Recurrent Bronchogenic Pseudocyst 24 Years After Incomplete Excision*

Report of a Case

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Bronchogenic cysts (BCs) are uncommon congenital anomalies. Due to the inherent complications, the mere presence of a BC should warrant surgical therapy. Partial excision of these structures leads to recurrence. Complete surgical excision using a thoracotomy or video-assisted thoracic surgery is the goal. We report a case of recurrent bronchogenic pseudocyst 24 years after initial excision. This case supports the argument for complete surgical excision of BCs at the time of diagnosis.

Key words: bronchogenic cyst; recurrent; surgery

Bronchogenic cysts (BCs) account for 10 to 15% of all mediastinal masses.1 These are uncommon congenital anomalies, which represent independent differentiation of a group of cells which separate from the developing lung bud.2,3 Management of these anomalies has ranged from expectant observation to surgical resection.2,4-6 Partial excision of a BC, since it was initially reported by Adams and Thornton in 1943, has been controversial.1,7,8 Recently, the increased popularity of minimally invasive video-assisted thoracic surgical techniques, however, has resulted in the resurgence of interest in “unroofing” and incomplete excision of BCs. We describe a patient with a bronchogenic pseudocyst recurring 24 years after partial surgical resection. This case illustrates the need for complete open surgical excision of BCs.

CASE REPORT

A 72-year-old man presented to Mayo Clinic Scottsdale with a chronic cough of 5 years in duration. The cough had become more prominent in the past 2 years. At times, the cough had been productive of 4 to 5 teaspoons of white-yellow sputum daily. These episodes had responded to short courses of antibiotic therapy. The patient had undergone partial excision of a benign BC in 1959. A chest x-ray film revealed a double density in the subcarinal region with slight widening of the carinal angle (Fig 1). CT of the chest showed a large subcarinal mass that extended inferiorly to occupy the space behind the left atrium (Fig 2). In light of the patient's history, the diagnosis of a recurrent bronchogenic pseudocyst was made. Pulmonary function testing revealed a reduction in the total lung capacity (4.68 L or 73% of predicted). Bronchoscopy showed splaying of the main carina and compression of the proximal left mainstem bronchus without obstruction. There was no evidence of communication of the cyst with the tracheobronchial tube.

Surgical excision was performed through a secondary postero-

FIGURE 1. Standard posteroanterior chest radiograph showing a large bronchogenic pseudocyst located in the subcarinal area. The arrows indicate the double density created behind the left atrium.

FIGURE 2. Computed tomographic scan shows the bronchogenic pseudocyst (arrow) that occupies the space behind the left atrium. The pseudocyst displaces the esophagus and the left mainstem bronchus to the left.

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lateral right thoracotomy incision. The mass could be palpated only after dissection of fibrous mediastinal adhesions. Since this represented a recurrent bronchogenic pseudocyst, extracapsular dissection was not possible. Due to the lack of a true capsule, it was reasoned that any attempts at enucleation of the pseudocyst would result in spillage of possibly infected cyst content into the mediastinum. Consequently, after careful identification of the anatomical landmarks, the pseudocyst was incised. About 40 mL of thick green mucus was aspirated from the pseudocyst cavity. This approach prevented any spillage. An approximately 2-cm square area of residual cyst wall was identified which was adherent to the posterior aspect of the left mainstem bronchus. This represented the active mucus-producing epithelium, which could have been left from the first procedure 24 years earlier. The remainder of the pseudocyst wall was made up of inflammatory tissue adherent to the mainstem carina superiorly, the left atrium anteriorly and inferiorly, and the esophagus posteriorly (Fig 3). The pseudocapsule, along with residual cyst wall, was removed without compromising any of the adjacent structures. The pleural space was drained for 48 h. The patient’s recovery from the thoracic surgical procedure was short, and his hospitalization was prolonged only due to complications related to a radial artery monitoring line. Follow-up 2 months later revealed complete resolution of his pulmonary symptoms.

**DISCUSSION**

Bronchogenic cysts are closed sacs, which result from the development of a group of cells that separate from the lung bud. When the separation occurs during early gestation, the cysts are located along the tracheobronchial tree. If separation occurs later in the course of development, the cyst is more peripheral and is located within the lung parenchyma. Although both groups of BCs may be associated with a patent tracheobronchial communication, this is seen more frequently in the peripherally located group. The BCs are spherical structures that are lined by ciliated columnar epithelium and may have bronchial mucous glands, smooth muscle, or hyaline cartilage within their walls. Unless infected, these cysts usually contain thick mucus.

