Benign esophagorespiratory fistula: A forgotten cause of chronic lung disease

THOMAS A HORAN, MD, JOHN D URSCHEL, MD

Benign acquired esophagorespiratory fistulas are an uncommon but eminently treatable cause of chronic lung disease. The diagnosis is easily made once it is considered as a possibility. Although medical and endoscopic treatment approaches are suitable in selected cases, surgical therapy is recommended for most patients.

Key Words: Esophageal diverticulum, Esophageal fistula, Tuberculosis

BENIGN ACQUIRED ESOPHAGORESPIRATORY fistula is a rare cause of chronic lung disease. The diagnosis, although simply made, is often delayed because this condition is not normally considered initially. To increase awareness of this cause of chronic lung disease we report a case of esophagorespiratory fistula complicating an esophageal traction diverticulum where the diagnosis eluded physicians for 10 years.

CASE PRESENTATION

A 56-year-old woman presented with a 10-year history of intermittent cough, fever and hemoptysis. Recurrent episodes of right lower lobe pneumonia had been successfully treated with antibiotics. At the age of three she had been hospitalized for Pott's disease of the spine. Her father had died of
tuberculosis. Dysphagia of recent onset prompted further investigation. A relationship between drinking liquids and coughing was not appreciated by the patient until diagnostic investigations were complete.

Sputum cultures were negative for mycobacterium and fungi. A barium swallow demonstrated a wide mouthed diverticulum in the subcarinal region with a fistula to the right bronchus intermedius (Figure 1). Flexible esophagoscopy confirmed the presence of a fistula and served to exclude a malignant etiology.

A right thoracotomy was performed. The diverticulum, fistula tract and adherent lymph nodes were excised. Esophageal and bronchial closures were butressed with an interposed intercostal muscle pedicle. Microscopic examination of the diverticulum demonstrated squamous epithelium surrounded by smooth muscle. The findings were compatible with an esophageal diverticulum of traction etiology. No further episodes of pulmonary infection have occurred in two years of follow-up.

**DISCUSSION**

Benign acquired esophagorespiratory fistulas may result from traumatic injury, foreign bodies, endoscopic instrumentation, prolonged tracheal intubation, granulomatous infections, and esophageal traction diverticuli (1). Fistulas caused by trauma, instrumentation, or tracheal intubation are usually characterized by an acute and dramatic presentation that contrasts with the chronic and insidious nature of fistulas of inflammatory or diverticular etiology. Fistulas of inflammatory and diverticular origin, having clinical and pathophysiological similarities, will be discussed together.

In granulomatous diseases, fistulization occurs as a sequela of chronic mediastinal lymphadenitis. Traction diverticuli, being secondary to granulomatous lymphadenitis, may fistulize by an extension of the original mediastinal inflammatory process or by unrelated diverticular ulceration and perforation (2). While granulomatous infections and esophageal diverticuli were relatively common etiologies of esophagorespiratory fistulas in the past, the reduction in tuberculous infection in the developed world has reduced both the prevalence of the problem and the diagnostic awareness of physicians. A delay of several years in diagnosis is not unusual (2).

The diagnosis of an esophagorespiratory fistula is suggested by a history of coughing after drinking. If the fistula is small, the temporal relationship of coughing after drinking may not be appreciated by patient or physician. An esophagram usually demonstrates the fistula. Bronchographic contrast agents are preferable to barium. Esophagoscopy and bronchoscopy will confirm the presence of a fistula and serve to exclude malignancy. Should difficulty be encountered in demonstrating the communication, installation of methylene blue into the esophagus during bronchoscopy is valuable (3).

Although surgical intervention was recommended for all benign esophagorespiratory fistulas at one time, medical and endoscopic treatment approaches have been successful in carefully selected patients. Management decisions are based upon the etiology and size of the fistula and the condition of the involved lung. Fistulas in the setting of active granulomatous infection may close with antituberculous or antifungal chemotherapy (4,5). If active infection has long passed, which is often the case, antimicrobial therapy is of limited value. Small esophagorespiratory fistulas have been obliterated by endoscopic application of adhesive agents, but experience is limited (6,7). If the fistula is large, or pulmonary resection for irreversibly diseased lung is required, surgical closure of the fistula is preferred.

Surgical management is favoured for most benign acquired esophagorespiratory fistulas. The principles of operative management include fistula division, esophageal and bronchial closure, interposition of local tissue between the two suture lines and concomitant resection of irreversibly diseased segments of lung. Fistula recurrence is unusual (8).

**REFERENCES**

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