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Case Reports

Intralobar Pulmonary Sequestration Supplied by Multiple Anomalous Arteries: Report of a Case

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Abstract Pulmonary sequestration is abnormal pulmonary tissue that has separated from the normal pulmonary parenchyma, is not connected to the tracheobronchial tree, and is supplied by a systemic artery. We describe herein a case of intralobar pulmonary sequestration found in a 66-year-old man who was admitted to our hospital with hemoptysis, coughing, and fever. Angiography showed that the branches of the 11th left intercostal artery and a bronchial artery had formed a hypervascular area in the lower part of the left lung. Bronchial artery embolization and subsequent embolization of the left 11th intercostal artery were performed in an attempt to control the recurrent hemoptysis. These treatments were unsuccessful, and he was transferred to our department of surgery after coughing up about 400 ml of fresh blood. A left lower lobectomy was performed. The resected lung contained a large feeding artery, some acute and partly organizing inflammatory lesions within collapsed lung parenchyma, and massive intra-alveolar hemorrhage in the peripheral area. The patient had an uneventful recovery and was discharged 22 days after his operation.

Key words Pulmonary sequestration · Surgical resection · Embolization · Anomalous systemic artery

Introduction

Pulmonary sequestration is an uncommon disease characterized by nonfunctioning abnormal pulmonary parenchyma that has no connection with the tracheobronchial airway and receives its blood supply from a systemic artery.

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Transarterial embolization has been the standard management for hemoptysis[1] caused by hemorrhage from a bronchial artery,[2] a bronchial artery of anomalous origin,[3] or a nonbronchial systemic artery without sequestration.[4,5] On the other hand, intralobar sequestration is usually treated surgically as early as possible.[6] However, the indications for transarterial embolization in patients with massive hemoptysis from intralobar sequestration[7] have not yet been properly defined.

Oxman[8] reported the case of a patient with severe hemoptysis as well as bleeding into the pleural space, the esophagus, and the sequestrated lung. This report describes an unusual presentation of pulmonary intralobar sequestration that received its blood supply from the branches of the 11th left intercostal artery and the left bronchial artery. The limited indications of transarterial embolization for intralobar sequestration are discussed following this case report.

Case Report

A 66-year-old man was admitted to our hospital with recurrent hemoptysis. On physical examination, diminished breath sounds were audible over the base of the right lung. The results of routine serum biochemical investigations were normal, and the tumor markers carcinoembryonic antigen (CYFRA 21, and carbohydrate antigen 19-9) were all negative. Sputum and blood cultures were also negative. A chest computed tomography scan revealed a mass lesion with a low-density area in the left lower lobe consistent with pulmonary sequestration (Fig. 1).

Bronchoscopy did not show any active bleeding site or blood clots in the bronchial tree. However, angiography revealed that the branches of the 11th left intercostal artery (Fig. 2) and the left bronchial artery (Fig. 3) had formed a hypervascular area in the lower part of the left lung. Pulmonary arterial angiography revealed

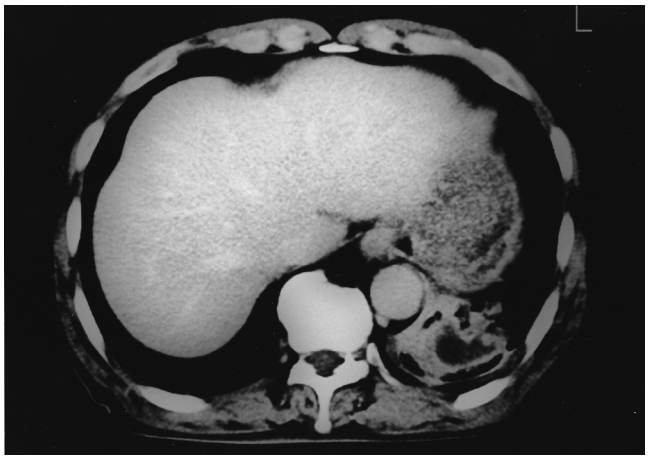

Fig. 1. Chest computed tomography (CT) scan demonstrated a mass lesion with a low-density area in the lower lobe of the lung

