

Pulmonary Sequestration Associated with Asymptomatic Aspergillosis

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Intralobar pulmonary sequestration associated with asymptomatic aspergillosis is a rare case. We describe the case of a 65-year-old woman with intrapulmonary sequestration, anomalous systemic arterial supply to the left lower lobe and aspergillosis who underwent left lower lobectomy and ligation of an anomalous artery by Video-Assisted Thoracoscopic surgery (VATS). Pathological examination showed the parenchymal distortion and chronic inflammation. Aspergillus were found in the cyst. VATS lobectomy for intralobar pulmonary sequestration is a safe and valid procedure. (Ann Thorac Cardiovasc Surg 2005; 11: 41–3)

Key words: pulmonary sequestration, aspergillosis, thoracoscopic

Introduction

Pulmonary sequestration involves an abnormal pulmonary tissue separated from the normal pulmonary parenchyma, not connected to the tracheobronchial tree and supplied by a systemic artery. Pulmonary sequestration itself is a rare disease. In addition, only a few studies have described pulmonary sequestration associated with mycosis, including aspergillosis, since the first report by Pryce in 1946.¹⁾ We report a case of an unusual presentation of pulmonary intralobar sequestration associated with asymptomatic aspergillosis.

Case Report

A 65-year-old woman with severe respiratory obstruction was admitted to our hospital with a massive hemoptysis. She was suspected to have a pulmonary sequestra-

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tion and was recommended for evaluation 13 years ago, but she refused at that time. On admission, the chest X-ray, showed a 5 cm solitary round shadow in the left lower lobe. A computed X-ray tomography (CT) of the chest (Fig. 1A) and aortography (Fig. 1B) demonstrated an abnormal artery arising from the thoracic aorta to supply the basal region of the left lower lobe.

These findings confirmed a diagnosis of pulmonary sequestration with anomalous systemic arterial supply to the left lower lobe and Video-Assisted Thoracoscopic surgery (VATS) lobectomy and ligation of anomalous artery were scheduled. A double-lumen tube was used to enable selective contralateral lung ventilation. Two 15 mm-incisions were made in the seventh intercostal space. A 50 mm-minithoracotomy was made in the sixth intercostal space on an anterior axillary line as an access port. The visceral pleura of the lower lobe of the left lung was covered with spider telangiectasia. An aberrant artery, which was 10 mm in diameter at its origin, was found originating from just above the diaphragm and divided following double ligation without any trouble, thereafter, left lower lobectomy was performed. The left pulmonary artery appeared to supply the upper lobe and segment 6 of the left lung. The patient had an uneventful postoperative course and was discharged on postoperative day 5. She was doing well without any symptoms 18 months after surgery.

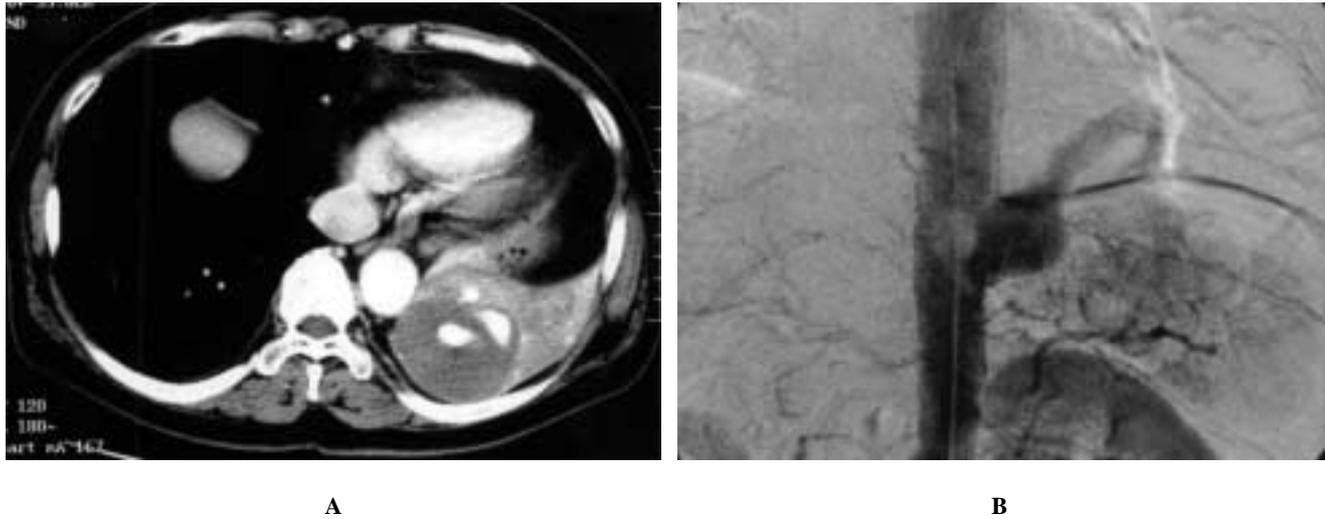


Fig. 1.

A: Chest computed tomography showed a huge aberrant artery with thrombus and sequestered segment of the left lower lobe of the lung.

B: Angiography showed an aberrant artery arising from the thoracic artery.

