Acquired Benign Esophagotracheobronchial Fistula*

Donald R. Judd, M.D., F.C.C.P.
and Theodore Dubuque, Jr., M.D.

Two patients with acquired benign fistulas between the esophagus and the tracheobronchial system are reported. In one, the fistula was the result of histoplasmosis; this is the fifth reported case due to histoplasmosis. We have also reviewed the etiology, diagnosis and therapy of such fistulas.

A cquired benign fistulas between the esophagus and the tracheobronchial system are uncommon. Although most such fistulas are the result of infection, only four have been attributed to histoplasmosis. Since no surgeon will have a large experience with this condition, we are reporting our experience with two cases of benign fistula; one due to histoplasmosis and one probably due to tuberculosis.

Case Reports

Case 1:
A 42-year-old white man was admitted on June 9, 1965, because of three similar coughing episodes. The first of these occurred one month earlier. He swallowed liquid, then felt a "gurgling sound in the throat." This was followed by severe nonproductive coughing. Each of these episodes lasted two to three days; in the interval he was entirely well. There had never been any difficulty with solid food. Because of seasonal allergy, he had received monthly injections of ACTH for the past six months.

Upon admission here there was no abnormal physical finding except for occasional wheezes in both lungs. The chest x-ray examination was normal except for mild emphysema. Roentgen-ray examination of the esophagus with barium contrast demonstrated an irregular traction diverticulum leading to a calcified mass (Fig 1 and 2). Bronchograms were normal except for indentation of the right intermediate bronchus by the calcified mass (Fig 3). No fistula was noted. Skin tests were positive for histoplasmosis and negative for tuberculosis, blastomycosis and coccidioidomycosis. Bronchoscopic examination demonstrated a red, granular, friable mass in the right intermediate bronchus just distal to the origin of the right upper lobe bronchus, occluding much of the intermediate bronchus. This mass was biopsied.

*From the Department of Surgery, St. Louis University School of Medicine and from the St. Louis University Surgical Service, John Cochran Veterans Administration Hospital and St. Mary's Hospital, St. Louis, Missouri.

Figure 1. Traction diverticulum in the midesophagus.

Esophagoscopy demonstrated a small mass of tissue in the mid-esophagus on the right anterolateral wall. This was also biopsied. Histologic examination of the bronchial and esophageal biopsies showed granulation tissue.

He was taken to the operating room with a presumptive diagnosis of benign esophagotracheobronchial fistula. Right posterolateral thoracotomy was performed. There was a calcified lymph node between the middle esophagus and right intermediate bronchus, densely adherent to both structures. The esophagus was dissected free by dividing the fistulous tract along the superior aspect of this lymph node. This left a defect in the esophageal wall, measuring 1.0 × 1.0 cm. This was closed by an inner layer of chromic catgut and an outer layer of silk sutures. The fistula and lymph node were then excised, leaving a 1.0 × 0.8 cm defect in the membranous portion of the right intermediate bronchus. This defect was closed with interrupted silk sutures and covered by a pleural flap.

Histologic examination of the fistula showed chronic inflammation. The lymph node was completely calcified; no histologic sections were possible. Three sputum cultures
area (Fig 4). Roentgen-ray examination of the esophagus with barium contrast demonstrated a fistula between the mid-esophagus and the left main bronchus (Fig 5). Bronchograms did not demonstrate the fistula, but showed severe bronchiectasis in the left lower lobe. The fistula could not be identified with certainty at bronchoscopy or esophagoscopy. A skin test was positive for tuberculosis and streptomycin and isoniazid therapy was started. Two weeks later, left thoracotomy was performed. There were dense adhesions between the lower lobe and the diaphragm and chest wall. These were divided and the fistula between the left main stem bronchus and mid-esophagus was isolated. It measured 8–10 mm in diameter and 10 mm in length. The fistula was divided and the two ends were sutured closed and mediastinal tissue interposed between them. Because of the previously demonstrated bronchiectasis, left lower lobectomy was performed. This lobe showed acute and chronic bronchitis with bronchopneumonia, abscess formation and bronchiectasis. Cultures for tubercle bacilli and fungi were negative. Postoperatively, he did well and has remained free of symptoms.

**DISCUSSION**

In 1965, Anderson and Sabiston\(^1\) found 127 instances of benign esophagotracheobronchial fistula. Since then, others\(^2-4\) have added 17 cases bringing the total to 144. These are differentiated from congenital fistulas which may persist into adult life before they are recognized.\(^3\) Most acquired benign

---

**FIGURE 2.** Arrows point to large calcified mass producing traction diverticulum.

were negative for tuberculosis. Cultures of the fistula and of the calcified lymph node were negative for tubercle bacilli and fungi. Serum sent to the Coccidioidomycosis Serology Reference Laboratory in San Fernando was negative for coccidioidomycosis on two occasions and positive for histoplasmosis. Postoperatively, he did well and barium swallow and bronchograms were performed at intervals of two weeks and again at five months postoperatively with no evidence of fistula recurrence.

**Case 2:**

A 35-year-old white man was admitted on August 15, 1964, because of a chronic cough which had been present intermittently for three years. The cough was often exacerbated by drinking liquids and sometimes by eating solid foods. There had been multiple respiratory infections in the past. In 1961, he had been hospitalized because of hemoptysis. Bronchoscopy then demonstrated bronchitis and bronchiolithiasis. Bronchograms then were normal. In the six weeks prior to this admission, he had noted pleuritic pain in the left lower chest and on one occasion, his temperature was 102° F.

