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Case Report
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Intralobar Pulmonary Sequestration with Successful Preservation of the Lung Lobe after Intraoperative Marking: A Case Report

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Abstract

Background: Pulmonary Sequestration (PS) is a relatively rare lung deformity. Surgical resection is the best treatment for asymptomatic PS. Although local resection should be the first choice for intralobular PS, there are still no published reports on how to distinguish the sequestered lung tissue from normal lung tissue and furthermore, how to remove it during local resection.

Case Report: This paper describes a 46-year-old female patient without the typical symptoms of PS or a history of tuberculosis. Enhanced computed tomography showed sequestered lung tissue with two abnormal blood supply arteries. After undergoing preoperative preparation, the patient was placed in the right decubitus position, and under general anesthesia, Video-Assisted Thoracoscopic Surgery (VATS) was performed; the patient was initially considered to have Intralobar Sequestration (ILS), with two abnormal blood vessels entering the posterior basal ganglia from the thoracic aorta. The diameters of the basal segments were 8 mm and 5 mm. A few minutes after the abnormal blood supply artery was severed, we observed a significant change in the color of the sequestered lung tissue, which was consistent with that of a healthy lung. Then, we confirmed the hypothesis that the electric hook placed as a marker was at the boundary between the two lobes. The postoperative course was uneventful, the pathological findings showed signs of sequestration of the left lower lobe, and she was discharged on the 5th day after surgery. CT examination of the chest at 6 months after surgery showed that the lungs remodeled well without inflammation.

Conclusion: Typically, the surgical treatment of pulmonary ILS combined with healthy lung tissue in the lobe will cause irreparable tissue loss to the patient. We attempted to mark the lung during surgery to ensure that as much healthy lung tissue was preserved as possible after complete resection of the sequestered lung tissue to improve the patient's postoperative quality of life. The patient recovered well after surgery, confirming the effectiveness of our procedure; this method provides an alternative for patients with PS who can be treated with partial resection; however, a large number of cases are still required to further support these findings.

Keywords: Intralobar sequestration; Dual artery blood supply; Intraoperative marking; Retention of lung lobes; Local excision

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Abbreviations

PS: Pulmonary Sequestration; ILS: Intralobar Sequestration; ELS: Extralobar Sequestration; CT: Computed Tomography; VATS: Video-Assisted Thoracoscopic Surgery

Background

Pulmonary Sequestration (PS) is a relatively rare pulmonary malformation. In PS, the pulmonary tissue in the lesion area is not connected with the bronchus, and the blood supply often comes from the aorta or its branches; this condition accounts for 0.15% to 6.4% of all congenital lung deformities. PS can usually be divided into two types: Intralobar Sequestration (ILS): Sequestration of lung tissue without independent pleura, with normal lung tissue wrapped in the same visceral pleura, most commonly occurs in the posterior basal segment; and Extralobar Sequestration (ELS): sequestration of lung tissue with its own independent pleura, separated from normal lung tissue [1,2]. PS is often characterized by cough, purulent sputum, etc. Surgical resection is the best treatment for asymptomatic PS [3,4]. Although most scholars believe that local resection should be the first choice for surgically treating conditional ILS, there are still no specific reports on how

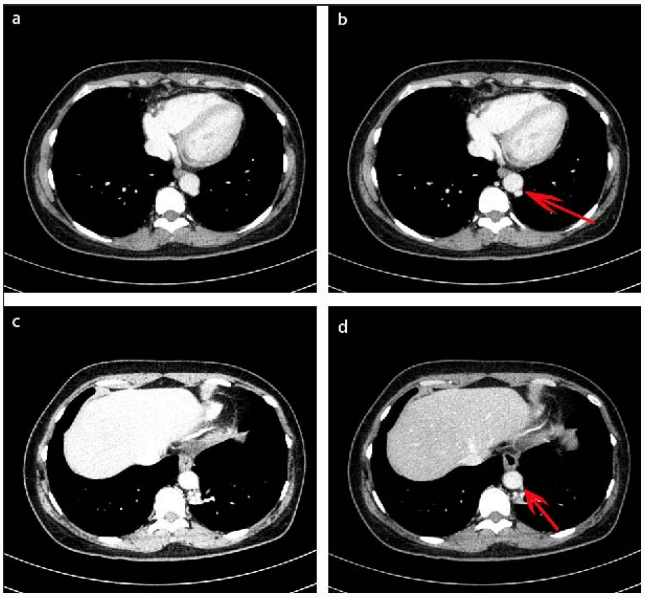

Figure 1: Preoperative chest CT of pulmonary sequestration showed two large vessels originating from the thoracic aorta with diameters of 8 mm and 5 mm, suggesting that the sequestered tissue that might be supplied by these two arteries.

to distinguish sequestered lung tissue from normal lung tissue and furthermore, how to further it during local resection [5-9].

