

Surgical Treatment of Infected Intralobar Pulmonary Sequestration: A Collective Review of Patients Older than 50 Years Reported in the Literature

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We report on the rare and surgical treatment of a senile patient of infected intralobar pulmonary sequestration. A 56-year-old male who had complained of headache, vomiting, cough, sputum production, and high fever was admitted to our hospital. Chest computed tomography (CT) showed an infected intralobar pulmonary sequestration as an 8×6 cm cystic mass with multiple air-fluid cavities in the left lower basal segment and severe pneumonia in the left upper and lower lobes around the mass. A 3-D CT showed an aberrant artery entering the consolidation from the descending aorta. A standard lower lobectomy was performed with a ligation of the aberrant artery with a diameter of 1 cm supplying the posterior segment of the left lower lobe. A histological examination of the lung revealed acute and chronic broncho-bronchiolitis with cystic dilatation consistent with intralobar pulmonary sequestration. We discuss the characters of senile patients compared with juvenile patients, with reference to a collective review of patients older than 50 reported in the literature. (Ann Thorac Cardiovasc Surg 2007; 13: 331–334)

Key words: intralobar pulmonary sequestration, aberrant artery, lower lobectomy

Introduction

Intralobar pulmonary sequestration is a rare congenital malformation disease, and patients often present with recurrent bronchitis, pneumonia, or hemoptysis. We report a case of left lower lobectomy in a 56-year-old male. Intralobar sequestration is typically diagnosed in juvenile patients. Because surgical treatment for intralobar pulmonary sequestration in patients older than 50 is very rare, we discuss it with reference to a collective review of such patients reported in the literature.

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Case Report

A 56-year-old male who had complained of headache, vomiting, cough, sputum production, and high fever was admitted to our hospital. He already had a previous history of pulmonary sequestration as a result of being diagnosed at another hospital 4 years previously, and he often presented with recurrent pneumonia. Chest computed tomography (CT) showed an infected intralobar pulmonary sequestration as an 8×6 cm cystic mass with multiple air-fluid cavities in the left lower basal segment and severe pneumonia in the left upper and lower lobes around the mass (Fig. 1). A 3-D CT showed an aberrant artery entering the consolidation from the descending aorta (Fig. 2). Laboratory findings showed a white blood cell count of 21,200/mm³ and C-reactive protein (CRP) of 27.3 mg/dl. Serum carcinoembryonic antigen (CEA) was 1.0 ng/dl, and carbohydrate antigen 19-9 (CA19-9) was 9 units/ml. We diagnosed the case as intralobar pulmonary sequestration of the left lower lobe with infection involving an aberrant artery arising from the descending aorta just above the diaphragm. The operation was performed

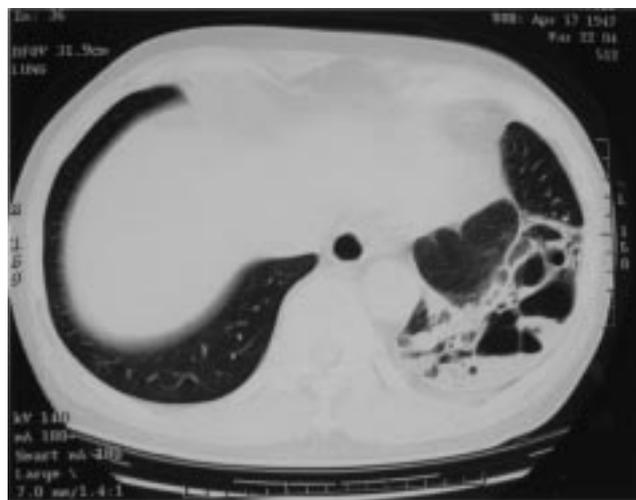


Fig. 1. Computed tomography shows an infected intralobar pulmonary sequestration as an 8×6 cm cystic mass with multiple air-fluid cavities in the left lower basal segment.

under general anesthesia with a double-lumen endotracheal tube. We could not perform a lobectomy under video-assisted thoracoscopic surgery (VATS), so we performed a standard lower lobectomy with ligation of the aberrant artery with a diameter of 1 cm supplying the posterior segment of the left lower lobe, because there was severe intrathoracic all-around adhesion between the mass and the chest wall. The resected specimen showed consolidated parenchyma containing a few cystic cavities (Fig. 3), and a histological examination of the lung revealed acute and chronic broncho-bronchiolitis with cystic dilatation consistent with intralobar pulmonary sequestration. The patient was later discharged in good health on the 20th postoperative day.

