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Original Article

Clinical profile and management of tuberculous bronchoesophageal fistula

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Abstract

Background and Objectives: Tracheoesophageal/bronchoesophageal fistula is a rare clinical condition, and occurs due to a variety of disease processes. This report describes the clinical profile, management, and outcome of bronchoesophageal fistulas due to tuberculosis in five patients. **Patients and Methods:** Patients diagnosed with esophageal tuberculosis over the last eight years were included. Details regarding the demographics, symptomatology, barium swallow, upper GI endoscopy, with biopsy and high resolution computed tomography of the chest were recorded for patients with tracheoesophageal fistula. The diagnosis was confirmed by acid fast bacilli (AFB) positive fluid aspirate/brush cytology from the fistula, lymph node biopsy showing caseous necrosis or AFB bacillus and tissue tuberculosis culture and polymerase chain reaction (PCR). **Results:** There were five patients (four males and one female) with a mean age of 43.8 ± 17 years (range, 17 to 59 years). The mean duration of symptoms was 38 ± 7 days. The most common symptom was coughing on swallowing followed by dysphagia. Two patients had concomitant pulmonary tuberculosis; two had human immunodeficiency virus (HIV) infection, and one was a post-renal transplant. The diagnosis of tuberculosis was established in all five patients with esophageal cytology, lymph node biopsy, and tissue tuberculosis PCR. All the patients were successfully treated with a combination of antituberculous drugs (five patients), glue application on fistula (one patient), Percutaneous endoscopic gastrostomy (PEG) tube insertion (three patients), and surgery (one patient). **Conclusions:** Tuberculous bronchoesophageal fistula is a rare complication and can be successfully managed predominatly with a combination of antituberculous treatment, PEG tube placement, and rarely surgery.

Key words Antituberculous treatment, bronchoesophageal fistula, coughing on swallowing, dysphagia, percutaneous endoscopic gastrostomy tube, PEG tube placement, tuberculosis

Introduction

Bronchoesophageal fistulae, which may be congenital or acquired, have been rarely reported in literature,[1-3] and their etiology has been associated with cancer, trauma, and infections.[1,4,5] The latter includes tuberculosis, histoplasmosis, syphilis, and actinomycosis.[1,4] Tuberculosis is the most common cause of infective bronchoesophageal fistula.[6]

It is rarely reported in literature,[7] however, its incidence appears to be increasing mainly among immunosuppressed patients, especially in developing nations. Bronchoesophageal fistula poses a challenge to the clinician for accurate diagnosis, which if made can offer the patient a potential cure from repeated pulmonary infections. The aim of this study is to report our experience with five patients of bronchoesophageal fistula over the last eight years. This report describes the clinical profile, management, and outcome of bronchoesophageal fistulas due to tuberculosis.

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Patients and Methods

Patients diagnosed with esophageal tuberculosis over the last eight years were included. Of them, five had bronchoesophageal fistula. The records of these patients were reviewed for demographic characteristics, clinical features, laboratory tests, imaging tools (chest radiograph, barium esophagogram, high resolution computed tomography (HCRT)), upper GI endoscopy, and biopsy.

Diagnosis was confirmed by an AFB-positive fluid aspirate/brush cytology from fistula, lymph node biopsy showing caseous necrosis or AFB bacillus and tissue TB culture and tuberculosis polymerase chain reaction.

Results

A total of 18 patients with confirmed esophageal tuberculosis diagnosed over the last eight years were evaluated; of them, five patients (27.7%) had bronchoesophageal fistula. There were four males and one female, with a mean age of 43.8 ± 17 years (range, 17 to 59 years). Out of five patients, three were immunocompromised (two were HIV positive - CD4 count <150 and one post renal transplant) [Table 1]. Mean duration of symptoms was 38 ± 7 days. Most common symptom was coughing on swallowing (three patients) followed by dysphagia in two, and dysphagia and coughing on swallowing in one. Two patients had concomitant pulmonary tuberculosis.

The diagnosis of tuberculosis was established in all five patients with esophageal cytology, lymph node biopsy, and tissue tuberculosis PCR. The barium swallow of one patient showed tracheoesophageal fistula and in the four others showed left bronchoesophageal fistula [Figure 1]. Esophagoduodenoscopy showed a fistulous opening, between 28 and 32 cm, with two patients having large openings (<5 mm) [Figure 2]. Three patients had surrounding superficial ulceration near the fistulous opening. High resolution computed tomography (HRCT) in all five patients showed paratracheal or subcarinal lymph nodes, and in three, necrosis. Tissue tuberculosis polymerase chain reation was positive in the last two patients.

All patients received antituberculous treatment and Ryles tube feeding initially. At the end of one month, one1 patient showed healing of the fistula and one patient underwent glue application with a hemoclip for recurrent aspiration pneumonia, because of a large fistula. The latter patient underwent surgery. However, three other patients did not show healing and in view of their immunocompromised state, PEG tubes were inserted and antituberculosis treatment continued. In two patients, the PEG tubes were kept for three months and in one patient it was kept for six months, as the patient developed hepatotoxicity and was on modified treatment and later changed to antituberculosis treatment. Healing of the fistula was confirmed by endoscopy and barium swallow. All the patients were successfully treated with antituberculosis treatment, which was given for 12 months.