Case Presentation

The case describes a 46-year-old female patient. She was admitted to our hospital because of a shadowy mass in the left lower lung found by physical examination performed in another hospital 3 months ago. The patient showed no typical symptoms of PS, such as cough, sputum or hemoptysis, and denied a history of tuberculosis. Enhanced thoracic Computed Tomography (CT) revealed an irregular high-density shadow in the posterior basal segment of the left lower lung, in which multiple thick vascular shadows were seen, and the trunk was from the descending aorta, suggesting PS of the left lower lobe (Figure 1). The preliminary diagnosis of PS was confirmed. After completing preoperative preparation, the patient was placed in the right decubitus position and underwent with Video-Assisted Thoracic Surgery (VATS) under general anesthesia. The local dense adhesions in the left thoracic cavity were gradually separated, and the adhesions were broken. Consolidation in the posterior basal segment of the left lower lung was observed, far from the hilum and confined, and the consolidated tissue and normal lung tissue shared the same visceral pleuron, which was preliminarily considered to be ILS (Figure 2a). The sequestered tissue was gradually resected from the pulmonary ligament, and 2 thick abnormal blood vessels arising from the thoracic aorta and entering the left lower lobectomy basal segment were visible, with diameters of 8 mm and 5 mm (Figures 2b-2f); a vascular malformation, marked hyperaemia in the basal segments of the left lung, and relatively obvious color differences with normal lung tissue (Figure 2a) were seen intraoperatively, resembling the characteristics of sequestered lung tissue. To maintain as much normal lung tissue as possible, we decided to use an electric hook to determine the boundary of the sequestered lung tissue before severing the abnormal blood supply (Figure 2b). The sequestered tissue was gradually freed to expose the blood vessels, and then the abnormal blood vessels were separated. A few minutes after the abnormal supply arteries were severed, we observed with the naked eye that the color of the sequestered lung tissue changed significantly to be consistent with that of healthy lung tissue (Figure 2g), thus confirming that the