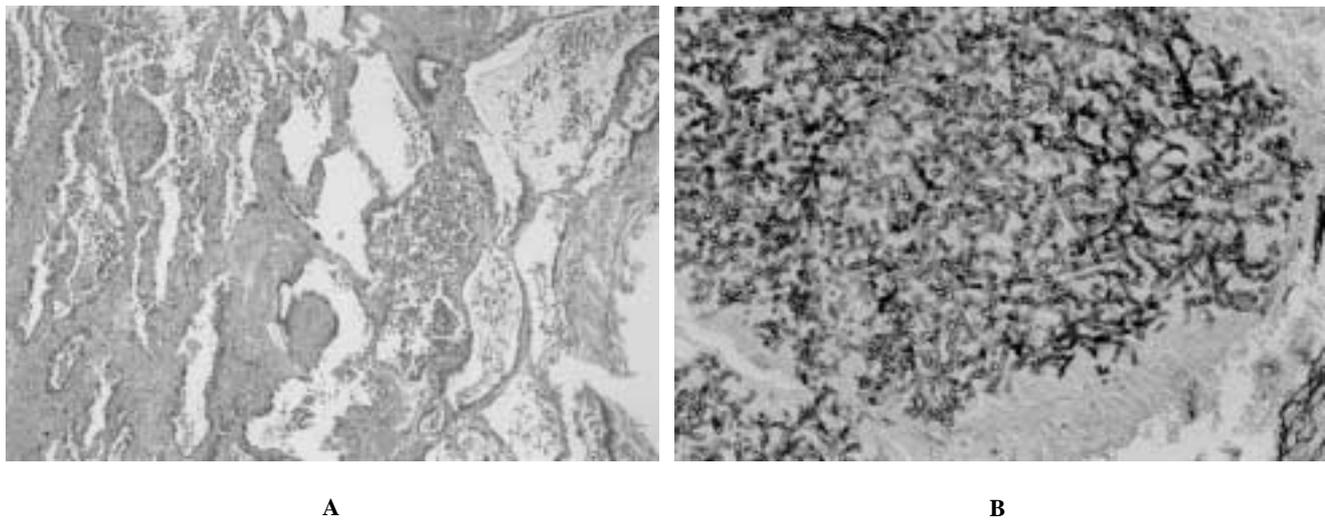


Fig. 2.

A: Microscopic findings showed a distorted pulmonary parenchyma with extensive replacement by dense fibrosis and chronic inflammation. Bronchioles filled with eosinophilic materials with numerous neutrophils (H & E, $\times 40$).

B: Hyphae of the aspergillus were observed in the bronchiole (Grocott's stain, $\times 200$).

The sequestered lung tissue shared common visceral pleura with the rest of the normal lung parenchyma. Pathological examination showed bronchioles filled with eosinophilic materials, and a distorted pulmonary parenchyma with extensive replacement by dense fibrosis and chronic inflammation. Several dilated bronchioles lined by cuboidal-to columnar epithelium, were filled with mucous, numerous neutrophils (Fig. 2A) and abundant aspergillus hyphae (Fig. 2B).

Discussion

In general, the clinical manifestation of intralobar sequestration is a chronic cough, sputum and recurrent attacks of pneumonia. Our case suffered from none of them except an occasional hemoptysis. However, a massive hemoptysis and severe respiratory obstruction caused by thrombus occurred suddenly. The cause of bleeding was unclear because there were no communication between

the sequestered lung and the normal bronchus by pathological examination. The rupture of abnormal vessels in the bronchioles or alveolar level was suspected.

Pryce et al.²⁾ classified the pulmonary sequestration into three types according to the extent of blood supply from an aberrant systemic artery into intralobar sequestration. Our case, in which an abnormal artery supplied only the sequestered lung, was type III of Pryce's classification.

There have been only nine reports on pulmonary sequestration associated with aspergillosis.^{3,4)} In 6 of nine reports, patients were symptomatic and the others were asymptomatic.⁴⁻⁶⁾ Although it is difficult to make the mechanisms of asymptomatic infection clear, we speculate that one of the mechanisms is disappearance of communication between the normal lung and sequestered lung following development of the communication in some way and colonization of trans-tracheal inhaled aspergillus.

The definitive treatment of pulmonary sequestration is surgical resection. The treatment for an asymptomatic pulmonary sequestration still remains controversial. However, in our case, aspergillosis was concealed and life-threatening hemoptysis developed. So we strongly recommend surgical resection of the sequestered lung. The use of VATS to perform therapeutic procedure of pulmonary sequestration has been reported by Wan et al.⁷⁾ We also have experienced surgery by VATS not only by standard lobectomy but also applied as alternative surgery.⁸⁾ We used a 5 cm-minithoracotomy to visualize the operative field directly. This method enabled us to ligate and divide an aberrant artery easily and safely. We also applied double ligation to the aberrant thoracic aortic branch. This procedure is more cost effective than the use of an endoscopic stapler-cutter and enables us to divide an ab-

errant artery at the level of the origin and to prevent aneurysmal change of an arterial stump. We conclude that VATS lobectomy for pulmonary sequestration is a safe and valid procedure.

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