Discussion

A pulmonary sequestration is a rare malformation in which pulmonary tissue is formed with no connection to the normal bronchial system or the aberrant arteries from the systemic supply, either within the lung (intralobar) or as a separate adjunct (extralobar). Pryce et al.¹⁾ classified intralobar sequestration by the extent of blood supply from an aberrant systemic artery in 1947. There are three types: an aberrant artery without sequestration (type 1), one supplying the sequestration as well as the adjacent normal lung (type 2), and one supplying only the sequestration (type 3). Because an aberrant artery supplied only the sequestered lung in our case, it was type 3 of Pryce's clas-

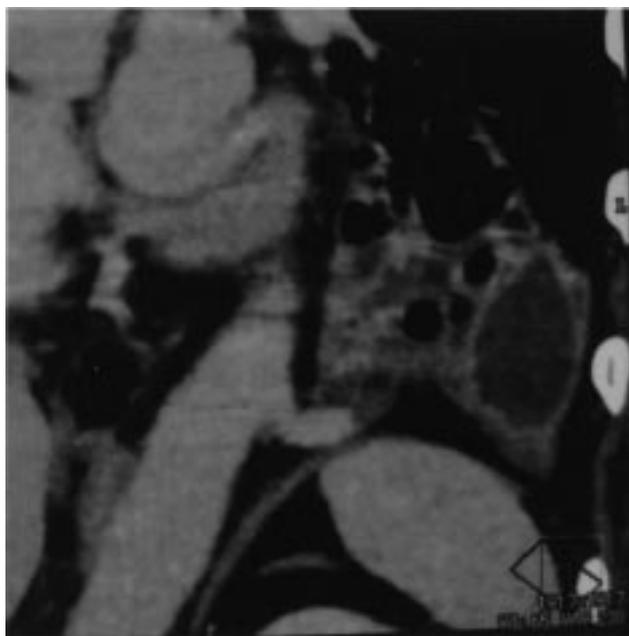


Fig. 2. A 3-D computed tomography shows the aberrant artery entering the consolidation from the descending aorta.

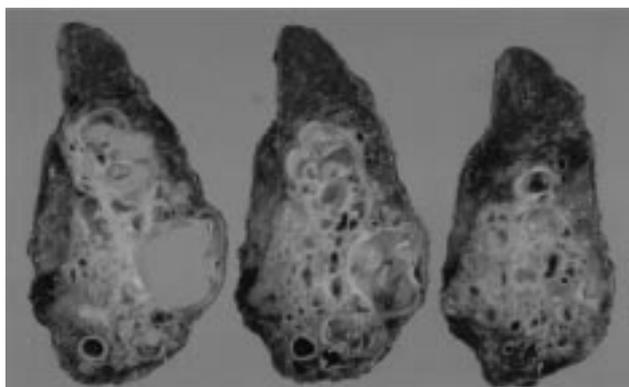


Fig. 3. A resected specimen showing a consolidated parenchyma containing a few cystic cavities, revealing acute and chronic broncho-bronchiolitis with cystic dilatation consistent with intralobar pulmonary sequestration.

sification.

Intralobar sequestration is diagnosed during the second decade of life or younger in approximately 50–60% of cases, and it is rarely found in patients older than 50.²⁾ In reviewing the literature for surgical cases of pulmonary intralobar sequestration in patients older than 50, we found only 10 since 1998, including our case (Table 1).^{2–10)} As shown in Table 1, patient ages ranged from 53 to 79; 5 cases were male and 5 female. Among senile patients it

Table 1. A review of the literature concerning the surgical case reports of pulmonary intralobar sequestration in patients older than 50 since 1998