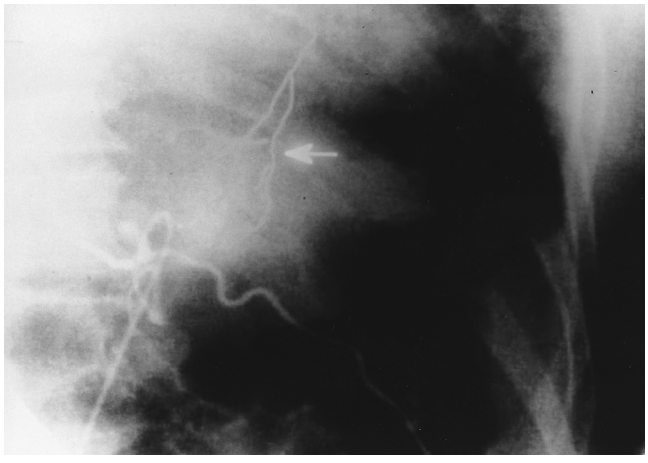

Fig. 2. Angiography showed that the branch of the left 11th intercostal artery (*arrow*) extended into the lower part of the left lung

that the lesion was a disconnected bronchopulmonary mass.

The patient initially underwent bronchial artery embolization (BAE) and subsequent transarterial embolization of the left 11th intercostal artery to control recurrent hemoptysis; however, 6 days later he coughed up about 400 ml of bright red blood and his condition became precarious. After his hemodynamic parameters had been stabilized, he was transferred to our department for surgical intervention.

At thoracotomy, the pleural space was found to be completely obliterated by dense adhesions, particularly in its caudal area. Intrapleural as well as extrapleural dissection was carried out and the whole left lung was freed. A left lower lobectomy was performed. Multiple cut sections of the resected lobe showed collapse of the

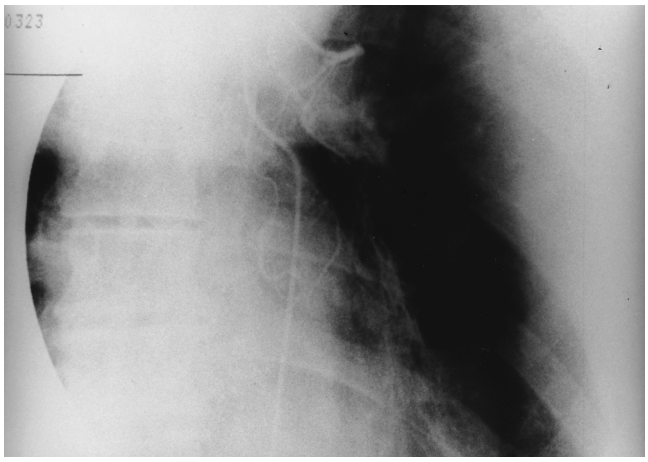

Fig. 3. Angiography revealed that the bronchial artery formed a hypervascular area in the lower part of the lung

parenchyma and peripheral massive intra-alveolar hemorrhage (Fig. 4, top). A large feeding artery considered to be the 11th intercostal artery was also noted. Microscopically, a feeding elastic artery with intimal fibrosis was seen to arise from the extrapleural space, penetrate the visceral pleura, and distribute into the lung parenchyma (Fig. 4, bottom). An area of secondary bronchiectasis accompanied by abscess-like acute inflammatory infiltrate with bacterial colonies was surrounded by organizing and fibrosing tissues. Culture from the abscess-like lesions was negative for mycobacteria.

The patient had an uneventful recovery and was discharged on the 22nd postoperative day.

Discussion

The term "sequestration" was first introduced by Pryce in 1946 to describe a disconnected bronchopulmonary mass with anomalous arterial supply.[9] Many variants of classic sequestration have been investigated by researchers since Pryce's original description. Pryce et al.[10] classified the extent of blood supply by defining the aberrant systemic artery in intralobar sequestration: as an abnormal artery without sequestration (type 1), an abnormal artery supplying the sequestered as well as the adjacent normal lung (type 2), and an abnormal artery supplying only the sequestered lung (type 3). The sequestration in our patient was categorized as type 3 according to Pryce's classification.

