Esophageal Actinomycosis Masquerading as Cancer in an Immunocompetent Patient

Sher N. Baig, MD1, Sadia Rehman, MD1, Mina Daniel, MD1, Vrushak Deshpande, MD2,3, George Abdelsayed, MD, FACG1, and Manuel Gonzalez, MD, FACG2

1Department of Internal Medicine, Richmond University Medical Center, Staten Island, NY
2Division of Gastroenterology, Department of Internal Medicine, Richmond University Medical Center, Staten Island, NY
3Division of Gastroenterology, New York Presbyterian-Brooklyn Methodist Hospital, New York, NY

ABSTRACT

A 79-year-old African American woman presented with acute hematemesis after progressive dysphagia for 6 weeks and 12-pound weight loss. She had no predisposing immunocompromising comorbidity such as the human immunodeficiency virus or active malignancy. Computed tomography showed air-fluid levels within the esophagus with partial obstruction. Upper endoscopy revealed a 1-cm mass lesion in the midthoracic esophagus, and biopsy results surprisingly showed esophageal actinomycosis. The patient’s symptoms resolved on antimicrobial therapy at a one-month follow-up, and the lesion was not seen on repeat endoscopy with biopsy at 3 months. We believe that inhaled corticosteroids for chronic obstructive pulmonary disease may have created the growth milieu by impairing local defenses. Correct inhaler technique, avoiding swallowing the water after mouth rinsing, and a spacer device are recommended to reduce esophageal corticosteroid exposure.

INTRODUCTION

All that looks like cancer is not always cancer. Esophageal actinomycosis frequently misleads as malignancy. Actinomyces is an anaerobic, gram-positive, filamentous rod, which very rarely infects the esophagus if the host is immunocompromised. The occurrence of this in immunocompetent patients is exceedingly rare, and the risk factors remain unknown because only a small number of cases have been published.1–3

CASE REPORT

A 79-year-old African American woman was admitted to our hospital for repeated bouts of small volume hematemesis. She had progressive, painless dysphagia for solids for 6 weeks and had lost 12 lbs. Her medical history was remarkable for squamous cell lung cancer 10 years ago, which was treated with curative lobectomy and smoking cessation. She had chronic obstructive pulmonary disease (COPD) and gastroesophageal reflux disease, but no history of chemotherapy or radiotherapy, tuberculosis, and diabetes mellitus. Her medications at presentation included budesonide-formoterol and albuterol inhalers. Her physical examination was normal. Complete blood count, renal, and liver function tests were normal, and serology for human immunodeficiency virus was negative. A record of a previous endoscopy taken before 4 years showed esophagitis. Differential diagnosis included carcinoma (esophagus, lung) and erosive esophagitis. Thoracic contrast-enhanced computed tomography showed air-fluid levels within the esophagus with partial obstruction and mild-wall thickening (Figure 1). Upper endoscopy revealed a 1 cm friable mass lesion in the midthoracic esophagus, 28 cm from the incisors (Figure 2). Gram stain showed gram-positive, filamentous rods (Figure 3). Special stains (Gomori methenamine silver [GMS]/periodic acid-Schiff) highlighted actinomyces as well as some hyphae and yeast forms of candida (Figure 4). The histopathological analysis of biopsy specimens also demonstrated the pathognomonic sulfur granules with abundant inflammatory exudates around the actinomyces colonies involving the surface epithelium.

The patient was educated about the proper inhaler technique and advised not to swallow the water after rinsing her mouth (which she was not meticulous about). She was also prescribed a spacer device to maximize the lung delivery of corticosteroid, thereby mitigating the amount...
reaching the esophagus. She was discharged on intravenous ceftriaxone, 2 g per day for 4 weeks, and oral fluconazole, 200 mg daily, for 3 weeks. The patient’s dysphagia had resolved at 1-month follow-up with the infectious disease clinic at which she was switched to oral amoxicillin for 6 months. She underwent a repeat endoscopy at 3 months. This time no gross pathology was visible, and the histology of the tissue sample did not show any evidence of persistent infection.

DISCUSSION

Actinomyces species are bacteria that behave like fungi. They frequently grow in the form of mycelia and are water and soil saprophophies. Actinomyces species grow as normal flora in the mouth and gastrointestinal tract. It is an anaerobic, gram-positive filamentous, branching rod. Nocardia shares the same characteristics but only actinomyces produces sulfur granules and only nocardia is acid-fast.4 Esophageal actinomycosis is an extremely rare event particularly in immunocompetent people.1–3 PubMed literature search found only 28 case reports indexed in the English language. Of these, 13 cases were reported in the United States. Two-thirds of patients were suffering from some form of predisposing immunocompromised state, for

Figure 1. Thoracic contrast-enhanced computed tomography showed air-fluid levels within the esophagus with partial obstruction and mild-wall thickening.