Discussion

Esophageal tuberculosis is rare, but appears to be increasing, especially in immunocompromised patients. *Mycobacterium tuberculosis* is the most common cause of infectious etiology of acquired bronchoesophageal fistula[6] and its management poses a challenge for the clinician. The pathogenesis of tracheoesophageal fistula[8-10] has been attributed to the following mechanisms: (i) Rupture of the caseonecrotic subcarinal lymph nodes into the esophagus and trachea; (ii) development of traction diverticula between the respiratory tree and esophagus; and (iii) erosion of primary tracheal ulcers into the esophagus.

Although traditionally the treatment of tuberculous esophagotracheal or esophagobronchial fistulae has been surgery, recent reports have favored antituberculous medication.[4,8] In recent times, it has been shown that bronchoesophageal fistula can be managed mainly

Table 1: Demographic and clinical profile of patients

Patient	1	2	3	4	5
Age	17	59	38	50	58
Sex	M	M	F	M	M
Symptoms	Dysphagia	Cough on swallowing	Cough on swallowing	Cough on swallowing	Dysphagia and cough on swallowing
Immunocompromised status	No	No	Yes	Yes	Yes (post-renal transplant)
PTB	Yes	No	No	Yes	No
Investigations					
Endoscopy: Site	30	28	30	32	30
Fistulous opening (small/large)	Large	Small	Large	Small	Small
HRCT (necrotic lymph node)	Yes	Yes	Yes	No	No
Esophageal brush/cytology	Positive	Negative	Negative	Positive	Negative
Lymph node biopsy	Not done	Pos	Pos	Not done	-
TB PCR	NA	NA	Negative	Positive	Positive

PTB=Pulmonary tuberculosis, HRCT=High resolution computed tomography, TB PCR=Tuberculosis polymerase chain reaction

Figure 1: Barium swallow-fistula between the esophagus and left bronchus

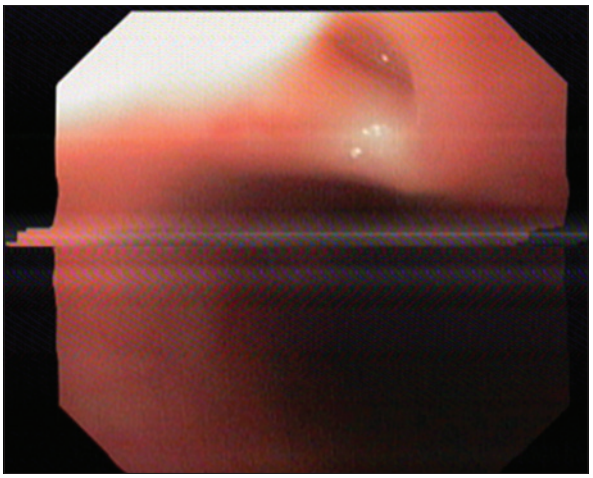

Figure 2: Endoscopic image-fistulous opening in the esophagus

conservatively, with antituberculous therapy and nutritional support. Nasogastric tube placement has been used to permit healing of the fistula. However, PEG tube placement may be preferable to a nasogastric tube to avoid physical contact of the catheter with the fistula, favoring mucosal repair and preventing gastroesophageal ascending reflux/colonization along the nasogastric catheter.

Patients with bronchoesophageal fistula usually present with recurrent lower respiratory tract infection, and cough, particularly following ingestion of liquids, is the most characteristic symptom. Some may present with cough, hemoptysis, weight loss, dysphagia, fever. Ono's sign refers to the uncontrolled coughing, on ingestion of liquids, and crepitations, posteriorly, over the sixth right intercostal space (IC) space, which is pathognomonic of tracheoesophageal fistula.[11]

However, surgical repair may be required for large fistulas complicated by recurrent pneumonia. Four out of five patients were cured with medical therapy, although in one, due to recurrent episodes of aspiration pneumonia, initially a hemoclip with glue application was tried and later surgical repair was done. In another study by Devarbhavi *et al.*, from India,[12] four cases of tracheoesophageal fistulae were managed effectively with medical treatment alone. In yet another study by Joanna Porter *et al.*, three immunocompromised patients were managed with antituberculous therapy and nasogastric tubes.[13]

Patients who require prolonged duration of enteral feeding were treated with PEG tube placement for feeding. In recent times, removable stents have also been used in the treatment of bronchoesophageal fistula; however, they are expensive. Pageno *et al.*,[14] treated a HIV patient with tracheoesophageal fistula for 127 days with enteral feeding (PEG). In our case, three patients were managed with PEG tubes.

Conclusions

Tuberculous tracheoesophageal fistula can be efficiently managed with antituberculous drugs, however, large fistulous openings with recurrent aspirations may require early surgical approach. Enteral feeding with a PEG tube is helpful in immunocompromised patients, who may require a longer duration of medical treatment for healing of the fistula.

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