Case Report

Benign broncho-esophageal fistula in an adult

K. N. Singh, S. Pradhan, B. Thapa, U. K. Shrestha, P. Sayami

Cardiothoracic and Vascular Surgery Unit, Department of Surgery, Tribhuwan University Teaching Hospital , Kathmandu, Nepal.

Correspondence to: Dr. K. N. Singh, Department of Surgery, Tribhuwan University Teaching Hospital , Kathmandu, Nepal.

e-mail: kumar.drnishant@gmail.com

Abstract

Tracheoesophageal and bronchoesophageal fistulas are usually diagnosed early in life after an infant presents with difficulty in feeding or recurrent pneumonia. These conditions rarely present in adulthood. We report the case of a 65-year-old man who presented with chest discomfort and cough particularly after meals for three months. Workup including a barium esophagogram revealed broncho-esophegeal fistula. Dissection of the fistula and pedicled intercostal muscle flap repair was done.

Introduction

Most bronchoesophageal fistulas (BEF) in the adult population develop as a result of locally advanced bronchogenic or esophageal malignancy. Benign causes of BEF are rare and in majority of cases due to trauma or infection, the most common being granulomatous disease.[1] These patients often succumb to sudden aspiration. In contrast, patients with benign BEF often have a relatively insidious clinical course. The combination of nonspecific symptoms and the rarity of benign BEF often result in long intervals without diagnosis and proper treatment.[2] Identification and treatment is required to prevent re-current infections, chronic lung changes, inflammation, and even pulmonary sepsis.[3]

Case Report

A 65 years male presented with chest discomfort and cough particularly after meals for the last three months. He could not recollect any incidence of swallowing bones or sustaining chest trauma. He had occasional dysphasia, particularly while consuming liquids and drooling of saliva for one and half months. He was a smoker with occasional consumption of alcohol for over 35 years. On presentation to the hospital, the patient had finished a course of intravenous antibiotics for his chest infection. Despite the treatment, he complained of increasing cough and nausea for a month but did not suffer from fever and vomiting.

Physical examination was unremarkable except for inspiratory crackles on the left side of his chest. Chest x-ray revealed calcified retrosternal mass in the anterior mediastinum. Axial CT images of the chest both with and without intravenous contrast were obtained which revealed loculated consolidation left lower lobe, calcified retrosternal anterior mediastinal mass with suspicious communication of left bronchus and esophagus.

Barium esophagogram revealed a communication between the lower esophagus and the left lower lobe bronchus. Upper gastrointestinal endoscopy showed an abnormal opening in the esophagus on left wall approximately 30 cm from incisors, although margin of the opening was smooth and mucosa was normal. Inflammatory changes were seen in the lateral segment of left lower lobe in Bronchoscopy with pus trickling from the periphery of same segment bronchus.

Patient underwent feeding jejunostomy because of poor nutritional status and oral feeding was restricted. His general condition gradually improved and after three weeks he underwent left posterolateral thoracotomy. A fistulous communication between lower oesophagus and peripheral bronchi was identified and in the left lower lobe, there was a localized lung abscess filled with foul smelling pus. Fistulous tract was excised and repaired with interposition intercostals muscle pedicle flap. His post-operative period was uneventful. On eighth post operative day, barium

