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Congenital bronchoesophageal fistula in an adult woman

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Tracheoesophageal or bronchoesophageal fistula (T/BEF), the abnormal communication between the esophagus and the tracheobronchial tree, may present at any time during the life span. In the neonate, T/BEF is a congenital malformation associated with some form of esophageal atresia in the vast majority of cases.^{1,2} Later in life, most T/BEFs are acquired.^{3,4} However rare, if the congenital T/BEF is not associated with esophageal atresia, it may remain silent until it eventually becomes symptomatic. Herein, we report the clinical case of a 53-year-old woman whose congenital T/BEF was diagnosed and treated at our institution.

Clinical Summary

The patient was referred to us as a result of a self-limited episode of minor hemoptysis of sudden onset. At anamnesis, she explained that she had experienced a pain in the upper abdomen 2 years before. Gallbladder stone disease and unilateral renal agenesis were diagnosed by echography, and chronic gastritis was diagnosed by esophagogastroscope. She was then cholecystectomized without postoperative complications. She did not disclose any other previous disease or complaint. Physical examination was normal on admission. Red blood cell, white blood cell, and platelet counts, coagulation profile; routinary serum; and urine analysis results were all within the normal range. Chest radiography and a computed tomographic scan of the thorax displayed an irregular area of lung parenchyma condensation in the upper segment of the left lower lobe. On fiberoptic bronchoscopy, the tracheobronchial tree was normal except for the presence of a blood clot that partly occupied the lumen of the left lower lobe bronchus. Bronchial washing specimens were sent for cytologic analysis, Gram and acid-fast stain, and bacterial and mycobacterial culture; all yielded

Figure 1. Preoperative esophagogram. The contrast medium passes from a diverticulum in the middle esophagus through an ascending fistula to the left lower lobe bronchus.

negative results. Esophagogastroduodenoscopy revealed the presence of a double hole in the left wall of the middle esophagus. Bouts of cough ensued every time the holes were instilled with serum saline, thus establishing the diagnosis of T/BEF. Initial treatment consisted of full oral fasting and total enteral nutrition for 3 weeks. Then, an attempt was made to close the fistula by endoscopic instillation of fibrin glue. However, an esophagogram showed the persistence of the T/BEF (Figure 1), and the patient was scheduled for an open surgical procedure. Dissection of the fistula, closure of both ends with an automatic stapler, division of the tract, handmade reinforcement of both sutures, and interposition of a pedicled pleural flap were performed through a left posterolateral thoracotomy. Postoperative evolution was normal. A second esophagogram showed no spilling of the contrast. The patient restarted oral feeding on the sixth postoperative day.

Comment

Congenital T/BEF seldom presents in adulthood because it is rarely left undiagnosed in the neonate: it is often associated with some other bronchopulmonary and/or foregut malformation (mainly with esophageal atresia).^{1,2} It is only in the absence of this

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association that a congenital T/BEF may remain silent and not be diagnosed until much later, when it becomes symptomatic. There is no explanation for the possible mechanism(s) that make a T/BEF remain silent for long. Occlusion by an imperforated web and/or the ascending (antigravitational) direction of the tract have been advocated.^{1,2} Besides, it is not known what the precipitating events are that cause symptoms to start. When this happens, symptoms are most often respiratory, acute, or chronic. Bouts of cough when swallowing, chronic wheezing, recurrent bronchopulmonary infections, hemoptysis, and hematemesis, either isolated or combined, should raise the suspicion of T/BEF. Diagnosis is made by endoscopy, esophagography, or both. Computed tomographic scanning of the chest is useful because repeated lung infections can lead to bronchiectasis. Fiberoptic bronchoscopy is also mandatory. The congenital nature of a T/BEF is admitted when an acquired cause is reasonably excluded. At pathology examination, an epithelium-lined tract with muscularis mucosae is conclusive. Once diagnosis is made, treatment should be aggressive and promptly instituted, because the acute worsening of the patient's condition due to potentially life-threatening complications, such as massive hemoptysis, is possible at any time. Treatment is as described previously. Endoscopic occlusion is an option to try on an individual basis. If bronchiectasis is present, then resection of the affected segment(s) or lobe(s) should be performed.^{3,4}

The case we present corresponded to a type I congenital T/BEF in Braimbridge and Keith's classification.⁵ It consists of a tract connecting the esophagus and a lobar or segmental bronchus, associated with a congenital esophageal diverticulum. Our patient also had unilateral renal agenesis. We could not identify any precipitating event. Notably, the patient had undergone an esophagogastroscope 2 years before that did not disclose esophageal disease.

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Video-assisted resection of bilateral intralobar pulmonary sequestrations

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Bronchopulmonary sequestrations (BPSs) are characterized by anomalous vascular and tracheobronchial connections and account for a small proportion of pulmonary malformations. Bilateral BPSs are extremely rare, and the traditional surgical approach has been sequential resections with staged thoracotomies. We report a single-stage, sequential, video-assisted thoracoscopic surgery (VATS) approach to bilateral intralobar pulmonary sequestrations (ILSs).

Clinical Summary

An otherwise healthy 7-year-old girl was referred with low-grade fevers, productive cough, and a right lower lobe infiltrate on standard chest radiographs. She was given 2 courses of antibiotics, with only partial resolution of her right lung infiltrate. Subsequent high-resolution computed tomographic imaging demonstrated bilateral lower lobe BPS (Figure 1, A). Arterial supply to both sides was from the abdominal aorta (Figure 1, B).

In the operating room, after achievement of general anesthesia with a single-lumen endotracheal tube and bronchial blocker, the patient was initially placed in the left lateral decubitus position. The lung was carefully inspected with a 5-mm, 30° thoracoscope through a 5-mm anterior ninth interspace thoracoport. A 5-cm access incision was made in the fifth interspace anteriorly, sparing the serratus anterior muscle. Visualization was improved by placing a figure-of-eight stitch in the tendinous portion of the diaphragm and drawing it down through the same ninth interspace thoracoport. A consolidated intralobar sequestration was noted in the posterior and lateral basilar segments of the right lower lobe, with the aberrant arterial supply located in the inferior pulmonary ligament. The arterial supply was dissected and divided with a vascular TA-30 stapler. A sequestrectomy was performed with multiple applications of an Endo GIA 3.5-mm linear stapler (Figure 2).

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