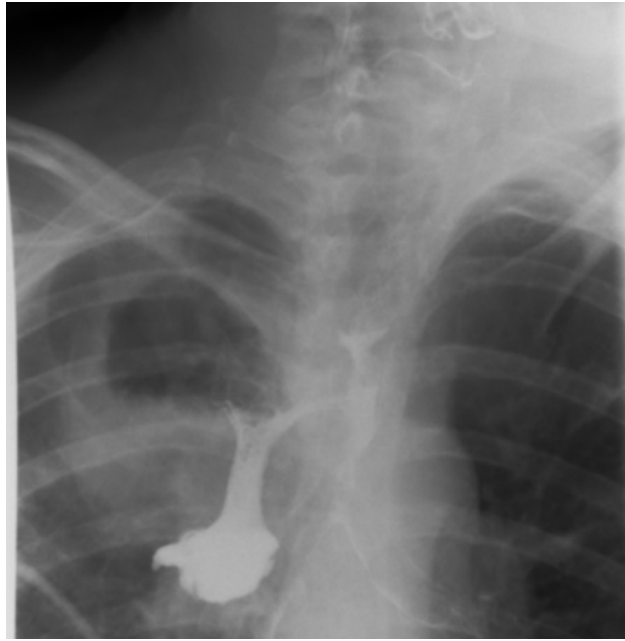


Figure 2. A barium esophagram shows an esophagopulmonary fistula.

An esophagogastroduodenoscopy (EGD) with possible stent placement was planned for the following day. An EGD was completed revealing moderate diffuse gastritis, a friable esophageal tumor from 22–29 cm, and evidence of a 1 cm esophagopulmonary fistula at 23 cm. Unfortunately, the patient went into respiratory distress during the procedure and required an abortion of the procedure prior to stent placement. The patient was then sent to the intensive care unit and required mechanical ventilation. After the pulmonary stabilization and clearance, the patient underwent EGD with the placement of a Boston Scientific WallFlex fully coated esophageal stent. This procedure was well tolerated without complications. A pureed diet was initially tolerated following the procedure with a slow advancement of diet and improving respiratory complaints.

Discussion

This case illustrates a patient with prior localized esophageal squamous cell carcinoma who developed a rare complication that led to debilitating symptoms. The case was complicated by medical noncompliance, and the concurrent COVID-19 pandemic potentially affected an immunocompromised host. Tracheoesophageal fistulas (TEFs) occur in <15% of patients with esophageal cancers, and, if untreated, they significantly increase morbidity and mortality (4). Esophagopulmonary fistulas account for 3–11%

of TEFs and are considered the least common subtype (5). The majority of TEFs are found in the middle 1/3rd of the esophagus. Distal esophageal fistulas are less common and are more prone to an abnormal connection with the major bronchi. Current standard therapy is dependent on whether or not concurrent airway stenosis is present, without which an esophageal stent alone is sufficient. If there is airway stenosis and esophagopulmonary fistula, then double stenting with a bronchial and esophageal stent is indicated.⁵ Prompt recognition of this rare type of TEF in patients with malignancy can lead to early palliative approaches and symptomatic relief.

Early recognition is of utmost importance for clinicians diagnosing TEFs in patients with esophageal cancer. A multidisciplinary strategy involving gastroenterology, oncology, pulmonology and occasionally thoracic surgery is essential to management. Without treatment, mortality increases as do dire consequences, including pulmonary abscess, pulmonary pseudoaneurysms and life-threatening hemoptysis.⁶ For patients deemed poor surgical candidates, an endoscopic approach with stenting can be a safe palliative alternative and can improve quality of life. Future studies are needed to establish the guideline-driven treatment of adults with acquired tracheo- and broncho-esophageal fistulas.

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The patient provided informed consent.

Conflicts of Interest

The authors declare they have no conflicts of interest.

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