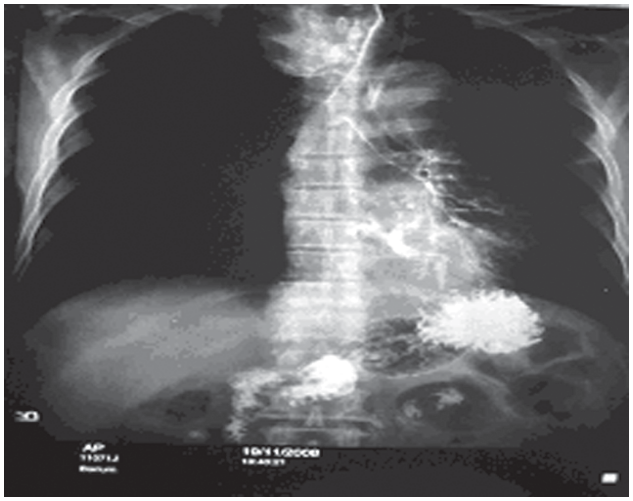

Fig. 1: Pre operative barium study showing broncho-esophegeal fistula

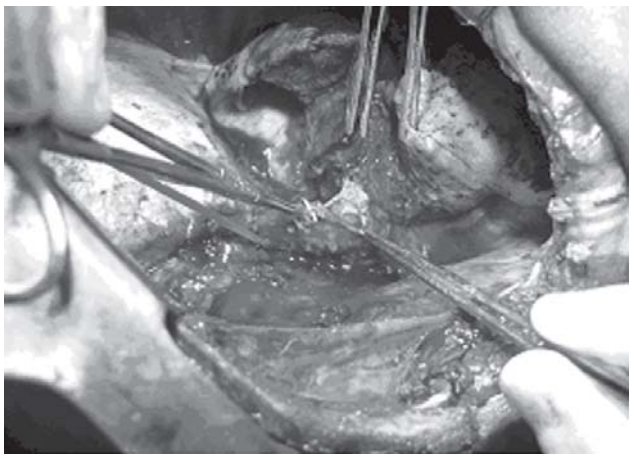

Fig. 2: Intra operative photograph showing fistula

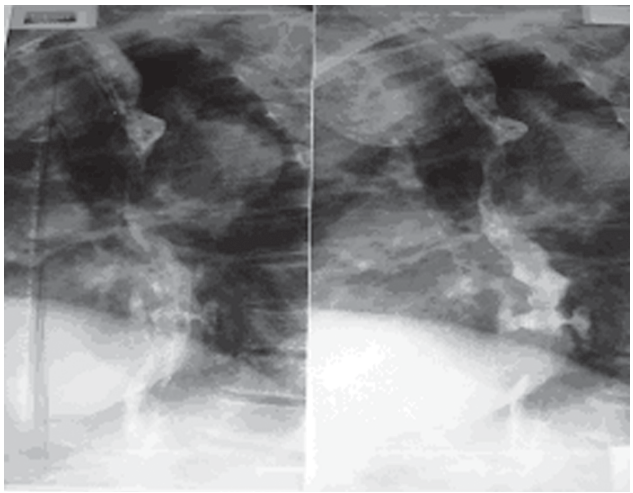

Fig. 3: Post operative barium Study

esophagogram was done. There was no leakage and the patient was allowed to take liquids, subsequently soft and finally a normal diet.

The patient returned for follow up in the clinic with no complaints of cough, chest discomfort or dysphagia.

Discussion

In 1916, Heiderich first demonstrated a fistula that coursed from the esophagus to the right main stem bronchus.[4] Since then, only about 120 cases of benign bronch-esophageal fistula (BEF) have been reported in the world literature. The largest multi-institution series, published in 1966, discussed 260 cases of benign and malignant fistulas in infants, children, and adults.[5] The largest single institutional series discussing benign BEF in adults was published in 1965 by Braimbridge and Keith, who reviewed three cases from their own experience and 20 cases that had been reported in the literature until then.[6] Based on this data, they suggested a classification system which is still in use. Type I fistula originates at the tip of a wide-necked esophageal diverticulum. Type II extends from the esophagus to a lobar or segmental bronchus. These are most common, comprising approximately 60% of all broncho-esophageal fistulas. Type III fistulas extend from a bronchogenic cyst to the esophagus and type IV from the esophagus to a pulmonary sequestration. Our patient had a type II fistula.

Broncho-esophageal fistula should be suspected in any patient who presents with persistent unexplained cough. This is a rare entity and presentation with relatively nonspecific symptoms and signs make the diagnosis challenging.

Barium esophagogram has an important role in the diagnosis of a tracheoesophogeal fistula at any age. However, in adults, a high degree of suspicion is required to prompt the clinician to even consider this diagnsosis. CT is a noninvasive study that can be useful in detecting unsus-pected adult tracheoesophageal fistulas. Malignant, iatrogenic and acquired tracheoesophageal fistulas are believed to be more common in adults than congenital tracheoesophageal fistulas. They are commonly associated with esophageal cancer, surgical complications and caustic ingestion.

Early and definitive surgical intervention with meticulous dissection of the fistula, division, and primary closure of bronchus and esophagus, along with interposition of viable tissue (eg, pedicled intercostal muscle flap, omentum, pericardial fat, or thickened pleura) is the procedure of choice.[7] Other treatment modalities include division of the fistula using a surgical stapling device and endoscopic

application of sodium hydroxide and acetic acid to both the bronchial and esophageal sides of the tract.[8, 9]

Conclusion

High degree of suspicion is required to make a prompt diagnosis of broncho-esophageal fistula. This is critical since early diagnosis and treatment can prevent longterm sequelae such as dysphagia, recurrent pneumonia, obstructive and restric-tive ventilatory defects, and airway hyperreactivity.[3]

References

1. Vasquez RE, Landay M, Kilman WJ et al. Benign esophagorespiratory fistulas in adults. Radiology 1988;**167:**93-93.
2. Abeel A. Mangi et al. Benign broncho-esophageal fistula in the adult, Ann Thorac Surg 2002;**73:**911–5.
3. Kovesi T, Rubin S. Long-term complications of congenital esophageal atresia and/or tracheosdophageal fistula. Chest. 2004 Sep;126**(3):**915-25.
4. Heiderich H. Pathologich-anatomische demonstrationen. Dtsch Med Wochenschr 1916;**42:**340.
5. Blackburn WR, Amoury RA. Congenital esophagopulmonary fistulas without esophageal atresia: an analysis of 260 fistulas in infants, children and adults. Rev Surg 1966;**23:**153–75.
6. Braimbridge MV, Keith HI. Oesophago-bronchial fistula in the adult. Thorax 1965;**20:**226–33.
7. Grillo HC, Wilkins EW Jr. Esophageal repair following late diagnosis of intrathoracic perforation. Ann Thorac Surg 1975;**20:**387–99.
8. Grillo HC, Wilkins EW Jr. Esophageal repair following late diagnosis of intrathoracic perforation. Ann Thorac Surg 1975;**20:**387–99.
9. Smith BD Jr, Mikaelian DO, Cohn HE. Congenital bronchoesophageal fistula in the adult. Ann Otol Rhinol Laryngol 1987;**96:**65–7.