BRONCHOESOPHAGEAL FISTULA AND DYNAMIC BRONCHOSCOPY

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Introduction

Fistulous communication between tracheobronchial tree and esophagus may present in adult life [1,2]. Cough after swallowing may herald the onset of a tracheoesophageal or a bronchoesophageal fistula [3]. Such fistulae are suggested by the clinical symptom of recurrent bouts of coughing after drinking. In highly suspicious cases, careful dynamic bronchoscopic examination will demonstrate presence of such fistula on deep inspiration [2]. Contrast studies of esophagus readily demonstrate such a fistula and confirm the diagnosis [3]. Esophagus should be investigated more frequently when patients have recurrent respiratory complaints. We present a case of bronchoesophageal fistula diagnosed by esophagography and dynamic bronchoscopy.

Case Report

A 35 year old lady reported with history of chest pain and cough for three months and fever of two weeks duration. Her symptoms used to increase after swallowing. She gave history of dyspeptic symptoms and long standing dysphagia for past 5 years. There was no history of tuberculosis, malignant disease, smoking, chest trauma or exposure to inhaled toxic agents. She earlier underwent upper gastrointestinal tract (UGIT) endoscopy twice in the past for dysphagia, which revealed narrowing at lower end of esophagus but smooth mucosa. Stomach and duodenum were normal. Biopsy from the narrowed esophageal lesion revealed columnar mucosal lining with plasma cell infiltration in lamina propria. No dysplasia, neoplasia or granuloma was detected.

On examination she weighed 42 Kg, had mild fever (temperature 99° F), pulse 86/min, blood pressure was 120/80 mm Hg and had respiratory rate of 20/min. There was no pallor, icterus, clubbing or lymphadenopathy. Examination of chest revealed coarse crepitations at right infrascapular region. There was no tender lump or hepatosplenomegaly on abdominal examination. Other systems were essentially normal.

Her haemoglobin was 10.3 gm% and total leukocyte count was

10800/cmm with P75L48E2M5, sputum was negative for acid-fast bacilli and Mantoux test was non-reactor. Biochemical parameters were normal. Plain radiograph of chest revealed few non-homogenous opacities in right lower zone. However, barium swallow (Fig-1) revealed hold up of barium at lower end of esophagus. After a delay, barium was seen going through a fistulous tract from the esophagus to the bronchial tree. Also seen were bronchiectatic changes in right lower zone. Fibreoptic bronchoscopy was done the day after esophagography. There was no abnormality during quiet breathing, but on deep inspiration a fistulous opening was seen in the posterior basal segment of right lower lobe (RLL). Furthermore, white flake of retained barium was exuding out of the fistulous tract. Thus deep inspiration (dynamic bronchoscopy) demonstrated a bronchoesophageal fistula.

High resolution computed tomography (HRCT) of chest confirmed the fistulous communication between esophagus and the posterior basal segment RLL (Fig 2). The latter was consolidated and bronchiectatic too. No lymph node enlargement was seen. She was treated with antibiotics, antacids and H₂ receptor antagonists and showed symptomatic improvement. She underwent thoracotomy and definitive surgery. Follow-up histopathology of excised fistulous tract showed fibromuscular tissue with chronic inflammation. There was no evidence of malignancy or tuberculosis.

Discussion

Fistulous communication involving tracheobronchial tree may be congenital or acquired. Congenital tracheoesophageal or bronchoesophageal fistulae, if not associated with esophageal atresia, may not appear initially until adult life [1]. There are scant isolated case reports of such an entity. Acquired fistula is usually encountered in adults and is secondary to neoplasm, trauma, instrumentation or radiotherapy. The abnormal communication can exist as tracheoesophageal, broncho-esophageal, broncho-pleural, broncho-vascular or broncho-peritoneal fistula [2]. Rajput et al recently demonstrated a rare transpleural fistulous communication between tracheobronchial tree and chest wall due to *Staphylococcus aureus* lung abscess as the

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Fig. 1 : Esophagography demonstrating narrowed lower end of esophagus communicating through a fistulous tract with posterior basal segment of RLL. Also seen are barium filled dilated and crowded bronchi (bronchiectasis) along with contrast in stomach.

underlying cause [4].

Gibson first reported congenital bronchoesophageal fistula in 1696 [1]. Congenital nature of these fistulae is controversial, but may be due to abnormal growth of the trachea during its severance from the esophagus; this abnormal growth produces a persistent communication between the tracheo-bronchial tree and the esophagus [5]. Following features suggest the congenital nature of such fistulae [1,6,7].