Author (year)	Age/Sex	Symptoms	Location	Origin of aberrant arteries (number/maximum diameter)	Operation
1) Tsunozuka and Sato (1998) ³⁾	75/F	Asymptomatic (abnormal shadow)	Left posterior basal segment	Descending thoracic aorta (3/2 mm, 2 mm, 2 mm)	Lower lobectomy
2) Yamanaka et al. (1999) ⁴⁾	68/F	Dyspnea, hemoptysis	Left posterior basal segment	Descending thoracic aorta (1/20 mm)	Basalectomy
3) Sakiyama et al. (2000) ⁵⁾	60/F	Lumbago segment	Left posterior basal segment	Descending thoracic aorta (2/7 mm, 3 mm)	Basalectomy
4) Lewis and Tsour (2000) ⁶⁾	66/M	Dyspnea	Left lower lobe	Left circumflex artery (1/unknown)	None
5) Singh and Nath (2001) ⁷⁾	53/M	Hemoptysis	Left posterior basal segment	Descending thoracic artery (1/30 mm)	Lower lobectomy
6) Petersen et al. (2003) ²⁾	79/F	Hemoptysis	Left lower lobe	Descending thoracic artery (1/1 mm)	Lower lobectomy & graft replacement of descending thoracic artery
7) Sakuma et al. (2004) ⁸⁾	60/M	Asymptomatic (abnormal shadow)	Right lower basal segment	Descending thoracic artery (1/unknown)	Video-assisted thoracoscopic wedge resection
8) Sato et al (2005) ⁹⁾	65/F	Hemoptysis	Left lower basal lesion	Descending thoracic artery (1/10 mm)	Video-assisted thoracoscopic lower lobectomy
9) Matsuoka and Nohara (2006) ¹⁰⁾	62/M	Asymptomatic (abnormal shadow)	Left lateral & dorsal basal segment	Descending thoracic artery (1/unknown)	Lower lobectomy
10) Hirai (2006)	56/M	Pneumonia	Left posterior basal segment	Descending thoracic artery (1/10 mm)	Lower lobectomy

has an equal gender distribution as well as among juvenile patients. In general, most senile patients have a chronic cough, sputum, and recurrent attacks of pneumonia similar to those in juvenile patients, as in our case.²⁾ Hemoptysis in association with infection is also a common presenting sign among senile patients, and 4 cases had symptoms of hemoptysis (40%). Savic et al.¹¹⁾ reported that more than 50% of patients become symptomatic after the second decade of life, and in 15% of patients anomaly caused no symptoms. Our reviews in patients older than 50 revealed that the number of symptom-free patients, 3 (30%), was higher than that reported by Savic et al. It thus appears that this reflects a higher likelihood for older adults to obtain chest radiographs for unrelated reasons. In juvenile patients, the arterial supply originates from the descending thoracic aorta predominantly, and it occurs in the left lung, most commonly in the basal segment of the lower lobe.⁴⁾ Also in senile patients, the aberrant artery was seen arising from the descending thoracic aorta in 9 patients (90%), and all lesions were in the left lower lobe (90%), most commonly

in the basal segment of the lower lobe, as in our case.

A diagnosis of sequestration requires a high index of suspicion. A chest radiograph may reveal a homogeneous lesion in the lung base in uncomplicated intralobar sequestration, although complicated sequestration may show as a cystic lesion with air-fluid levels. Intralobar sequestration diagnosed typically in juvenile patients is often misdiagnosed both because of a lack of recognition of chest radiographic findings and a low expectation of the disease in senile patients.

The main treatment for pulmonary sequestration should be surgical resection with a ligation for an aberrant artery because of recurrent pulmonary infection and the unfavorable cardiac influence caused by the existing aortopulmonary shunt, though limited success has been reported with the use of a simple ligation technique for the feeding artery. However, before surgical resection it is important to delineate the abnormal lung parenchyma and identify any aberrant vessels for the reduction of serious hemorrhage during operation. Traditionally, angiography is required for a definitive diagnosis, but we be-

lieve that recent advances in the diagnosis value of CT (helical CT, dynamic CT, and 3-D CT) may be sufficient to provide noninvasive alternative diagnostic methods for pulmonary sequestration. In particular, 3-D CT provides accurate information on the aberrant vascular supply and can replace angiography in the preoperative diagnosis, as in our case.

Also in senile patients, the definitive treatment of pulmonary sequestration is surgical resection. Our review found that 9 cases (90%) received surgical resection in patients older than 50, including asymptomatic patients. Six cases underwent a lower lobectomy, which involved the graft replacement of the descending thoracic aorta in 1 case and VATS in 1 case. Two cases underwent basalectomy, and 1 underwent wedge resection using VATS. The first use of VATS in the surgical treatment of pulmonary sequestration was reported by Wan et al.¹²⁾ in 2002. We also believe that VATS provides a more patient-friendly approach and minimal invasiveness for senile patients, though we were obliged to select a standard lower lobectomy because of severe intrathoracic all-around adhesion between the mass and the chest wall. Furthermore, we believe that in the treatment of severe infected intralobar pulmonary sequestration, such as in our case, segmentectomy or ligation of the aberrant artery is not sufficient, and that the most favorable treatment is lobectomy in order to avoid contamination.

Because a surgical treatment of intralobar pulmonary sequestration in patients older than 50 is very rare, we report and discuss the characters of senile patients compared with juvenile patients, with reference to a collective review of the literature.

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