Esophagobronchial Fistula Complicating Isolated Esophageal Crohn’s: A Case Report

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Abstract

Isolated esophageal Crohn’s is a rare condition with few reported cases; there are even much fewer documented case reports of esophagobronchial fistula complicating esophageal Crohn’s. We present a case of isolated esophageal Crohn’s with esophagobronchial fistula presenting with hemoptysis. Diagnostic considerations and therapeutic options are detailed. We recommend chest computerized tomography (with computerized virtualization of endoluminal data when available) as the imaging modality of choice, as well as traditional studies: barium swallow, bronchoscopy and upper gastrointestinal endoscopy, for a complete elucidation of all associated clinical entities. We conclude by making the case for entertaining a diagnosis of esophagobronchial or related fistulas in patients with Crohn’s disease who present with interval development of respiratory symptoms or signs especially when these are persistent or recurrent.

Keywords: Esophageal Crohn’s; Esophagobronchial fistula; Hemoptysis

Introduction

Crohn’s disease (CD) is an idiopathic chronic inflammatory bowel disease characterized pathologically by the development of chronic segmental mucosal and transmural inflammation, granulomata and fistulae with adjacent organs. Clinically, CD presents with a myriad of constitutional, systemic and gastrointestinal symptoms and signs. Although any part of the alimentary canal may be affected, CD has a great predilection for the ileum, colon and the anorectal regions of the alimentary tract. CD involving the esophagus is uncommon [1].

There are few reported cases of isolated esophageal CD (probably under-reported) [1-3], and even much fewer reported cases of esophageal CD with esophagobronchial fistula. The estimated incidence of esophageal CD is 0.3-2% and affects as much as 6.5% of the adult and pediatric population with CD [1, 4, 5]. Britenden and colleagues in Leeds reported a case of cough and recurrent pneumonias complicating esophagobronchial fistula in a patient with CD [6]. Dyer et al are among the few who have documented cases of esophageal CD complicated by fistula with the respiratory tract [7, 8].

The history is often that of interval development of respiratory symptoms or signs (coughing upon eating, hemoptysis, recurrent pneumonias) in the setting of pre-diagnosed esophageal or gastrointestinal CD. The diagnosis demands combinations of barium swallow, chest-CT, and endoscopies of respiratory and upper gastrointestinal tract, as well as histopathologic confirmation of biopsied tissue from these endoscopic studies. Intervention is necessary to prevent pneumonic, and hemorrhagic complications. Treatment of lower esophageal Crohn’s complicated by esophagobronchial fistula includes definitive surgery: esophago-gastrectomy and cervical esophagostomy with interval immunomodulatory therapy to control synchronous and metachronous esophageal, gastrointestinal and extra-intestinal disease. We present a case of isolated esophageal Crohn’s complicated by the development of esophagobronchial fistula.

Case Report

AB, a 30-year-old Asian male presented to the ER with hematemesis and hemoptysis of 2 days duration. Medical history was significant for 10-year history of esophageal CD in remission for the past 6 years. The patient was not taking any medication. He reported the absence of fever, night sweats, chills or recent weight loss. There was no recent or remote smoking history. On physical examination, he appeared...
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- with cervical esophagostomy was performed. The biopsy of finding into left main stem bronchus. An esophagogastric fistula was found to have an esophagobronchial fistulous tract extending from the esophagus adjacent to trachea and left bronchus, with minimal bronchial wall thickening suspicious for esophagobronchial/trachea fistula. As patient’s history, chest-CT and endoscopic studies were nearly conclusive, a barium swallow study was not conducted. Intra-operatively, the patient was found to have an esophagobronchial fistulous tract extending into left main stem bronchus. An esophago-gastrectomy with cervical esophagostomy was performed. The biopsy of the tract showed mucosal bridging, filiform polyposis and focal transmural scarring consistent with CD at esophagobronchial fistula site.

The patient did well post-operatively and was discharged to follow-up with the gastroenterology and cardiothoracic surgery units, with plan to institute medical immunomodulatory therapy.

**Discussion**

In the case presented above, we were not burdened with the arduous task of making the initial diagnosis of isolated esophageal CD as the patient had the diagnosis at presentation. A description of the chronology of events preceding the admitting diagnosis is outside the scope and intent of this paper. The diagnosis of an isolated esophageal CD is often a sequel of endoscopy and histopathology of biopsied tissue done for work-up of upper gastrointestinal symptomatology or in patient with extra-intestinal features of CD [5]. In certain circumstances, absent the extra-intestinal manifestations of CD, an incidental endoscopic finding results when investigating a patient for usual symptoms relatable to the upper gastrointestinal tract [5]. Differentiating isolated mid-lower esophageal CD clinically from more common differentials like functional dyspepsia, peptic ulcer disease, GERD and gastroesophageal hernia is difficult [2]. Histopathology, imaging and occasionally, serology and microbiologic techniques may be needed to clinch a definitive diagnosis or at least exclude some differential diagnoses.

The case demonstrates the potential for extension of the inflammatory process associated with CD to, and the development of fistulous connections with visceral and body structures adjacent to the esophagus. Reider and colleagues allude to this potential complication in the course of CD [9]. CD almost exclusively affects the distal two-thirds of the esophagus (with only one reported case of the isolated upper-third involvement) [2]. The relation of the distal two-thirds of the esophagus to the tracheobronchial tree and lungs, the pericardium and the left atrium anteriorly, its surrounding structures in the inferior mediastinum and the respiratory diaphragm inferiorly create the potential for contiguous extension of esophageal inflammation and the development of fistulous connections.

The gravity of inflammatory extensions ranges from clinical trivialities like subclinical para-esophageal inflammation to catastrophic presentations like pan-mediastino-pleuropericarditis, esophageal perforation and fistula formation (with severe recurring aspiration and pneumonias) and life-threatening hemorrhage. It sounds logical to infer that the much less common esophageal involvement in CD and the effectiveness of current medical therapies may explain the rarity of these complications. The differences in thoracic and abdominal anatomy make the potential for development of life-threatening complications more likely with esophageal disease (than with intestinal disease), were the incidence of esophageal and gastrointestinal CD similar.

The complaint of hemoptysis, the finding of fresh blood in the left bronchus on bronchoscopy and the CT-chest report demonstrating extraluminal air and para-aortic/paraesophageal consolidation are consistent with the presence of esophageal perforation and/or esophagobronchial/tracheal fistula. These findings are comparable to radiologic findings by Chaky et al (2008) in their report on chest-CT diagnosis of adult bronchoesophageal fistula [10] and Erasmus et al (2002) report on esophageal perforation [11]. Chaky et al
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mid-third esophageal disease into the inferior mediastinum
events in this case is the subclinical transmural extension of
examination, serologic inflammatory markers and imaging
malnourished and/or taking anti-inflammatory and immuno-
may not be evident in patients with chronic disease who are
chronicity of the inflammation and the tendency to wall-
measurement to accurately assess extravascular loss is required.
the amount of internal hemorrhage. Serial hemoglobin mea-
necessitate appropriate investigation.
Conflicts of Interest
We hypothesize that the most probable chronology of
events in this case is the subclinical transmural extension of
mid-third esophageal disease into the inferior mediastinum
with subsequent metachronous fistula tract development into
the left bronchus. Erosion into the left bronchus resulted in
bronchial hemorrhage and hemoptysis, heralding the devel-
ment of the fistula. We reason that the fistulous process
involved a non-major branch of the left bronchial artery. The
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Competing Interest and Financial Disclosures
The paper received no form of funding or financial support
from any public or private organization. The authors have no
conflict of interest issues to declare.

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