- a) Duration of symptoms, usually since childhood.
- b) Absence of past history of infection or malignant disease.
- c) Tracheal or proximal bronchus level of the fistula.
- d) Completed recovery after resection of the fistula.
- e) Associated other congenital malformations eg. sequestration.
- f) Presence of mucosa (usually of the squamous type) and muscularis mucosa, together with absence of adherent lymph nodes.

Braimbridge and Keith classified such congenital respiratory-esophageal fistula into four types [6].

Type 1 : Esophageal diverticulum with a large ostium and a fistula at its tip.

Fig. 2 : HRCT thorax demonstrating contrast in the dilated bronchi of posterior basal segment of right lower lobe and esophagus along with the intervening fistulous tract

Type II : A short tract running directly from the esophagus to the bronchus or trachea.

Type III : A fistulous tract connecting the esophagus to a cyst in the lobe, which in turn communicates with the bronchus.

Type IV : A fistula that runs into a sequestered segment or lobe.

In a typical form, the fistula extends from the antero-lateral aspect of the lower part of the esophagus to a right segmental bronchus as happened in our case. Azoulay et al reported that 4 out of their 7 cases had communication between segmental bronchus and were right sided [1]. Presenting symptoms include cough (96%), recurrent lung infection (56%) and bouts of coughing when swallowing liquids - Ohno's Sign (65%), pathognomonic of the condition [6].

The clinical profile of our patient did not support tuberculosis or malignancy as the underlying cause. Trauma due to instrumentation eg. upper GIT endoscopy too appears unlikely as no dilatation of esophagus was attempted at any stage and post endoscopy period was uneventful. She had a long standing dysphagia of 5 years suggesting that fistula was probably of a congenital origin. Development of cough and lung infection suggested bronchoesophageal fistula, which was confirmed by esophagography and dynamic bronchoscopy.

The fistula may be demonstrated incidentally in some cases and also present for the first time in adult life [1]. The long, silent interval until adulthood has been explained due to an occlusive valve like mucosal flap, which may close during swallowing because of

the oblique direction of fistulous tract, or spasm of the muscular layer occluding the lumen during swallowing [7]. Bekoe and co-workers suggested that the late onset of symptoms may be due to the fact that mild initial complaints were not thoroughly investigated until complications appeared [8].

Esophagography is the most useful investigation and provides a diagnosis in more than 65% cases. Other diagnostic techniques include esophagoscopy, bronchoscopy, instillation of methylene blue into esophagus during bronchoscopy or into the trachea during esophagoscopy [1,7]. Esophagography in fact clinched the diagnosis of fistula in this case (Fig-1).

Dynamic bronchoscopy [2] : Although bronchoscopy is by design a dynamic procedure, the dynamism applies more to the instrument and the bronchoscopist during the procedure. A normal appearing bronchial tree may show an abnormality on deep inspiration. Our case showed no apparent abnormality during quiet breathing whereas deep inspiration revealed a broncho-esophageal fistula. In addition retained barium was seen coming out of the fistulous tract from the posterior basal segment of RLL. Computed tomography has replaced bronchography to assess the coexistent pulmonary lesion. Bronchography in fact may fail to contribute to the diagnosis and actually lead to a false diagnosis of bronchiectasis [1]. HRCT confirmed the fistulous communication between esophagus and posterior basal segment of RLL and excluded malignancy and tuberculosis as underlying causes.

There is no controversy about the necessity for surgical management of these fistulae. Division of two main ends and complete resection are the main methods. However, a pleural or a muscular flap can be inserted between the esophagus and the respiratory tract to prevent re-fistulization. The diseased lung tissue should be resected at the same time [1]. Our patient too underwent excision of fistula along with in-

section of pleural flap. Communication between tracheobronchial tree and esophagus may persist until adult life. This condition is usually benign but may lead to life threatening complications if left untreated. Recurrent pulmonary suppuration, eg. pneumonia, bronchiectasis and lung abscess are the most common complications. Such respiratory-esophageal fistula should be suspected when these complications occur. Esophageal contrast studies usually confirm the diagnosis. Surgery is the treatment of choice and prognosis is excellent. Esophagus should be investigated more frequently when patients have recurrent respiratory complaints, since pulmonary symptoms may reflect oesophageal disease.

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