Figure 2. Endoscopy showing a friable luminal mass at the mid-distal esophagus (28 cm from incisors) which was biopsied.

Figure 3. Gram stain showing gram-positive, filamentous, rods of actinomyces (arrow).

Figure 4. Hematoxylin and eosin stain showing the pathognomonic large, oval, dark, cotton-ball like sulfur granule of actinomyces (arrow). They are periodic acid-Schiff positive, devoid of sulfur, and consist of densely aggregated microcolonies of actinomyces and cellular debris. Inflammatory infiltrate is seen around the sulfur granules.
example, advanced human immunodeficiency virus, cancer, chemotherapy, organ transplant, end-stage renal disease, or esophageal trauma.1–3,5 Actinomyces causes eroding abscesses after trauma to the mucous membranes of the mouth or gastrointestinal tract. The infection is named according to the area of the body through which the abscess erodes: cervicofacial, abdominal, and thoracic actinomycoses are known classifications.5

Esophageal actinomycosis causes subacute to chronic granulomatous infection which can manifest as esophagitis, esophageal ulcer, an abscess, trachea-esophageal fistula, sinus tracts, or stricture.1–4,6 Dysphagia and odynophagia are the 2 most common complaints.7 Actinomycosis coexisting with carcinoma of the esophagus has also been reported.8 Actinomycosis is a great imitator with the potential to mislead physicians to wrong diagnoses.2 Owing to the progressive obstructive course of symptoms, esophageal actinomycosis raises red flags, thereby triggering an extensive malignancy workup. This patient was otherwise immunocompetent and her history of lung cancer was a strong risk factor for another malignancy. We believe that her use of inhaled corticosteroids for COPD may have created the milieu for the growth of actinomyces and candida in the esophagus. According to gamma scintigraphy studies in the literature, up to 43% of inhaled corticosteroids are indeed deposited in the esophagus.9 Improper inhaler techniques, especially swallowing the water after mouth rinsing, may further increase the esophageal delivery of corticosteroids.

The diagnosis of esophageal actinomycosis involves imaging studies and tissue sampling for histopathological analysis. Microscopy reveals yellow sulfur granules. They do not contain sulfur but are composed of microcolonies of actinomyces and cellular debris.4 Sulfur granules are only observed in 50% of cases, so their absence does not exclude actinomycosis.1 GMS stain highlights these filamentous bacteria. Actinomyces species are difficult to isolate and require prolonged anaerobic culture for up to 3 weeks, but the yield can be as low as 24% and therefore most often not helpful.5 Actinomyces are hardly ever the sole pathogens because other organisms, for example, candida frequently coexist.1 16s rRNA sequencing can provide a quick and accurate molecular diagnosis with sensitivity and specificity above 96%.1,3,6

An initial course of intravenous penicillin G (every 4 to 6 hours) for 4 to 6 weeks is recommended. Ceftriaxone (1 to 2 g every 24 hours) is a reasonable alternative for the ease of daily dosing. Either parenteral regimen is followed by oral penicillin V (2 to 4 g per day, every 6 hours) or oral amoxicillin (1.5 to 3 g per day, every 8 hours) for 6 to 12 months.10 For penicillin-allergic patients, tetracycline and macrolides can be used.8 Surgery is reserved for actinomycosis complicated by a necrotic ulcer, fistula, or abscesses.1 Patient education and counseling are the cornerstones of successful therapy because of the prolonged course of antimicrobial treatment. Patients on inhaled corticosteroids must be educated about the correct inhaler technique and advised against swallowing the water after mouth rinsing. A spacer device should be prescribed to increase the amount of inhaled medication that reaches the airways which would, in turn, reduce esophageal exposure.

Esophageal actinomycosis is a rare cause of dysphagia and can be mistaken for cancer. This case report underscores the importance of attention to underlying local factors in an otherwise immunocompetent patient. Inhaled corticosteroid deposition in the esophagus in asthmatic or patients with COPD, for instance, may increase the susceptibility to actinomyces by depressing esophageal mucosal defenses, although a causal relationship cannot be established because of the current paucity of data. The use of a spacer device should be encouraged to minimize the risk.

DISCLOSURES

Author contributions: SN Baig wrote the manuscript and is the article guarantor. S. Rehman and M. Daniel helped with data collection. V. Deshpande reviewed the literature. G. Abdelsayed revised the manuscript. M. Gonzalez provided the images.

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