Case report

Tuberculosis presenting as bronchoesophageal fistula

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A B S T R A C T

We report a case of bronchoesophageal fistula associated with tuberculosis. A 25 year old woman presented to us with 3 month history of cough worsening with deglutition. Radiological examination revealed mediastinal lymphadenopathy and bronchoscopy with esophagoscopy confirmed the presence of fistulous communication with features of endobronchial tuberculosis. Histological examination of bronchial biopsy specimen showed non necrotic granuloma with the PCR positive for Mycobacterium tuberculosis in her bronchial secretions. She was begun on antituberculous treatment and became asymptomatic after 2 months. Bronchoscopy done during follow up after 4 months showed normal bronchial lumen with disappearance of fistulous tract. Imaging showed resolution of lung lesions.

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Introduction

Noncancerous causes of bronchoesophageal fistula (BEF) are rare and in a majority of the cases are due to trauma or infection, the most common being granulomatous disease [1]. The combination of mediastinal lymphadenopathy and cough following intake of food should alert the treating clinicians about possibility of tuberculous bronchoesophageal fistula. Conventionally, BEF require surgical resection of the fistulous tract. However, a few case reports have suggested that tuberculous BEF can be effectively treated with medical management alone.

Case report

A 25 Year old woman, with no premorbid illnesses presented with a history of cough during eating for 3 months duration and mucoid, non blood tinged sputum production for 1 month. She reported an 8 kg weight loss. There was no history of shortness of breath, chest pain, vomiting or choking. There was also no history of foreign body aspiration, ingestion of toxic or corrosive substances or any surgical procedures in past. Her younger sister was detected to have pulmonary tuberculosis 1 yr back and completed antituberculous treatment. She was not evaluated for Tuberculosis at the time of her sister’s diagnosis. She had been married for 9 months, had regular menstrual cycles and no history of high risk behaviour.

On examination she exhibited mild pallor. There was no icterus, cyanosis, clubbing or lymph node enlargement. Pulse rate – 86/min, regular; Blood pressure – 110/70 mmHg; respiratory rate – 16/min; and she was afebrile. Cardiovascular, respiratory, gastrointestinal, nervous system examination were within normal limits. Investigations showed WBC 11,200/cmm, with 64% neutrophils and 31% lymphocytes, Hgb – 10 g/dl, platelet count 230,000/cmm and ESR was 55 mm/h. Renal and liver function tests were within normal limits. Retroviral screening and autoimmune markers were negative. Sputum AFB was negative. Tuberculin skin test showed an induration measuring 15 × 15 mm.

Barium swallow showed a fistulous communication between esophagus and bronchial tree (Fig. 1). Esophagoscopy was performed which revealed a 30 mm ulcer with irregular borders with communication into respiratory tract, 25 cm from the oral cavity. Computed tomography scan of thorax with three dimensional reconstruction was done which showed mediastinal lymphadenopathy, with erosion of posterior wall of left main bronchus, with a fistulous tract into anterolateral wall of esophagus (Fig. 2A–C). There was also centriflobular nodules in bilateral lung parenchyma with tree in bud appearance. Bronchoscopy revealed inflamed mucosa which revealed granulomatous inflammation on biopsy. AFB staining of bronchial secretions was negative, but tested positive for M.tuberculosis by PCR. Cultures done on bronchial secretions showed growth of M. tuberculosis.

She was started on antituberculous treatment modified according to weight and nasogastric feeding started. Treatment comprised of an intensive phase for the first two months in which four antituberculous drugs (Isoniazid, Rifampicin, Pyrazinamide, Ethambutol) were given. This was followed by a continuation phase for next four months consisting of two drugs (Isoniazid and

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Rifampicin). Bronchoscopy was done after completing 4 months of treatment and showed normal bronchial lumen with disappearance of fistulous tract. Computed tomography of thorax showed resolution of lung lesions without any fistula. She completed 6 months of antituberculous treatment and remains asymptomatic till date.

Discussion

BEF poses a challenge to the clinician for accurate diagnosis which if confirmed can offer the patient a potential cure from repeated pulmonary infections. Patients with BEF usually present with recurrent lower respiratory tract infection. The most characteristic symptom is paroxysmal cough particularly following ingestion of liquids. Some patients are able to avoid the paroxysms of cough by swallowing in the supine position (Ono’s sign) [2]. Other symptoms include fullness of stomach with air following expiration. The fistula does not usually give rise to physical signs.

BEF are divided into two broad categories as congenital and acquired. Brainbridge and Keith classified congenital BEF into four types depending on the site of the fistulous tract [2]. 49% of acquired BEF are malignant in etiology and the rest are secondary to benign causes such as trauma, tuberculosis, actinomycosis and esophageal diverticulosis. (Table 1) [3].

The development of BEF in tuberculosis and other granulomatous diseases are related to mediastinal lymph node involvement [4]. Inflammation in and around these enlarged lymph nodes lead to involvement of neighboring structures or organs particularly the esophagus and the trachea near its bifurcation resulting in periesophagitis and peritracheitis. Subsequent healing with scar
formations may produce a typical traction diverticulum of the midesophagus [5]. If, however, necrosis and caseation occur in the lymph nodes with local abscess formation, secondary rupture into the esophagus, trachea or main stem bronchi results in fistula. There was evidence of numerous necrotic mediastinal lymphnodes in the computed tomography performed in our patient. Henceforth, fistula would have been caused by the erosion of lymphnode rather than a primary bronchial tuberculosis.

The therapy of esophagobronchial communication is usually surgical, performed by division of the fistulous tract and resection of any portion of the lung irreversibly damaged by the suppurative process. If the fistulous tract originates from lymph nodes with no parenchymal complication, simple ligation and resection of the fistula can be performed [6]. However, in our case surgical treatment was not required. In a similar study in 3 patients infected with human immunodeficiency virus presenting with tuberculous bronchoesophageal fistula, antituberculous chemotherapy and nasogastric feeding resulted in healing of all fistulae [7]. Thus, tuberculous BEF if diagnosed early, both the causative process and the complicating fistula may be effectively treated with antituberculous chemotherapy without the need of surgical intervention [8].

**Conflict of interest**

We, authors here by declare that we have no conflict of interest regarding the publication of this article. This article has not been published elsewhere or has not been sent to another journal.

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