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 esophagogastrosomy 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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Figure 1. A, Coronal computed tomographic image demonstrating bilateral bronchopulmonary sequestrations. B, Coronal computed tomographic image, with *white arrow* identifying left arterial feeding vessel and *black arrow* identifying right arterial feeding vessel, both originating from the abdominal aorta.

A 20F chest tube was placed, the lung was inflated under direct vision, and the incisions were closed in layers. The patient was then turned to the right lateral decubitus position, access to the left pleural space was obtained through a 5-mm ninth interspace thoracoport, and a 5-cm access incision was made in the fifth interspace anteriorly. The lung was inspected, and a similar intralobar sequestration was observed in the posterior basilar segment of the left lower lobe. The aberrant arterial source in the inferior pulmonary ligament was isolated and divided, and sequestrectomy was performed in similar fashion. A single left chest tube was placed, and the patient was transferred to the pediatric intensive care unit. Both chest tubes were removed on the second postoperative day, and the patient was discharged to home on the fourth postoperative day. Final pathology showed bilateral intralobar sequestrations with severe inflammation.

Figure 2. Excised right intralobar bronchopulmonary sequestration.

Discussion

ILSs are characterized by nonfunctioning lung tissue that lacks normal communication with the tracheobronchial tree. The arterial supply to ILSs is most often from the abdominal aorta, and venous drainage is usually through normal routes to the left atrium. Although children with BPSs are at risk for associated abnormalities, they occur less with intralobar than with extralobar sequestrations.

To date, there have been 12 reported cases of bilateral BPS in the literature, with only 5 intralobar cases.¹ All operative cases were approached with staged sequential thoracotomies.¹ More recently, surgical resection of BPSs has been accomplished with a VATS approach.²⁻⁴ Lobectomy, sequestrectomy, and wedge resections have all been described, with excellent results and a small number of conversions to thoracotomy.⁵ Because disease in this patient was limited to the basilar segments of the lower lobes, it was possible to perform sequestrectomies and preserve lung parenchyma. This unique case of bilateral ILS demonstrates the feasibility of a single-setting, sequential VATS approach to minimize postoperative pain and limit multiple or prolonged hospitalizations.

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