CASE REPORT

Spontaneous bronchoesophageal fistula in an adult – A possible delayed sequela of pulmonary tuberculosis

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Abstract  Spontaneous bronchoesophageal fistula in the adult is a rare clinical entity. Most bronchoesophageal fistulae are due to malignancy, prolonged endotracheal intubation or trauma. Granulomatous infections like tuberculosis, HIV and mediastinitis are rare causes of acquired bronchoesophageal fistula. We report a case of a 50 year old man, treated for pulmonary tuberculosis 15 years ago, who developed a spontaneous bronchoesophageal fistula between the mid-esophagus and right main stem bronchus, having no history of malignancy or trauma. Surgical closure of the fistula was done and post operative recovery was uneventful. In this case, the bronchoesophageal fistula probably developed as a delayed sequela of pulmonary tuberculosis as the patient had no active signs of pulmonary tuberculosis clinically or histopathologically.

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Case report

A 50 year old male chronic smoker was referred to us with history of paroxysms of cough and expectoration following intake of liquids and intermittent fever since 3 months. The patient had pulmonary tuberculosis 15 years ago for which he was treated and became asymptomatic after that. The patient had no history of trauma, prolonged mechanical ventilation or malignancy. On examination the patient had tachycardia, mild pallor and bilateral basal crepitations on chest auscultation.

Preliminary investigations including blood counts, erythrocyte sedimentation rate, chest X-ray and sputum for acid fast bacilli were normal. A barium swallow showed a fistulous communication between the mid-esophagus and the right main stem bronchus (Fig. 1). Upper GI endoscopy confirmed the fistulous communication between the esophagus and right main stem bronchus 28cms from incisors. CT scan of the chest showed no evidence of active tuberculosis in the patient other than fibrotic foci and hilar and mediastinal lymphadenopathy. The patient was subsequently explored via a right anterolateral thoracotomy through the fifth intercostal space. The mid-esophagus was dissected, incised longitudinally with a 4 cm incision and communication with right main stem bronchus identified by instilling saline in the open esophagus (Fig. 2).

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Due to extensive fibrosis and adhesions in the posterior mediastinum, dissection and excision of the fistulous tract was not attempted. The fistulous tract was obliterated by plugging it with a patch of autologous pericardium and the ostium approximated with 3–0 multifilament absorbable suture. The esophagus was closed in two layers over a nasogastric tube. Tube feeding was started on the third postoperative day along with parenteral nutrition. Post operative recovery was uneventful. Oral feeding was started after 3 weeks. Biopsy of peribronchial lymph node was suggestive of chronic inflammatory pathology with no evidence of malignancy or active tuberculosis. The patient has been asymptomatic over a follow up period of 9 months and his postoperative barium swallow has been devoid of any fistulous communication.

Discussion

An acquired bronchoesophageal fistula (BOF) is a rare but serious complication of malignancy and trauma. An established patent tract from the airway to the upper-gastrointestinal tract bypasses the normal protection offered by the laryngeal reflexes. Malignancy is the most common cause of acquired bronchoesophageal fistulae. Of these, 77% are attributable to esophageal tumors and 16% are secondary to pulmonary primaries [1–3]. Of the non-malignant etiology of acquired BOF in literature, more than 75% are the result of endotracheal cuff-related trauma in patients subjected to prolonged mechanical ventilation [2]. Secondary erosion of the tracheal and esophageal walls occurs with a 0.3–3% incidence in mechanically ventilated patients. Other causes include trauma to the chest and upper airways, iatrogenic trauma and ingestion of poisonous and caustic substances. Most common etiology of pediatric traumatic BOF is the ingestion of button-type batteries [4]. Although rare, acquired BOF can occur as a consequence of tuberculosis, HIV infection, mediastinitis, histoplasmosis and syphilis.

Patients of BOF usually present with recurrent lower respiratory tract infections. The most characteristic symptom is paroxysmal cough, particularly following ingestion of liquids (Ono’s sign). Some patients are able to avoid the paroxysms of cough by swallowing in the supine position [5]. Other symptoms include fullness of stomach with air following expiration. The fistula does not usually give rise to physical signs but chronic bronchopulmonary suppuration can cause clubbing of fingers, basal crackles or pleural effusion.

The development of BOF in tuberculosis and other granulomatous diseases like histoplasmosis and syphilis are related to mediastinal lymph node involvement [6]. Inflammation in and around these enlarged lymph nodes leads to the involvement of neighboring structures, particularly the esophagus and the trachea near its bifurcation, resulting in periesophageagitis and peritracheitis. Subsequent healing with scar formation may produce a typical traction diverticulum of the midesophagus [7]. If, however, necrosis and caseation occur in the lymph nodes with local abscess formation, secondary rupture into the esophagus, trachea or main stem bronchi could result in the formation of a fistula. Other complications related to mediastinal lymphadenopathy may coexist, including compression of major bronchi by lymph nodes, secondary formation of lung abscesses, bronchiectasis or the middle lobe syndrome [8]. Erosion of a bronchus with intrusion of calcified fragments can give rise to broncholithiasis. Enlarging granulomas may lead to compression of the esophagus and extensive fibrosis resulting in fibrocalcific mediastinitis with superior...
vena caval compression. Primary tuberculous infection of the esophagus is a rare disease and usually occurs following tuberculous mediastinal lymphadenopathy.

Treatment of bronchoesophageal fistula is surgical and done by division of fistulous tract and lung resection if a portion of the lung is irreversibly damaged by the suppurative process. In this patient both lungs were grossly healthy though excision of the fistulous tract would have been hazardous given the close proximity of vital structures.

This case has been labeled a spontaneous bronchoesophageal fistula as a definite underlying cause could not be identified except for the past history of pulmonary tuberculosis. A bronchoesophageal fistula developing as a delayed sequela of pulmonary tuberculosis is a very rare phenomenon. Less than 30 patients with tuberculous bronchoesophageal fistulae have been described in the literature [6,8].

Conflict of interest

We have no conflict of interest to declare.

References