In general, the clinical manifestation of intralobar sequestration is a chronic cough, sputum, and recurrent attacks of pneumonia, usually caused by pyogenic infections. Having reviewed 540 published cases, Savic et al.[6] reported that 16% of sequestered lungs had multiple

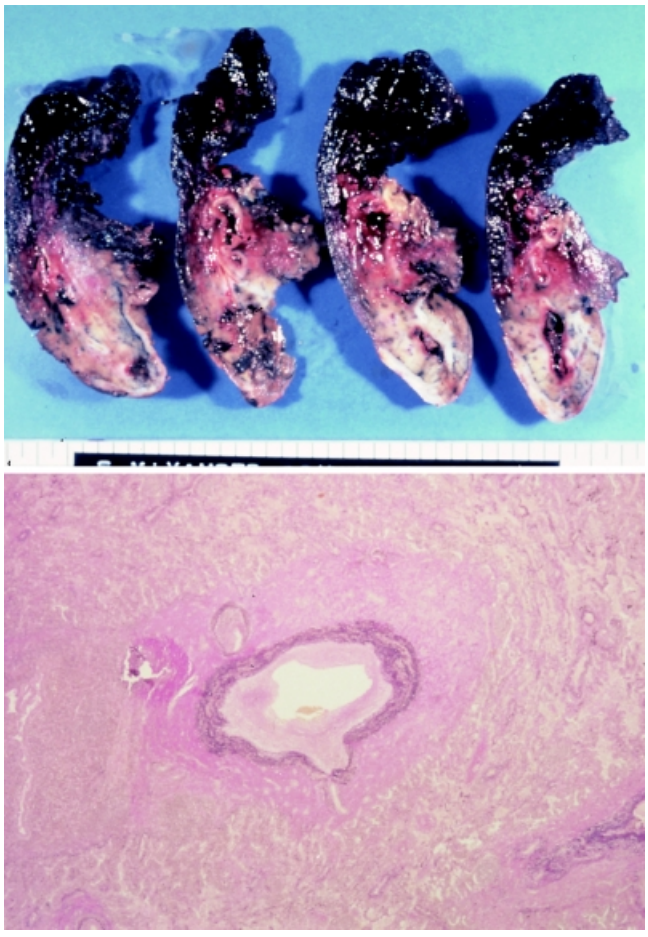

Fig. 4. Top Multiple cut sections of the resected lobe showed collapsed parenchyma and massive hemorrhagic lesions in the periphery. **Bottom** Microscopic examination revealed intimal fibrosis of an aberrant large elastic artery (Elastica van Gieson stain, ×25)

anomalous arteries. The sequestered lung in our patient was supplied by branches of the 11th left intercostal artery and the left bronchial artery. BAE is currently the standard treatment for massive or recurrent hemoptysis. Our review of the literature revealed many articles demonstrating the effectiveness of BAE in the control of hemoptysis.[11] The important role of the thoracic collateral circulation and intercostal arteries in the source of bleeding of intrathoracic lesions[12] was also reported. Hayakawa et al.[13] reported a high rate of hemoptysis control and a high cumulative survival rate in patients with hemoptysis treated by embolization of the feeding artery. However, failed BAE has been reported in patients with aspergillosis, lung abscess, and pulmonary sequestration during long-term follow-up.[13] Nevertheless, BAE remains the treatment of choice for life-threatening massive or recurrent hemoptysis in patients with pulmonary sequestration. BAE failed to control the hemoptysis in our patient due to incomplete embolization of the intercostal artery, and surgery was ultimately required. The resected lung revealed massive intra-alveolar hemorrhage with hemosiderin-laden macrophage as the cause of recurrent hemoptysis.

On one hand, the visceral pleural bronchial artery that develops secondarily, accompanied by bronchial artery or lung abscess, is a muscular systemic artery. On the other hand, the large elastic artery that is distributed with no relation to the pulmonary bronchial tree, usually recognized in pulmonary sequestration, was identified in our case. Thus, the histological diagnosis was made.[14,15] In patients with pulmonary sequestration supplied by multiple anomalous arteries, if BAE fails to stop bleeding from only one of the supply arteries, then there is strong possibility of recurrent hemoptysis, which may prove fatal. Accordingly, we consider that a surgical procedure in terms of lobectomy may well be the treatment of choice except when BAE is successfully performed for all the supply arteries. Moreover, segmentectomy or ligation of an anomalous artery is not enough, even if the patient is asymptomatic. Thus, it is important to delineate the abnormal lung parenchyma and identify any aberrant vessels before surgical resection is carried out.

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