Benign bronchoesophageal fistula: Report of four cases

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Benign bronchoesophageal fistula (BEF) is rare and may be characterized by nonspecific symptoms that may delay a correct diagnosis.¹ As soon the diagnosis of BEF is made, immediate surgical treatment is necessary to avoid complications. We present 4 cases of benign BEF that we encountered over a 20-year period.

Clinical Summary

Four patients, 2 male and 2 female, mean age 44.0 ± 25.2 years, underwent surgery for BEF. Clinical and diagnostic characteristics are in Table 1. The initial symptomatology was elusive in all patients: recurrent pulmonary infections, mild dysphagia, and asthma-like crises. These symptoms lasted for several months before the appearance of coughing on liquid ingestion, which is diagnostic for BEF. Plain chest radiography and computerized tomographic (CT) scan showed an irregular mass in the lung fields but were not

decisive in the diagnosis of BEF. Bronchoscopy and esophagoscopy were not always diagnostic, frequently showing only inflammation in the esophagus or bronchus, or both. The definitive diagnostic tool was esophagography with leakage of contrast medium into the bronchus.

Surgery was carried out through a thoracotomy in 3 patients (left in 1 patient and right in 2 patients) and through a cervicotomy in 1 patient. The BEF was excised, and bronchial and esophageal openings were closed with interrupted sutures. A pleural flap was added in 3 patients. Additional surgery, as a consequence of BEF, was required in 3 patients, and a left pneumonectomy, because of bronchial carcinoma, was carried out in another patient. Pathologic examination showed congenital origin in 1 patient, in whom the bronchus and the esophagus were connected through a bronchogenic cyst (Figure 1). Tubercular infection caused the BEF in 2 patients, and trauma was the cause in a young girl who was struck in the neck by her bicycle handlebars.

At a mean follow-up of 12 ± 5.6 years (range 7-20 years), all patients are in good health with no BEF recurrence.

Discussion

Benign BEFs are rare. They can be congenital or acquired. Congenital BEFs, classified by Braimbridge and Heith,² result from failed tracheoesophageal separation in the early stage of embryonic development and, if they are not associated with esophageal atresia, they can be silent for many years until symptoms appear.¹⁻³ A bronchogenic cyst, as in our third patient, is sometime present in congenital BEF. An epithelium-lined tract with muscularis muco-

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Received for publication Oct 10, 2006; accepted for publication Nov 9, 2006.

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J Thorac Cardiovasc Surg 2007;133:1378-9

0022-5223/\$32.00

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doi:10.1016/j.jtcvs.2006.11.007

TABLE 1. Clinical and diagnostic characteristics

	Patient 1	Patient 2	Patient 3	Patient 4
Age (y)	45	8	61	62
Sex	Male	Female	Male	Female
Clinical diagnosis	Likely	Suspect	Likely	Likely
Chest x-ray film	Negative	Negative	Negative	Negative
Chest CT scan				
Plain	Negative	Suspect	Negative	Suspect
After esophagogram with contrast medium	Suspect	Diagnostic	Negative	Diagnostic
Esophagography	Diagnostic	Negative	Diagnostic	Diagnostic
Endoscopy				
Bronchial	Negative	Negative	Negative	Negative
Esophageal	Suspect	Negative	Diagnostic	Diagnostic
Bronchial location	Right main stem	Right upper	Right lower	Right lower
Esophageal location	Medium	Proximal	Distal	Medium
Etiology	Infective	Traumatic	Congenital	Infective
Surgery	Suture + pedicled flap	Direct suture	Suture + pedicled flap	Suture + pedicled flap
Additional surgery	Left pneumonectomy*	Lobectomy	Lobectomy	Segmentectomy

CT, Computed tomographic. *Because of lung cancer.

Figure 1. The probe connects the esophageal (arrowhead) and bronchial (arrow) openings through a bronchogenic cyst (double arrows).

sae and the absence of relevant inflammation around the fistula are conclusive for congenital BEF. Acquired BEFs are more common than congenital types and can be of traumatic, inflammatory, infective, and iatrogenic origin. They may appear at any time during the life span.¹⁻⁴

Over a long period of time, patients with BEF may have nonspecific signs such as vague thoracic or epigastric pain, heartburn, mild dysphagia, weight loss, and recurrent pulmonary infections. Coughing after liquid ingestion is a more specific sign, frequently associated with productive sputum, fever, and hemoptysis. Usually symptoms depend on the fistula size, being more evident in short, wide communications and more discrete in narrow fistulas.

A wide array of diagnostic tools is available in the diagnosis of BEF (Table 1). Plain radiography and CT scan of the chest give information on the lungs and mediastinum, but they may not confirm the clinical suspicion. Endoscopic examinations of bronchi and esophagus may fail to identify the anomalous orifice but

may reveal inflammatory changes, may help in the surgical approach, as in our second patient, and may exclude a malignancy.¹⁻⁴ Esophagography with dilute barium is almost always decisive in the diagnosis of BEF even if sometimes, despite thorough investigation, the diagnosis is made only at surgery. Once diagnosed, benign BEFs require immediate surgical treatment because, if left untreated, they may pose risks of chronic sepsis, airway and lung tissue destruction requiring additional surgery (as was the case in 3 of our patients), and fatal massive hemoptysis.

The principles for successful fistula management are as follows: control of sepsis, establishment of a good nutritional status, pulmonary support and rehabilitation, and surgical repair. Through a thoracotomy, direct suturing or excision of the BEF with interposition of a pedicled flap is the preferred method of treatment. Cervical approach, as in our second patient, should be reserved for higher BEFs, which are indeed very uncommon. Additional lung segmentectomy or lobectomy is necessary when lung parenchyma is involved. Endoscopic treatment and even medical treatment alone have also been described.⁵

In conclusion, a high index of suspicion in every patient with chronic pulmonary infections of obscure etiology is important in establishing a correct diagnosis. Prompt surgical repair gives the best chances of success.

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