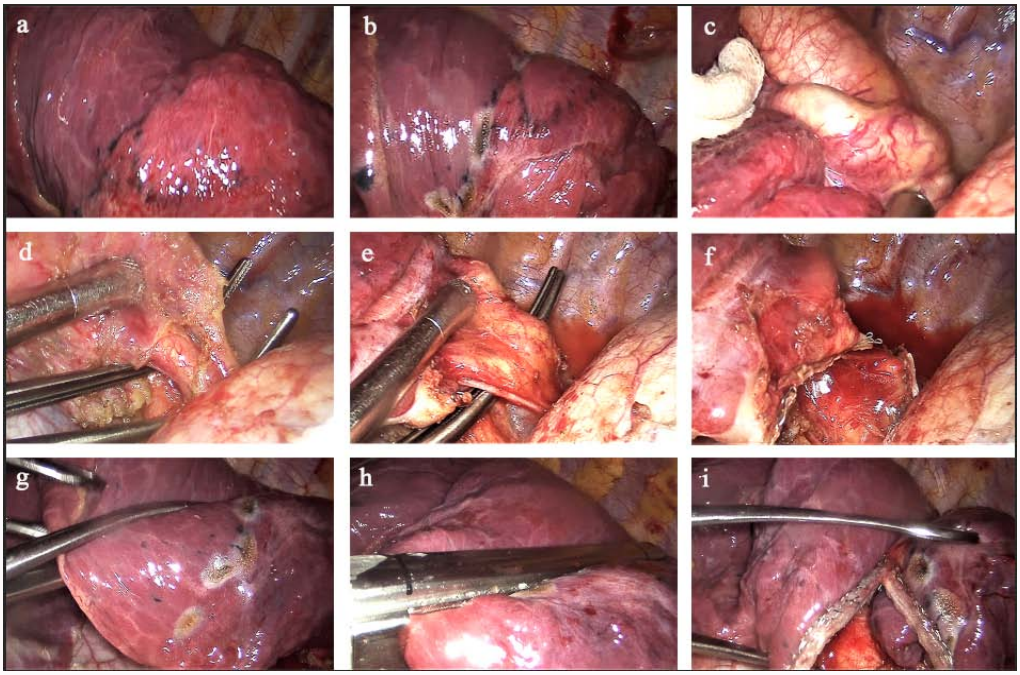

Figure 2: Exploration of the chest cavity. a. Surgical exploration showed that the consolidated tissue in the left lower lung shared the same visceral pleura with the normal lung tissue, and the initial consideration was ILS. A clear color boundary existed between the normal lung tissue and sequestered lung tissue. b. Electrocoagulation marks were made along the color boundary. c and d. The first abnormal blood supply artery. e. The second abnormal blood supply artery. f. The second supply artery after rupture. g. Several moments after severing the two blood supply arteries, the color boundary between the normal lung tissue and sequestered lung tissue disappeared, leaving only our pre-marked boundary. This indirectly confirmed our estimation of the color boundary, that is, the boundary between two different tissue types. h and i. The sequestered lung tissue was excised according to the present boundary before the artery was severed.

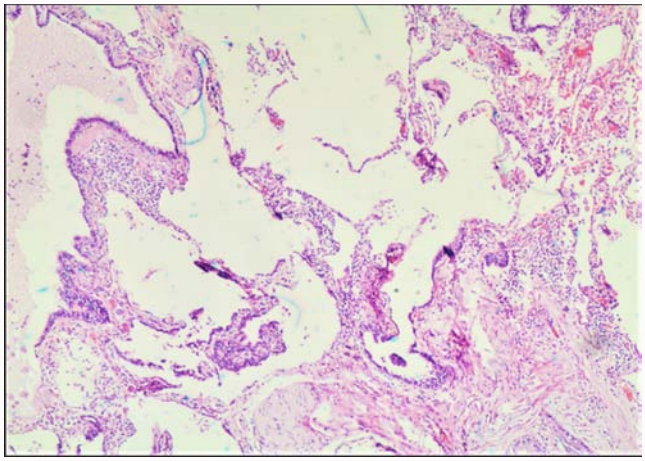

Figure 3: Pathological examination showed ILS in the left lower lobe.

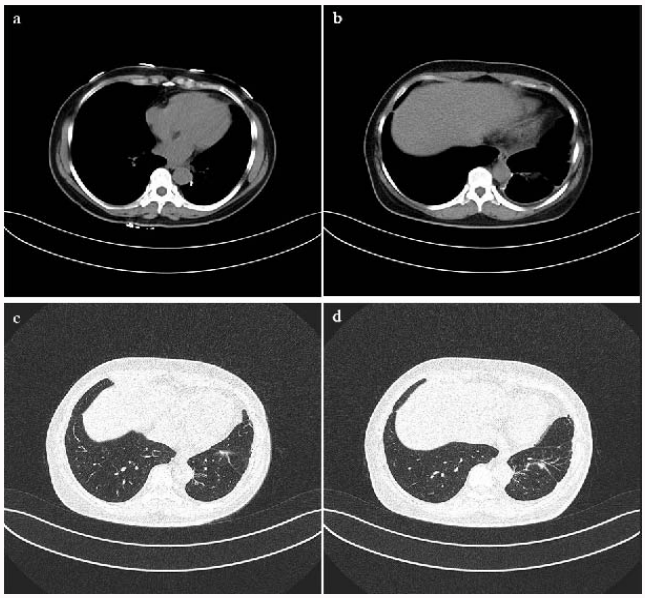

Figure 4: The chest CT scan showed good pulmonary reinflation, and there were no inflammatory changes 6 months after the operation.

electric hook marker was placed at the boundary between the two tissue types. The lower lung ligament was further dissected to the level of the lower pulmonary vein, and the basal segment of the lower lung was removed by wedge resection along the preset edge (Figure 2h and i). Water was introduced to test for a leakage in the lung. No air leakage was found at the cutting edge of the lung with the water column up to 40 cm. The patient returned to the ward after surgery, and the resected specimen was examined and sent for pathological examination. Pathological examination showed ILS in the left lower lobe (Figure 3). The postoperative course was uneventful, and the patient was discharged 5 days after the operation. The chest CT scan showed good pulmonary reinflation, and there were no inflammatory changes 6 months after the operation (Figure 4).