At the time of admission, there was some elevation of the left hemidiaphragm. A pleural friction sound was present in the left lower chest and rales were heard over this area. Chest x-ray film demonstrated a pulmonary infiltrate in the left lower lobe with a nodular mass in the left hilar

**FIGURE 3.** Right intermediate bronchus is indented by calcified mass.
fistulas are caused by trauma or infection. Either direct or indirect trauma may produce a fistula. In 1957, Coleman⁵ was able to find nine cases of esophagotracheal fistula due to nonpenetrating chest trauma. Since then, Wychulis and associates⁴ and Killen and Collins⁵ have added two cases each of esophagotracheal fistula due to steering wheel compression of the chest. Fistulas have also resulted from direct injury of the esophagus. In Coleman’s review of benign acquired fistulas, five cases were due to esophageal dilatation. Wychulis and his associates⁴ reported six cases of fistula after lye ingestion; three without dilatation, and three as a result of esophageal dilatation.

More fistulas are produced by infections than by trauma. The esophagus, trachea, and bronchial system are surrounded by lymph nodes. If these nodes become inflamed, they may enlarge, calcify, or both and erode into a bronchus or the trachea and the esophagus. A variant of this mechanism is the production of a traction diverticulum of the esophagus by an adherent inflamed lymph node. This node may subsequently erode into the trachea or bronchial system and the diverticulum resulting in a fistula. Since the fistula often appears some time after the infection has subsided, the exact cause of the nodal inflammation may be difficult or impossible to determine. Because of this, most of the fistulas of infectious origin are attributed to nonspecific infections. The granulomatous infections are the most common specifically recognized etiologic agents. Syphilis, tuberculosis, and the fungal diseases have all been responsible for fistula formation. Coleman⁶ found 13 cases of fistula due to syphilis. He also found 14 cases due to tuberculosis. Our second case may be due to tuberculosis.

Fungus infections have rarely been diagnosed as the cause of esophagotracheobronchial fistula. Actinomycosis was implicated in one case⁶ and histoplasmosis was thought to be the cause of fistula in only four cases.¹,²,⁷ We believe that the fistula in our first case was the result of histoplasmosis. The presence of calcified nodes, positive skin tests for histoplasmosis alone, and positive serum titers for histoplasmosis make the diagnosis of histoplasmosis reasonably certain.

The symptoms of such fistulas depend upon their location and their size. Classically, the swallowing of liquids results in severe coughing. This may be intermittent, as in our first case. The fistula may result in hemoptysis and in some series, such hemoptysis has been profound. The fistula may result in pulmonary infection with pneumonia, bronchiectasis, or abscess. Wychulis and associates⁴ reported 36
cases of fistula. Of these, 18 had pneumonia, nine had bronchiectasis, three had empyema, three had mediastinal abscess, and one had a lung abscess.

Diagnostic evaluation centers on determining the presence of a fistula and establishing its benign character. When swallowing liquids results in paroxysms of coughing, the diagnosis of a fistula is almost certain if the neurologic examination of the throat is normal. Usually the fistula can be demonstrated with roentgen examination of the esophagus using some contrast material such as thin barium, diatrizoate sodium (Hypaque), or iodized oil (Lipiodol). Esophagoscopy and bronchoscopy are important parts of the evaluation. It may be possible to visualize and biopsy granulation tissue around the fistula, as in our first case. Bronchoscopy has been more productive than esophagoscopy in visualizing the orifice of a fistula. In some instances the injection of methylene blue into the esophagus during bronchoscopy has been helpful. Bronchograms are also important. Only rarely do they demonstrate the fistula, but they are necessary to demonstrate the presence of bronchiectasis and to aid in determining the need for pulmonary resection at the time of surgical repair of the fistula.

Spontaneous closure of these fistulas is rare. The best treatment is division of the fistula and resection of irreversibly diseased lung. Lesser surgical procedures such as endoscopic cauterization are not likely to succeed. The timing of surgical intervention depends upon the patient's tolerance to the fistula and whether or not any pulmonary or bronchial infection can be expected to improve with antibiotic treatment. Since spontaneous closure is rare, the fistula should be treated as early as possible. Delay may result in irreversible lung disease.

Most of these fistulas have been approached by a right posterolateral thoracotomy incision. If the preoperative evaluation establishes the need for resection of the left lung, then left posterolateral thoracotomy should be used. The esophagus is mobilized and the fistula is isolated, then excised. The defects in the esophagus and bronchus or trachea are closed. A flap of mediastinal pleura or other mediastinal tissue is interposed between the sutured structures. Any required pulmonary resection is performed.

Anderson and Sabiston found adequate information concerning treatment and results for 94 of the 127 cases in their review. Twenty-nine were treated non-operatively; five of these underwent spontaneous cures, six were unimproved and 18 died. There were 53 patients treated by resection of the fistula; 47 were cured and six died. Wychulis and associates resected the fistula in 14 of their 36 patients. Thirteen were cured and one patient died.

References

Reprint requests: Dr. Judd, 1325 South Grand Boulevard, St. Louis 63104.

An Unusual Case of Cyanotic Heart Disease*

Atrial Septal Defect with Acquired Tricuspid Insufficiency

Robert G. Fish, M.D., Stewart M. Scott, M.D., John T. Joyner, III, M.D. and William M. Nelson, M.D.

A rare case of tricuspid insufficiency due to bacterial endocarditis in association with a previously asymptomatic atrial septal defect is presented. Intense cyanosis, polycythemia and clubbing of digits were the major presenting clinical features. The classic physical findings of tricuspid insufficiency were not present. Hemodynamic data, blood gas analysis, and cine-angiographic studies are included. Surgical correction of the cardiac defects was successful.

This report describes the clinical picture and successful surgical treatment of a patient with an atrial septal defect and tricuspid insufficiency due to bacterial endocarditis. Prior to cardiac catheterization and cineangiography, the exact nature of

*From the Medical Service and Cardiovascular Surgical Section, Veterans Administration Hospital, Oteen, North Carolina.