CASE REPORT

Successful medical management of tuberculous broncho-oesophageal fistula

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SUMMARY

Broncho-oesophageal fistula (BEF) of benign aetiology is rare. BEF is a rare complication of intrathoracic involvement with tuberculosis. A high index of suspicion and appropriate investigations can lead to achieving an early diagnosis following which appropriate management can be timely instituted. Surgery can be avoided if the condition is recognised in early stages. We present a case of a young female patient with tubercular mediastinal lymphadenopathy complicated by left broncho-oesophageal fistulisation. Timely initiation of conservative medical management was followed by an uneventful recovery.

BACKGROUND

Broncho-oesophageal fistula (BEF) represents an abnormal connection between the bronchial tree and the oesophagus. They are classified into two types—congenital and acquired. Acquired cases of BEF are generally secondary to malignancy, infections or trauma. Benign causes of BEF are rare. In endemic countries, a large proportion of these may be secondary to mediastinal involvement by tuberculosis.^{1 2} Other infective causes of BEF include histoplasmosis, actinomycosis and syphilis.³ A characteristic history of paroxysms of cough on drinking fluids points towards this diagnosis which can be confirmed by performing a barium oesophagogram. Awareness about this unusual complication in tuberculosis endemic countries can lead to appropriate and timely diagnosis and management.

CASE PRESENTATION

A 15-year-old female patient presented to the pulmonary medicine outpatient clinic with a history of paroxysmal coughing following oral intake of solids and liquids (more for liquids) for 1 month duration. There was history of significant loss of

appetite and the patient had lost nearly 5 kg weight in the preceding 1 month. There was no history of fever, chest pain, haemoptysis or preceding trauma. There was no history of blood transfusion or high-risk behaviour. There was no history suggestive of any cranial nerve involvement or myopathy.

General physical examination was unremarkable and there were no palpable peripheral lymph nodes. Systemic examination including a detailed neurological examination was normal. Tuberculin test was strongly positive with an induration of 20 mm with ulceration. Three sputum smear examinations were negative for acid-fast bacilli. Contrast-enhanced CT scan of the thorax (figure 1) showed enlarged mediastinal lymph nodes predominantly at the right paratracheal (left panel) and subcarinal location (right panel). The nodes appeared heterogeneous with internal areas of peripheral rim enhancement around the necrotic areas. Subsequently, Barium oesophagogram (figure 2) was performed which demonstrated tracking of barium into the left bronchial tree immediately below the level of carina confirming the presence of oesophageal-left bronchial communication.

A diagnosis of tuberculous mediastinal lymphadenopathy with ruptured subcarinal lymph node into the left main bronchus and oesophagus was considered. Flexible bronchoscopic examination of the airways (figure 3 and video 1) demonstrated a small fistulous opening (3×3 mm) having friable edges with air bubbling, on the medial aspect of the proximal left main bronchus approximately 1 cm from the carina. Upper gastrointestinal endoscopy examination also confirmed the presence of a fistulous opening on the oesophageal aspect at 25 cm from the incisors. Endoscopic ultrasound guided fine-needle aspiration from the subcarinal lymph node station yielded frank pus which on cytopathological analysis demonstrated positivity

Figure 1 Contrast-enhanced CT scan of the thorax demonstrating enlarged and necrotic appearing mediastinal lymph nodes at the right paratracheal (left panel) and the subcarinal lymph node stations (right panel).

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Figure 2 Barium oesophagogram examination showing opacification of the left bronchial tree by barium confirming the presence of broncho-oesophageal fistula.

for acid-fast bacilli. Gene-Xpert Mtb-RIF test on lymph node aspirate showed positivity for *Mycobacterium tuberculosis* which was sensitive to rifampicin.

A diagnosis of tubercular left BEF was confirmed. Conservative medical management with nasogastric tube feeding and four drugs antitubercular therapy was initiated. The patient improved and gained weight. A check flexible bronchoscopic and endoscopic examination was performed after 6 weeks of therapy and a significant reduction in the size of the fistula was documented. After 4 months of therapy, repeat endoscopic and flexible bronchoscopy examinations demonstrated that the fistula had healed completely. Nasogastric tube was removed and the patient was initiated on normal oral diet. The patient tolerated the oral diet without any complications and continues to be asymptomatic until now.

Figure 3 Flexible bronchoscopy examination demonstrating the presence of a fistulous opening in the proximal left main bronchus at approximately 1 cm from the carina.