Although most BCs are located along the tracheobronchial tree, they have been reported in the neck, within the pericardium, in the prestenal space, and below the diaphragm. In 1948, Mater classified the tracheobronchial location of BCs as subcarinal, right and left peritracheal, right and left hilar, and paraesophageal types. The BCs are most frequently seen in the subcarinal region.

Until recently, most patients with BCs were thought to be asymptomatic; however, in a recent review of 86 patients from four institutions over a 20-year period by St. Georges and colleagues, 72.1% of patients were symptomatic. This finding has been supported by other investigators. The most common symptoms associated with BCs have been substernal pain, dysphagia, dyspnea, cough, and hemoptysis in a decreasing order of frequency.

A number of complications have been reported with BCs. Infection is the most common, with other reported complications including recurrent respiratory obstruction, fistulae to various structures in the chest, hemorrhage into the cyst cavity, superior vena caval syndrome, arhythmias, obstruction of the pulmonary artery, cyst rupture, cyst recurrence following incomplete excision, and malignant transformation. Adenocarcinomas and sarcomas arising from the cyst wall have been reported. A standard chest radiograph may result in suspicion of a BC; however, CT scans can clearly show the cyst and its relationship to other structures in the mediastinum. Although magnetic resonance imaging may confirm the presence of a BC, it has not been found to provide any more information than CT scans. In the presence of bronchial obstruction, bronchoscopy can confirm the compressional nature of the cyst.

The indication for surgical intervention has been merely the presence of the BC. This largely has been the result of the finding that most BCs become symptomatic or eventually are associated with complications. In the presence of complications, or in the setting of a recurrent BC, morbidity and mortality of surgical procedures have been significantly higher. The patients with recurrence usually have undergone an incomplete resection of the cyst. The retention of any actively secreting cyst wall has resulted in the formation of a pseudocyst. Most frequently, this has been observed in the subcarinal location.

Percutaneous or transtracheal needle aspiration has been advocated as an alternative to excision. These techniques have proven to be unsatisfactory with cyst recurrence in virtually all patients. Mediastinoscopy has been used for piecemeal removal of accessible cysts. Obviously, not all cysts are accessible by mediastinoscopy, and any remaining cyst wall will result in cyst recurrence. Disastrous complications have been observed with the use of this technique in the setting of an infected cyst. Video-assisted thoracic surgery has revolutionized the treatment of many pulmonary and mediastinal anomalies. Perhaps the most significant
benefits of this minimally invasive thoracic surgical procedure have been increased patient comfort and decreased postoperative morbidity. Video-assisted thoracic surgery has been advocated as a technique for unroofing and partial excision of BCs. Unfortunately, video-assisted thoracic surgery suffers from the shortcomings outlined for techniques using the mediastinoscope. Furthermore, video-assisted thoracic surgery is not applicable to all locations where BCs are found and may be associated with increased cost because of the longer operative times.

The patient reported herein exhibited many of the salient features associated with BCs. Twenty-four years after incomplete excision, he presented with a pseudocyst resulting from retention of a portion of the original cyst wall. At this time the patient was elderly, had greater risk factors for surgical intervention, and had experienced many episodes of pulmonary infection which had responded to antibiotic therapy. A recurrent pseudocyst should be entered and drained without spillage of cyst contents into the mediastinum. This maneuver allows for a controlled excision of the pseudocyst. A true capsule, as is seen in a primary bronchogenic pseudocyst, is not present because of the pathophysiology of pseudocyst formation. Attempts at “shelling out” of the pseudocyst will result in injury to the structures that form the wall of this structure. Sharp dissection of the pseudocapsule away from the bronchus, esophagus, and the posterior wall of the left atrium as well as complete excision of any active residual cyst wall are paramount to a successful outcome. The greater technical difficulties and inevitable formation of a pseudocyst after partial excision present a convincing argument for complete open resection of a BC.

Barring any medical contraindications, all patients with BCs should undergo surgical excision. This approach should be taken even in asymptomatic patients due to the risk of complications. In asymptomatic patients without other medical risk factors, open surgical excision of BCs can be accomplished with virtually no morbidity or mortality. Significantly higher complication rates are seen in the presence of complicated cysts or recurrence. Complete surgical excision is the goal. This can be performed through a right or left thoracotomy or video-assisted thoracic surgery depending on cyst location. Following complete excision, any defects in the tracheobronchial tree should be closed primarily and covered with a pedicle of pleura, pericardial fat, or intercostal muscle. Intrapulmonary BCs should be excised as a “wedge,” sparing most of the pulmonary parenchyma.

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