Discussion

Patients with ILS often present with cough, sputum, and even hemoptysis [10]. The patient in this case was confirmed to have ILS after surgery, but there was no typical clinical manifestation of PS before surgery. This suggests that the possibility of ILS cannot be ruled out solely on the basis of negative clinical manifestations. Enhanced thoracic CT can clearly display abnormal blood supply arteries to the sequestered lung tissue, so this method has become the first choice for the diagnosis of PS [11]. The abnormal blood supply arteries mainly arise from the thoracic aorta; in addition, they can include the abdominal aorta, intercostal artery, subclavian artery, or even coronary artery [12]. The abnormal arteries are usually hidden in the inferior pulmonary ligament, and there can be 1 to 3 abnormal arteries, or even more in some cases [3]. This report described a rare case of PS with a blood supply from both arteries of the descending aorta. Therefore, for patients with PS, we should repeatedly review the relevant imaging data before surgery to find as many of the supplying arteries as possible. Careful separation during surgery should be carried out for vessels that appear to be abnormal arteries so that dangerous situations caused by the omission of supplying arteries can be avoided during surgery. Thoracoscopic surgical resection (especially thoracoscopic anatomical lobectomy) is a good option for simple PS without pleural or hilar adhesions [13]. In this case, although there were local adhesions in the pleural cavity, the prognosis of the patient after thoracoscopic surgery was good; suggesting that patients with PS but no serious adhesions can undergo thoracoscopic surgery to achieve better treatment results. The procedure for ILS is usually a complete lobectomy of the sequestered lung and its associated lobes. In this case, however, the authors believed that the sequestered lung tissue of this patient was located in the posterior basal segment of the left lower lung far away from the hilum, there was with limited lesions, and there was a clear color boundary between the sequestered lung tissue and healthy lung tissue, making it easier to distinguish between the two tissues. Therefore, the surgical resection extent can be determined based on these characteristics, and the boundary could be marked by cauterization before the abnormal blood supply artery was severed to ensure complete resection of the sequestered lung tissue; moreover, local resection can be selected to retain healthier lung tissue. Compared with total lobectomy, this procedure improves the patients' postoperative quality of life. Although it has been reported [8,14] that wedge resection and segmental resection are feasible alternative to lobectomy in some cases of ILS, the specifics of how to perform each operation have not been further clarified, and the complexity of the surgery may lead to a prolonged operation time. The key to surgery is to distinguish between normal lung tissue and sequestered lung tissue. In ILS, approximately 95% of the pulmonary veins return to the pulmonary circulatory system. This produces a pathophysiological change called a left-to-left shunt, causing differences in the oxygen content of the surrounding normal lung tissue. Therefore, during the operation, there was a relatively obvious color boundary between the two tissues (the sequestered lung was pink, and there was no carbon dust on the surface). To confirm the color boundary, we first marked this boundary with an electric cautery knife, severed the blood supplying artery, and observed that the boundary gradually disappeared. This indicates our estimation of the color boundary between the two tissues was correct. Therefore, we removed the sequestered lung tissue along the boundary, leaving as much healthy lung tissue as possible. The patient was successfully discharged from the hospital after the operation, and no related complications occurred.

Conclusion

PS is divided into ILS and ELS. In the surgical treatment of ILS, the lobes belonging to the sequestered lung tissue are usually removed together, but because of the simultaneous removal of healthy lung

tissue within these lobes, the patients experience irreparable tissue loss. Therefore, we attempted to more carefully mark the lung during surgery to ensure that as much healthy lung tissue as possible is preserved after complete resection of the sequestered lung tissue, thereby improving the patient's quality of life after surgery. The patient recovered well after surgery, which confirmed the effectiveness of our procedure; this method provides an alternative for patients with PS who can undergo partial resection; however, a large number of cases is still required to further support these findings.

Declarations

Ethics approval and consent to participate: The participants provided informant consent and agreed to participate. Ethical approval was obtained from the Ethics Committee of the Affiliated Hospital of Zunyi Medical University. Written consent was obtained from the patient and his relatives for the publication of this article, the related medical history and the imaging data.

Consent for publication: Written consent was obtained from the patient and his family for the publication of this manuscript and the use of related images. All authors confirmed that written informed consent from the patient and his family members was obtained for the publication of this case report and any related images. The written consent may be reviewed by the editor-in-chief of this journal.

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