DISCUSSION

Acquired causes of BEF include malignancies involving the oesophagus or the adjacent structures; granulomatous infections; trauma; postinterventions like prolonged endotracheal intubation; endoscopic procedures or impaction by foreign body following radiotherapy and corrosive poisoning.⁴ Congenital BEF is a rare bronchopulmonary foregut anomaly which is generally diagnosed in the neonatal period.⁵ Their proposed pathogenesis is persistence of attachment between the tracheobronchial tree and oesophagus due to abnormal growth of trachea during its separation from the oesophagus.⁶ The commonest location is lower third of oesophagus communicating with the right bronchial tree.⁷

There are no specific clinicoradiological features differentiating congenital and acquired BEF. In surgically resected specimens, absence of normal mucosal lining (bronchial or oesophageal epithelium) in the fistulous tract points towards an acquired cause. Brunner proposed criteria to differentiate congenital from acquired fistulae which include absence of past or present inflammation in the surrounding tissues, absence of adherent lymph nodes and presence of an epithelium lined tract with a muscular layer.⁸

Most of the acquired cases are malignant in origin secondary to oesophageal or lung cancer.⁴ The proposed mechanism of development of a BEF in tuberculosis is the involvement of the mediastinal lymph nodes. Inflammation involves the surrounding tracheobronchial tree and oesophagus leading to either rupture of abscess with caseous necrosis or fibrous scar causing traction diverticulum of the oesophagus with subsequent fistula formation at the tip of the diverticulum. In the recent years, a number of cases of tuberculous BEF in association with HIV infection have been reported.⁹ Tuberculous BEF in children are rare and have been described in reports to be associated with high fatality.¹⁰ Symptomatic presentation of BEF in childhood can either be subacute with repeated episodes of aspiration or an acute presentation with respiratory failure requiring mechanical ventilation.¹⁰

Diagnosis of BEF is usually elusive and often delayed due to the lack of specific signs or symptoms. Patients may present with bouts of cough after drinking fluids (Ono's sign) which is a characteristic symptom. Some patients present with non-specific symptoms of recurrent lower respiratory tract infections, haemoptysis and retrosternal chest discomfort. Barium oesophagogram is the most sensitive means of diagnosis. Bronchoscopic examination and an esophagoscopy should be performed in

Video 1 Flexible bronchoscopy examination demonstrating a small fistulous opening on the medial aspect of proximal left main bronchus.

these patients. CT scan of the thorax allows evaluation for neoplasm's, lymphadenopathy or other associated anomalies. Sometimes, the thoracic CT may also demonstrate a communication between the oesophagus and the bronchial tree.

Treatment of BEF traditionally involved surgical resection or ligation and suturing of the fistulous tract. Patients with recurrent pulmonary infections and bronchiectasis may require resection of the affected lung segment.¹¹ However less invasive, endoscopic procedures can be attempted in poor surgical candidates with malignant fistulae including injection of fibrin glue, application of haemoclips and stenting.¹² Medical management with antitubercular therapy and nasogastric feeds for patients with tubercular BEF has been reported as a successful modality

of treatment.¹³ However, medical management alone may not lead to closure/healing of large-sized fistulae.¹⁰ Also, treatment of tuberculous BEF in the acute phase of the disease can often be very difficult due to the associated risk of airway compromise which might require oesophageal and/or airway stenting.¹⁰ The importance of a high index of suspicion and early diagnosis for conservative management of tuberculous BEF cannot be over emphasised. The need for surgery and its associated complications may be averted with early diagnosis as in our patient.

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Contributors KV was involved in the manuscript preparation; KM and AM were involved in the manuscript preparation and flexible bronchoscopy; S performed upper gastrointestinal endoscopy and endoscopic ultrasound-guided fine-needle aspiration. All the authors contributed equally towards preparation of the manuscript and patient management.

Competing interests None.

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Learning points

- Broncho-oesophageal fistula (BEF) is a rare complication of intrathoracic involvement with tuberculosis and may also be its presenting manifestation.
- In endemic countries, a high index of suspicion of this complication should be maintained in patients presenting with characteristic symptoms especially paroxysmal coughing immediately following intake of liquids.
- Early diagnosis of tuberculous BEF can allow timely institution of conservative medical management which can avert surgery and lead to an uneventful recovery in most of the patients diagnosed early.

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